CASE REPORT

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Probable Nonconvulsive Status Epilepticus after Drainage of a Chronic Subdural Hematoma in a Patient with Moyamoya Disease

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A 52-year-old woman with hypertension and moyamoya disease presented with chronic subdural hematoma (CSDH). The presumed cause of bleeding was ascribed to administrated antiplatelet agents. She responded slowy and clumsily to verbal commands and had right arm weakness. After surgery, her clinical condition improved. But two days after surgery, her symptoms became aggravated and a convulsive seizure was noted within 24 hours. Brain magnetic resonance imaging showed no organic lesion except a small amount of residual CSDH. In addition, there was no laboratory evidence of metabolic brain disease. Moreover, after the administration of an antiepileptic drug (phenytoin), her manifestations disappeared. Therefore, the authors presume that her symptoms were resulted from nonconvulsive status epilepticus (NCSE), despite a lack of ictal period electroencephalographic findings. The authors were unable to find a single case report on postoperative NCSE in Korea. Therefore, the authors report this case of nonconvulsive status epilepticus after drainage of a CSDH in a patient with moyamoya disease.

KEY WORDS : Chronic subdural hematoma · Moyamoya disease · Nonconvulsive status epilepticus.

Introduction

Nonconvulsive status epilepticus (NCSE) can be defined as a condition of ongoing or intermittent clinical epileptic activity without convulsions for at least 30 minutes. Without electroencephalography (EEG) correlate, the diagnosis remains probable, whereas with EEG correlate it should be regarded as definite[6]. Some patients postoperatively show an altered mentality or behavioral changes. In such cases, the majority of physicians would recommend an imaging study to evaluate any intracranial lesion and a laboratory study to determine the presence of a metabolic brain disease. However, if no abnormality is found, it is not easy to manage these patients. Moreover, although epilepsy is commonly associated with such features, it is not easy to consider epilepsy in cases with no evidence of convulsion. In the literatures, NCSE is considered to constitute almost a quarter of status epilepticus cases, and the annual incidence of an episode of NCSE varies from 1.5 cases to 18.5 cases per 100,000 of the population[1,14,16]. According to this, the estimated number of domestic cases ranges from 720 to 8,880 per year. Moreover, a delay in diagnosis in postoperative patients presenting with NCSE maybe harmful. Nevertheless, we were unable to find a single case report on postoperative NCSE in Korea. Therefore, the authors report this case of nonconvulsive status epilepticus after drainage of a chronic subdural hematoma (CSDH) in a patient with moyamoya disease.

Case Report

A 52-year-old woman with hypertension and moyamoya disease presented with right arm weakness with an onset three days prior to visit. She responded slowly and clumsily to verbal commands and her right arm muscle power was grade IV. Three years previously, she had experienced a transient ischemic attack which presented with similar symptoms to this episode, and was diagnosed as having moyamoya disease after cerebral angiography. She was being prescribed clopidogrel (75 mg q.d.) and aspirin (100 mg q.d.). One year before this presentation, she had suffered a similar event which persisted

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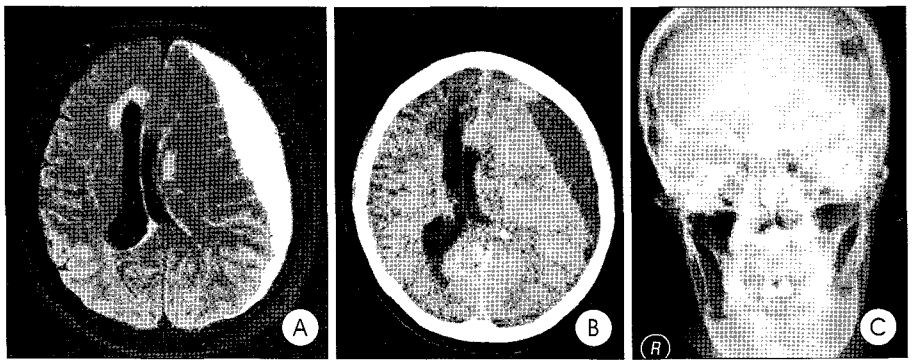

Fig. 1. A, B : Initial magnetic resonance and computerized tomographic images showing chronic subdural hematoma. C : Postoperative plain skull X-ray showing small craniotomy and an indwelling catheter in the subdural space.

