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Case Report

Dense array EEG estimated the epileptic focus in a patient with epilepsy secondary to tuberous sclerosis complex

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Abstract

Purpose: Tuberous sclerosis complex (TSC) is a leading cause of epilepsy, with seizures affecting almost 80–90% of children. We used the concordance between magnetic resonance imaging (MRI) and dense array electroencephalography (dEEG) findings to detect epileptic focus in a patient with TSC.

Methods: A 9-year-old boy with TSC exhibited daily choking spells. As we could not detect the seizure onset area with conventional scalp electroencephalogram (EEG) and long-term video monitoring, we performed dEEG and captured his regular seizures.

Results: dEEG estimated that the clinical seizure activities from the right frontal region. This patient underwent focus removal, tuberectomy of the right frontal lobe, and removal of a subependymal giant cell astrocytoma. He has been seizure free for 7 years and 10 months.

Conclusion: dEEG was useful for estimation of the placement of intracranial electrodes in a patient with TSC.

This method may be useful for pre-surgical evaluation of epilepsy treatment.

Keywords: Tuberous sclerosis complex; Epilepsy; Dense array EEG; Spatial resolution; Epileptogenic zone

1. Introduction

Tuberous sclerosis complex (TSC) is a leading cause of epilepsy, with seizures affecting almost 80–90% of children [1]. Only one-third of these patients will achieve freedom from seizures with antiepileptic medication [2]. Even though surgery can be effective [3], for individuals with multiple epileptogenic foci, achieving freedom from seizures without adverse effects from surgical treatment might not be easy in TSC populations. As is well known in epilepsy surgery, the greater the concordance of information, the better the results tend to be [4]. However, as discordant neuroimaging, neurophysiological and clinical findings are commonly seen in TSC, we generally seek for some concordance of information to perform surgical intervention.

Dense array electroencephalography (dEEG) is a method in which electroencephalogram (EEG) is recorded using more electrodes, up to 256 channels, than are utilized with standard techniques that typically employ 19–21 scalp electrodes. dEEG recordings of seizure onset have shown good predictive ability for seizure localization [5].

We used the concordance of magnetic resonance imaging (MRI) and dEEG findings to detect seizure onset in a patient with TSC.

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Ethics approval

Written informed consent for publication of case details was obtained from the guardian. This study was approved by the ethics committee at Seirei Hamamatsu General Hospital.

2. Patient and methods

The patient was a 9-year-old boy with TSC. He exhibited choking spells followed by loss of awareness. The seizure frequency was three to 10 times per day. He had a severe intellectual disorder with a developmental quotient 17 level.

His regular seizures were captured by scalp long-term video monitoring. However, we could not identify from where these seizures arose due to a nearly generalized amplitude attenuation pattern. The interictal scalp EEG showed diffuse high amplitude spike-waves in the bilateral frontal regions. Therefore, the patient underwent 256-channel dEEG (Electrical Geodesics, Inc., Eugene, OR, USA), that covered the face and neck as well as the cranium.

We employed this technique for recording seizures, with recordings of a duration of 6 h. The EEG amplifier characteristics included a band-pass of 0.1–400 Hz and sampling rate of 1000 Hz. The average referenced dEEG waveforms were examined with topographic waveform plots, a method that allows inspection of the geometric distribution of the potential fields. We analyzed the interictal spikes based on the linear inverse method of local autoregressive average, and projected the Montreal Neurological Institute Typical Brain as probabilistic cortical gray matter. We also analyzed the ictal EEG onset with visual inspection.

