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Case Report

Seizure frequency can be reduced by changing intracranial pressure: A case report in drug-resistant epilepsy☆

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ABSTRACT

A relationship between seizures and intracranial pressure (ICP) has been proposed, but not clearly identified. Whether changes in ICP can evoke seizures remains controversial. We report the case of a 23-year-old man who had undergone shunt surgery in childhood and later presented with focal impaired awareness seizures and behavior arrest. Seizures were uncontrolled despite 3 years of pharmacotherapy, but suddenly stopped after shunt removal. Our case supports the hypothesis that drug-resistant epilepsy can be influenced by changes in ICP. In particular, this case indicates that elevations in ICP may help reduce some seizures.

1. Introduction

All diseases affecting the brain are known to carry the potential to evoke epileptic seizures [1]. In particular, a relationship between seizure and intracranial pressure (ICP) has been proposed, but not clearly identified [1–4]. Most studies have confirmed that ICP increases significantly after seizure [2–4], but conversely, whether changes in ICP can evoke seizures remains controversial. Furthermore, many reports have described seizures in children with hydrocephalus, but to the best of our knowledge none have indicated a relationship between changes in ICP and drug-resistant epilepsy. We describe a patient with focal epilepsy in whom seizures decreased after a change in ICP.

2. Case report

Our patient was a 23-year-old, right-handed male born by vaginal delivery. He had no contributory family history. At 4 years old, a cerebellar tumor had been removed and diagnosed as medulloblastoma. He had undergone chemotherapy and radiotherapy, and a ventriculo-peritoneal (VP) shunt had been positioned in the right lateral ventricle. Postoperatively, he did not display any symptoms. He was taken off medications, grew up normally and entered university.

At 20 years old, he experienced a seizure for the first time in the form of behavioral arrest and staring for 60–120 seconds. He was unaware during these episodes and subsequently unaware that a seizure had occurred. From that point, seizures occurred approximately three times per week.

He visited our department 6 months after the first seizure. Electroencephalography (EEG) showed epileptiform discharges over the right temporal area. A variety of anti-seizure drugs was prescribed, including levetiracetam, lamotrigine and carbamazepine, but seizures were unable to be controlled.

We conducted simultaneous video-EEG recording to determine the focus of epilepsy and confirmed focal impaired aware seizures. Sharp-and- slow waves occurred over the right temporal area at electrode positions T2, T4 and F8 during seizures (Fig. 1). Magnetic resonance imaging (MRI) showed high T2/fluid-attenuated inversion recovery (FLAIR) intensity from the ventricle to the cortex in the right posterior-temporal lobe, representing a characteristic transmantle sign seen with focal cortical dysplasia (Fig. 2-A). No evidence of cerebral infarction was found, and brain perfusion appeared normal. Fluorodeoxyglucose position emission tomography (FDG-PET) showed hypometabolism in the right temporal lobe, corresponding to the focal abnormal lesion on MRI (Fig. 2-B). A multidisciplinary conference was held. Focal impaired awareness seizures with behavior arrest with a presumed etiology of right focal cortical dysplasia was diagnosed, according to the 2017 International League Against Epilepsy (ILAE) classification [5].

The VP shunt comprised an Integra™ LPVII™ fixed-pressure valve that had been placed on the right side of the head at 4 years old. The reservoir of the shunt system could not pump and was considered non-functional. We therefore removed the shunt prior to subdural electrode insertion, to reduce the risk of infection. During surgery, no exogenous

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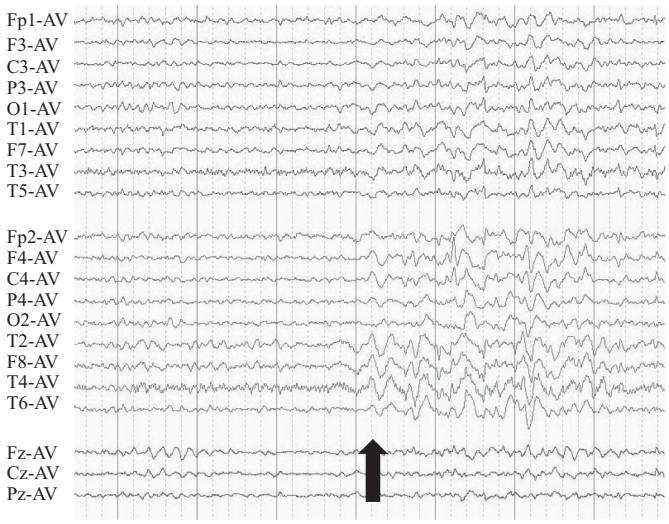