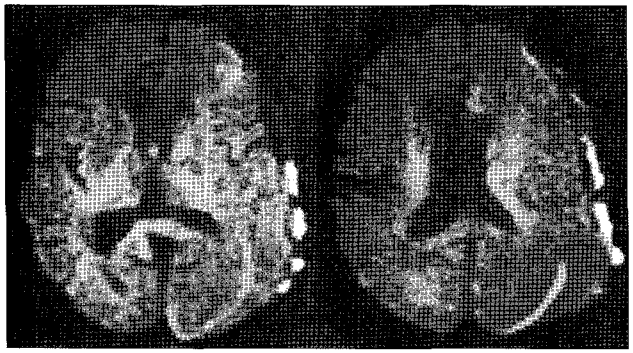

Fig. 2. Post-ictal diffusion magnetic resonance image showing no remarkable organic lesions except for small amount of residual subdural hematoma.

for one day. The authors presumed that her symptoms were ischemic in nature because of moyamoya disease, but magnetic resonance (MR) imaging showed CSDH in the left side intracranial space (Fig. 1A, B). Under local infiltrating anesthesia, the authors stripped galea from subcutaneous tissue and removed a small bone flap (5×3 cm) around left parietal eminence. A H-shape dura incision was performed, and on incising the dura liquefied blood gushed out. A drain catheter was then inserted under the subdural space (Fig. 1C). Dural openings were closed by suturing the dural edge and galeal edge of the donor strip on each side, and the bone flap was reapproximated with wires. Immediate after surgery, her clinical conditions were improved, and the authors prescribed valproic acid (300 mg t.i.d.) and stopped antiplatelet agents.

However, two days after surgery, her initial symptoms became aggravated, but emergency brain MR imaging showed no organic lesions other than a small amount of residual CSDH (Fig. 2). Also, there was no laboratory evidence of metabolic brain disease. The authors recommended a nasal oxygen supply and skipping antihypertensive drugs administration to maintain a moderate hypertensive state (around 150/100 mmHg). However, her clinical condition did not improve. Follow up brain MR imaging after 9 hours also produced no remarkable findings, and three days after surgery, she presented with a convulsive seizure of her right side arm and leg. The convulsion disappeared after a bolus injection of lorazepam (4 mg, i.v.); the duration of the convulsion was about one minute. The authors presumed that the untreated NCSE had produce the convulsive seizure. After injecting a loading dose of phenytoin (18 mg/kg), her manifestations (mutic slowing, clumsiness and right arm weakness) disappeared gradually, and one day after phenytoin loading, scalp EEG revealed diffuse continuous irregular slow waves. Follow up scalp EEG three days after phenytoin loading showed improved findings of intermittent slow waves. The author's did not check the ictal EEG because NCSE was not considered before the convulsion. Eight months after surgery, there was no evidence of ictus or neurological deficit.

Discussion

Definition and diagnosis

Towards the end of 19th century, Charcot described somnambulism patients, who are now believed to have been suffering from NCSE. However, NCSE was first described in the 1940s after the invention of EEG[12]. Nevertheless, there are no well constructed definitions of NCSE as compared with the international league against epilepsy (ILAE) system for convulsive seizures. In general, NCSE can be defined as a condition of altered mental status or behavior from baseline without convulsion, which lasts for more than 30 minutes with EEG evidence of seizures[6]. A response to antiepileptic drugs (AEDs) is helpful for diagnosis[2,7], but it should be noted that response to AEDs may take several hours or even days. "An altered mental status or behavior from baseline" differentiates NCSE from other epileptiform encephalopathies, such as, subclinical seizure, electric seizure and subtle status epilepticus[7]. These epileptic encephalopathies show epileptiform discharges on EEG that result from underlying brain damage with obscure clinical manifestations. On the other hand, epileptic discharge in NCSE causes direct neuronal damage with symptoms. Much animal data shows that epileptic discharge over 30 minutes may induce permanent neuronal damage and drug (AEDs) resistance. Therefore, seizures "lasting more than 30 minutes" should be regarded as status epilepticus and not as episodic seizures, which usually last two minutes or less[10]. This time criterion is also used in convulsive status epilepticus. Although, this definitions appears simple, its clinical implications are not obvious. Persons at risk of NCSE are those with behavioral