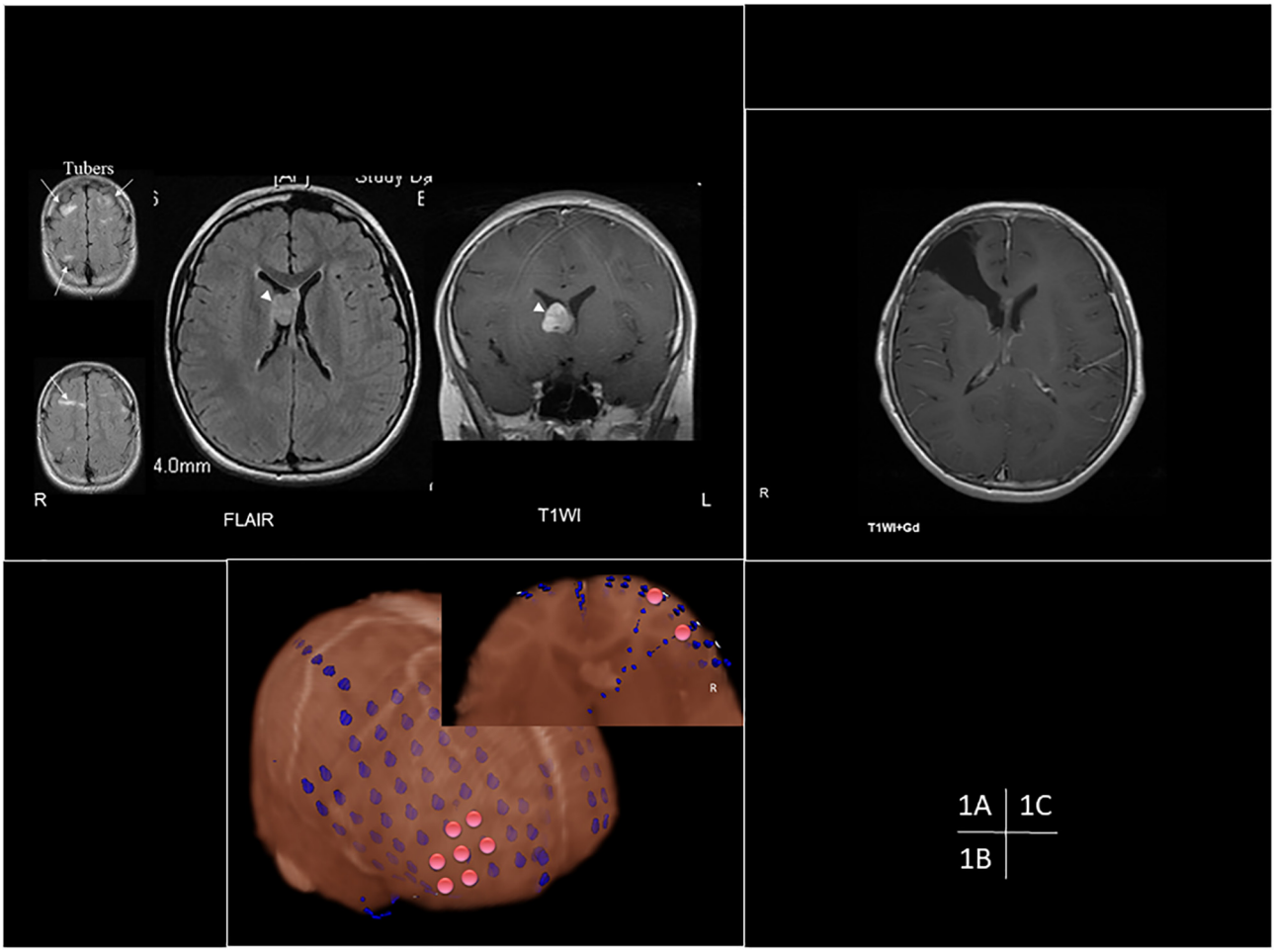

Fig. 1. Brain magnetic resonance image (MRI). Multiple cortical tubers were seen (arrows). The largest tuber was in the right frontal lobe. Gadolinium-enhanced MRI showed a subependymal giant cell astrocytoma near the right foramen of Monro (1A). Intracranial electrode monitoring and depth electrode monitoring showed that the seizure arising from the right frontal lobe (pink circle). The area which pink dotted circle showed high frequency oscillations at the beginning of the seizure (1B). This patient underwent resection of the right frontal focus, tuberectomy, and removal of the subependymal giant cell astrocytoma. He has been seizure free for more than 7 years. MRI showed the right frontal cortical resection, right frontal lobe tuberectomy, and removal of the SEGA (1C). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

3. Results

MRI of this patient revealed multiple tubers and a gadolinium-enhanced subependymal giant cell astrocytoma (SEGA) near the right foramen of Monro. One of the largest tubers was found in the right frontal lobe (Fig. 1A).

Interictal dEEG detected right frontal epileptiform discharges and left frontal epileptiform discharges independently (Fig. 2). Ictal dEEG showed that his regular seizures were indicative of a right frontotemporal focus (Fig. 3). Based on these findings, we estimated that the region of seizure onset was the right frontotemporal region.

Subsequently, the patient underwent extra-operative subdural electrode monitoring. The high frequency oscillation which evolved to beta activities about 1 sec before the clinical manifestation was detected in his right frontal lobe near the largest tuber. We regard this area as possible epileptogenic zone [6]. He underwent resection of the right frontal focus, right frontal lobe tuberectomy, and removal of the SEGA. The patient has been seizure free for 7 years and 10 months.

Histopathology revealed large heterotopic neuronal proliferation resulting in grotesque cells, balloon cells, and gliosis in the cortex. The number of neurons, which were mainly composed of balloon cells, was increased in the white matter.

4. Discussion

Brain tissues includ~~ed~~ dysplastic lesions composed of giant cells, misaligned dysmorphic neurons, and atypical astrocytes. These abnormal neuronal populations exhibit reduced GABAergic inhibition and are thus intrinsically epileptogenic [7]. Large tubers are reportedly more likely to be an epileptogenic focus compared to smaller tubers [8]. In this case report, the tuber we removed was the largest tuber and was concordant with these reports.