Fig. 1. Representative example of electroencephalogram (EEG). During simultaneous video-EEG recording, we confirmed four complex partial seizures. Sharp and slow waves always occurred from the temporal area at T2, T4 and F8 during the seizure (arrow). Interictal epileptiform discharges were also recorded at T2, T4 and F8.

material entered the field and no antibiotic solution was used for irrigation.

After shunt removal, seizures suddenly stopped but he complained of headache. Computed tomography (CT) revealed ventricular enlargement. Headaches worsened, so we performed VP shunt replacement 9 days after removal of the original shunt. Since the probability of complications associated with changes in intracranial pressure seemed high, the planning of intracranial electrode placement was canceled.

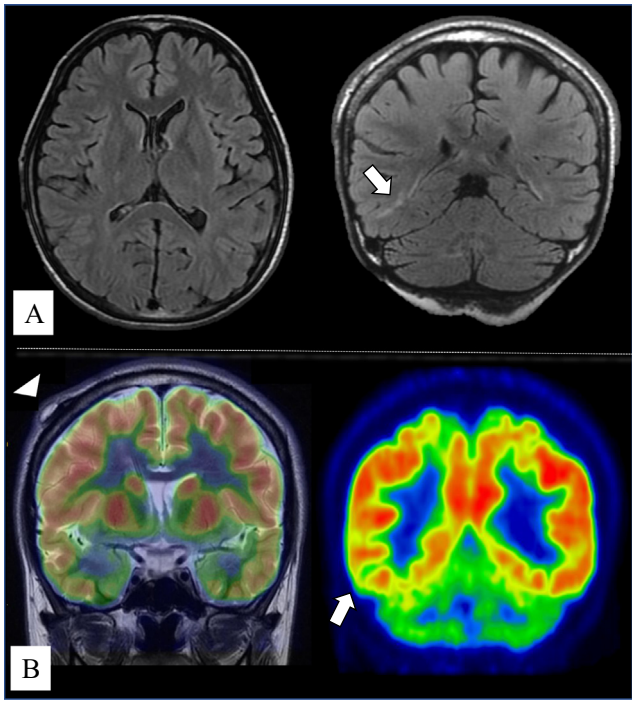

Fig. 2. Representative MR and ^{18}F-FDG PET images. A: MR images (FLAIR axial and coronal) show an abnormal hyperintense signal (arrow) extending from the ventricle to the cortex in the right posterior-temporal lobe. No other abnormalities (including cerebral infarction) are seen, and cerebral perfusion appears to have remained normal. B: Images from ^{18}F-FDG PET show hypometabolism in the right temporal lobe (arrow), corresponding to the focal lesion on MRI. No abnormal metabolism is seen in other areas, including the vicinity of the shunt catheter (arrowhead).

Headaches gradually disappeared along with good decompression of the ventricles. Seizure frequency dramatically decreased, and only lamotrigine was continued at discharge from our hospital.

We presumed that changes in ICP and seizure recurrence were closely related, and so gradually raised the opening pressure of the shunt valve, paying attention to the presence of headache. Seizure has since been well controlled coinciding with pressure adjustments (Fig. 3). Only perampanel was added to control residual seizures at 12 months after discharge, although frequency of seizures was significantly decreased compared to pre-operatively.

3. Discussion

This case indicates that changes in ICP and focal epilepsy seem to have a close interaction. Seizures were unable to be controlled for 3 years with appropriate pharmacotherapy, but stopped just after the shunt removal.

Many publications have suggested some correlation between seizures and ICP, particularly from the perspective that seizures can increase ICP. One hypothesis is that the increase in metabolism caused by seizures results in depletion of energy reserves. To compensate for this, cerebral blood flow increases and thus raises the ICP [1,2,4,6]. Some reports from animal experiments have confirmed sudden rises in ICP during seizures [3,7].