changes that may have been ascribed to other conditions, for example, to head trauma, stroke, a postoperative state, a postictal state of convulsive seizure, metabolic disorders, or drug intoxication. And behavioral changes may vary widely from subtle change of personality (loss of humor, reduced tendency to smile, acting strange etc.) to coma. Even among neurologists, "EEG evidence of seizures" is a topic of debate. In most instances, periodic lateralized epileptiform discharges are not interpreted as seizures. However, when clinical correlations are taken into consideration, the record may be interpreted as epilepsy[4]. A patient with hyperammonemia could show triphasic waves on EEG, and show improved clinical symptoms after benzodiazepine administration, and thus hyperammonemia could be confused with NCSE[4].

Classification and prognosis

NCSE has traditionally been divided into two groups : (1) the absence status epilepticus (ASE) or generalized NCSE, or (2) complex partial status epilepticus (CPSE) or focal NCSE[5,6]. But this classification does not reflect the underlying disease or symptom severity, and patients from slightly obtunded to comatosed might classified as same category. Thus, there is a need for more sophisticated classification, though there probably are as many types of status epilepticus as there are seizure types. The reasons for neurologic morbidity associated with convulsive status epilepticus are related to associated adverse systemic effects (e.g., hypotension, hypoxia, acidosis, and hypoglycemia) and direct neuronal damage resulting from ictal electrical discharge[11,15]. However, NCSE does not cause systemic effects, moreover ictal discharge in patients with ASE is inhibitory in nature; absence attack usually presented as brief motion arrest. On the other hand, most seizures present with hyperactivity such as tonus or clonus, and therefore, ASE is not believed to induce permanent neuronal damages. In CPSE, the lack of permanent neuronal deficit is debatable. Like ASE, CPSE does not cause systemic effects, though animal data demonstrate that electrical discharge without convulsion can cause permanent neuronal damage[9]. Some authors have advocated that this experimental data is applicable in humans and that the two conditions (NCSE and underlying acute brain injury) are synergistically detrimental and increase brain injury[3,7,8,17]. Conversely, animal data is not directly transferable to humans, because in these studies convulsive status epilepticus was induced without convulsions (e.g. for studies that utilized paralysis and physiologic controls)[2]. Moreover, the major neurologic sequela are usually due to the underlying insult rather than to NCSE per se[2,4,13]. Kaplan[4] advocated that NCSE should be stratified based on underlying pathophysiologies, and reported that NCSE in the elderly has a poor prognosis because of underlying causative processes and medical complications, whereas ambulatory patients show little morbidity. However, the treatment strategy for NCSE remains an issue of debate because its precise prognosis is unknown. Some have recommended aggressive treatment according to convulsive status epilepticus to prevent permanent neuronal damage[3,7], whereas other are of the opinion that the adverse effects of AEDs outweigh any obscure therapeutic gain, and thus, have advocated that aggressive treatment is contraindicated[2,4].

Our patient had hypertension and moyamoya disease and presented with CSDH. Bleeding was due to antiplatelet agents. After surgery, her clinical condition improved, but two days later her symptoms were aggravated and progressed to convulsive seizure within 24 hours. However, brain MR imaging and laboratory tests showed no evidence of culprit lesions. After administering an antiepileptic, these manifestations disappeared. Therefore, the authors presumed that in the described case symptoms have resulted from the nonconvulsive status epilepticus, despite a lack of ictal period electroencephalographic findings.

Conclusion

NCSE should be suspected and EEG checked in patients with an altered mentality or behavioral change without evidence of organic brain disease or metabolic encephalopathies. The authors stress that early diagnosis and proper medication probably minimizes the potential risk of NCSE.

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