Several investigators have characterized the EEG patterns of TSC. Westmoreland examined EEG findings from 361 patients [9]. A high incidence of abnormalities was identified, with 78% of the patients having epileptiform features. Approximately 12% of the population had a normal EEG, and 10% had slow abnormalities.

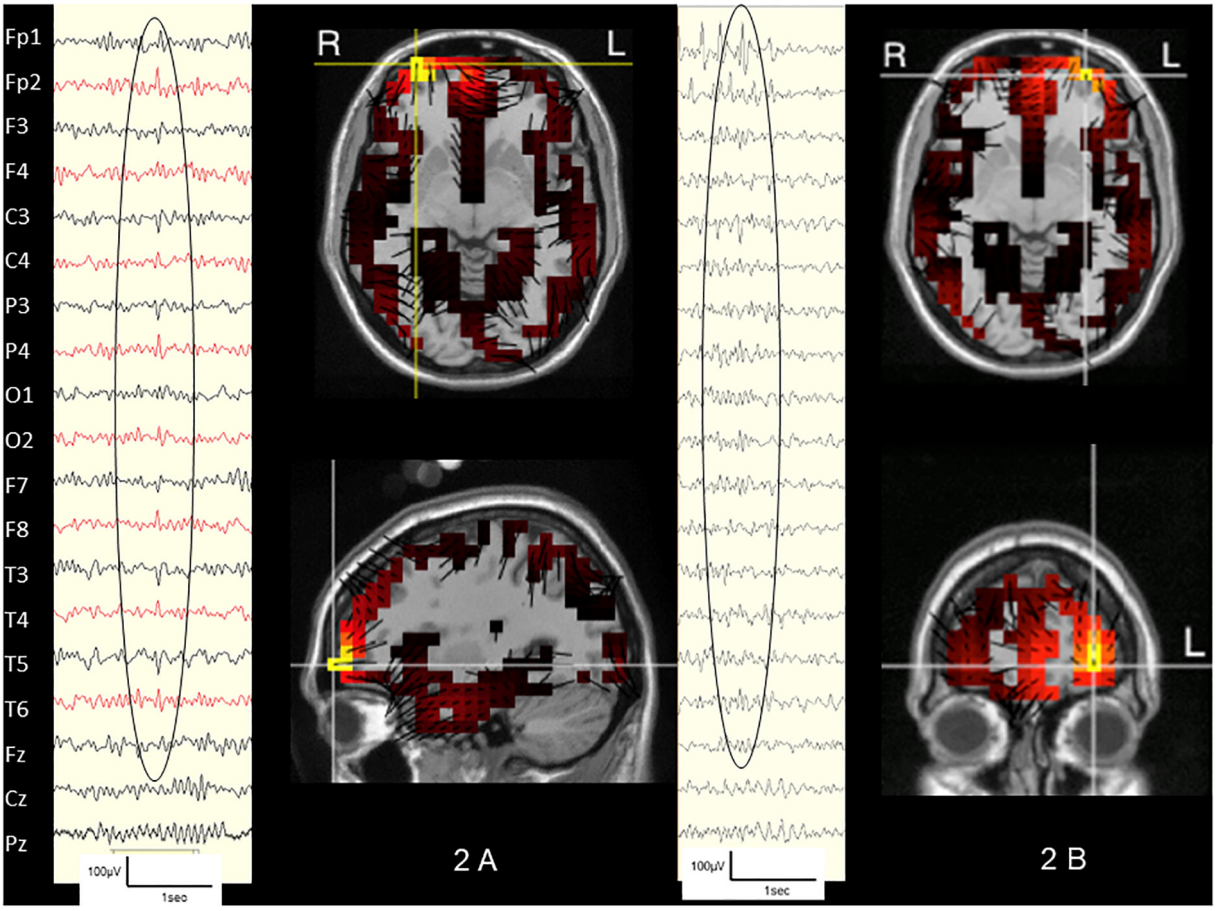

Fig. 2. Interictal dense array electroencephalogram (dEEG) findings Scalp EEG shows diffuse spike-waves with frontal predominance. This spike source was estimated with dEEG, which was superimposed on a standard MRI that showed the right frontal focus (2A) and left frontal focus (2B).