In contrast, whether changes in ICP can cause subsequent seizures remains controversial. Idiopathic intracranial hypertension is a rare syndrome characterized by isolated elevation of ICP of unknown cause. Common symptoms include headache, nausea, vomiting, pulsatile tinnitus and visual disturbance, but seizures are uncommon in these patients [8,9]. This fact may indicate that seizures are not caused by elevations in ICP. On the other hand, some reports have concluded that intracranial hypotension subsequent to cerebrospinal fluid (CSF) leakage could be a cause of seizure [10,11]. In addition, Agrawal et al. reported a patient suffering from frequent postural seizures [12]: seizures occurred only when sitting up, and resolved when lying down. The patient had undergone VP shunt placement in infancy, and seizures resolved after changing the shunt valve to one with a higher opening pressure. These reports may indicate that intracranial hypotension predisposes to seizures.

The present case suggests that intractable epilepsy can be influenced by changes in ICP. Particularly in this case, elevations in ICP might reduce seizures. Many reports have considered the relationship between ICP and seizures, but almost all have looked at children with hydrocephalus or patients with brain trauma. We could not find any reports about patients with drug-resistant epilepsy. Our case indicates that some cases of drug-resistant epilepsy also have a relationship with ICP, although the mechanisms underlying such a relationship are not well understood.

The Monro-Kellie hypothesis states that the sum of volumes of the brain, CSF, and intracranial blood is constant [13]. As a result, any increase in CSF volume is accompanied by a decrease in intracranial blood flow. Seizures are associated with increased blood flow, so increased ICP, and hence decreased intracranial blood flow, might have reduced the frequency of seizures in the present case.

Moreover, the brain tissue microstructure or mechanical properties are known to change with the development of obstructive hydrocephalus, leading to changes in brain stiffness [14]. Increased brain stiffness may amplify the effects of changes on ICP. Shunting is also thought to lead to brain stiffening. These factors may explain why subtle changes in ICP dramatically affected the frequency of seizures in our patient.

In this case, the shunt could potentially have provoked seizures independent of ICP, although the mismatch in locations of the shunt site and epileptic discharges do not seem support such a relationship. Moreover, Bourgeois et al. [15] reported that in the majority of 255 children with seizure and hydrocephalus, the first seizure occurred within 1 year after shunt insertion. In the present case, the first seizure did not occur until 16 years after shunt insertion. For these reasons, we

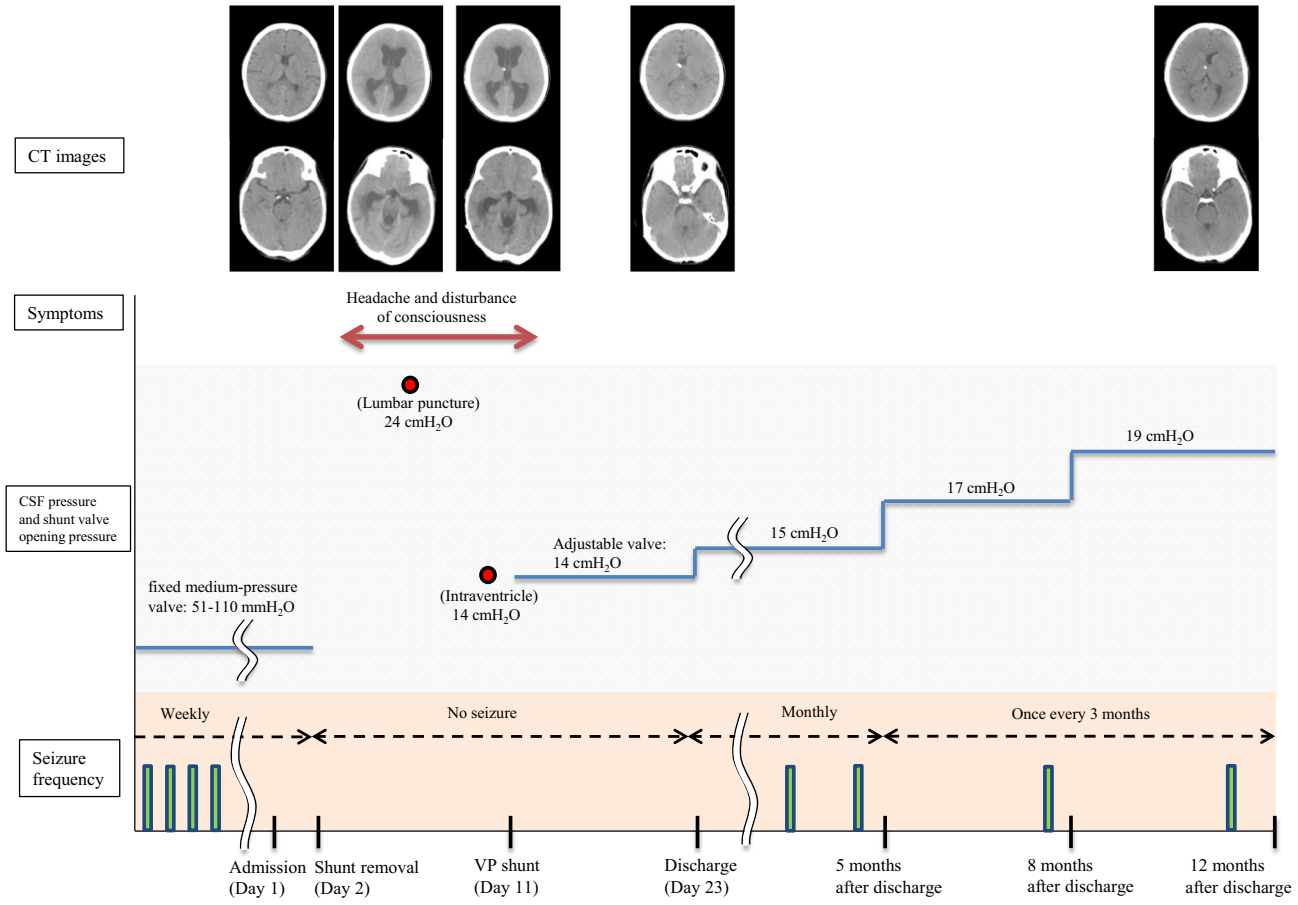

Fig. 3. Summary of the clinical course of the patient. Seizures were occurring three times a week at the time of the first visit to our department. His seizures were uncontrolled despite 3 years of pharmacotherapy. To reduce the risk of infection for further epilepsy surgery, we removed the old shunt. After removal (Day 2), seizures suddenly stopped, although he instead complained of headache. Ventricular enlargement was revealed on CT, and indicated increased intracranial pressure. We therefore performed VP shunt replacement (Day 11). Headaches gradually disappeared along with good decompression of the ventricles. Frequency of seizures was significantly decreased compared to pre-operatively, and only lamotrigine was continued as of discharge from our hospital. Seizures have since remained well controlled (about once every 3 months) with pressure adjustments.

think the shunt itself was hard to consider as the main cause of seizures in this case.

Some limitations must be acknowledged. First, this case did not represent planned research, and we thus could not measure ICP continuously throughout the clinical course. Second, although epileptiform discharges were confirmed in the temporal region far from the operated site, whether shunt surgery itself might have affected seizure occurrence is unknown. Finally, this is a case in which the results cannot be generalized to a wider population. Additional tests should be performed to clarify the details of mechanisms connecting changes in ICP to drug-resistant epilepsy.

The relationship between seizure and ICP is clearly complicated, but our case supports the hypothesis that drug-resistant epilepsy can be influenced by changes in ICP. In particular, we assume that elevations in ICP might help reduce some seizures, although the precise mechanisms remain unclear. Given our results, for patients suffering from drug-resistant epilepsy who have a shunt, we suggest attempting an adjustment of shunt valve pressure to a slightly higher level before prescribing additional drugs. This option is easily tried and non-invasive for patients.

4. Conclusion

We report a patient with focal epilepsy in whom seizures decreased after an increase in ICP. The clinical progress of the case supports the hypothesis that drug-resistant epilepsy can be influenced by changes in ICP. In particular, our case indicates that elevations in ICP might help reduce some seizures. Further research is needed to clarify the detailed relationship between ICP and focal epilepsy.

Disclosure

The authors report no conflicts of interest concerning the materials or methods used in this study or the findings specified in this paper.

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