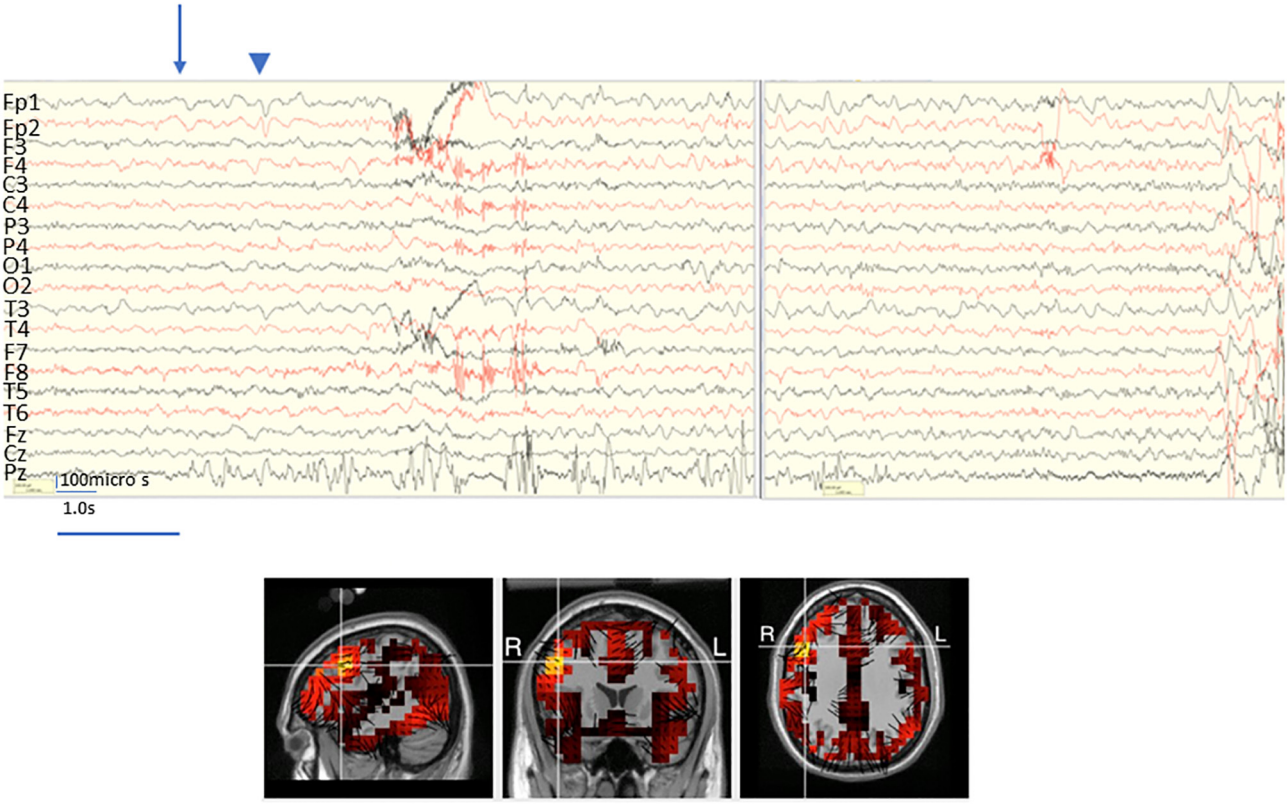

Fig. 3. Ictal electroencephalogram (EEG) change. There were generalized amplitude attenuation for about 3 s (underlined) followed by rhythmic delta activities with sharp waves (arrow). At the time of this rhythmic delta activities, the patient stated to exhibit clinical manifestation. These activities change the morphology to the rhythmic theta activities. The beginning of this ictal activities analyzed by dEEG is suggestive of right frontal focus (Arrow head).

Of those with epileptiform features, 35% had focal epileptiform discharges, 33% had multifocal discharges, 22% had hypsarrhythmia, and 10% had generalized discharges. A similar study of 60 patients showed that 84% of the recordings were diffuse slowing [10]. Thus, many of TSC patients with epilepsy do not have only a single focus as determined with conventional EEG.

Conventional scalp EEG recordings utilize the international 10 to 20 system in which only 19 to 21 electrodes are typically applied to the scalp. This method results in interelectrode distances of several centimeters and thus produces relatively poor spatial resolution. As a consequence, the initial noninvasive EEG evaluation of the patient is sometimes inconclusive [11]. According to Tao et al. [12], conventional scalp EEG requires an area of at least 10 cm^2 of cortex to display an epileptic discharge. In contrast, Yamazaki [5] suggested that dEEG requires an area of only 5 cm^2 of synchronized activity. Therefore, dEEG could obtain the seizure onset area quicker than the scalp EEG before the vast spreading of the epileptiform discharges.

The concordance between the largest cortical tuber of the brain MRI and dEEG findings lead to seizure freedom of a pediatric patient with TSC. Therefore, dEEG might be one of the modalities to seek for concordance of presurgical information.

5. Conclusion

dEEG was useful for estimation of the placement of intracranial electrodes in a patient with TSC. dEEG may be useful for pre-surgical evaluation of epilepsy treatment.

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