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Case Report
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High-Frequency Oscillations Led to the Diagnosis of Frontal Lobe Epilepsy in a Case of Large Corpus Callosum Lipoma: A Case Report

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Abstract

This case involves a 15-year-old girl who had first experienced brief attacks of impaired consciousness when she was 14 years old. Subsequently, frequent occurrence of similar seizures accompanied with occasional foot stomping caused her to visit our hospital. Although there were no abnormal neurological findings, head computed tomography and magnetic resonance imaging revealed a 50 mm × 35 mm × 35 mm intracranial tumor deep in the interhemispheric fissures of the bilateral frontal lobe. The initial Electroencephalogram (EEG) did not show any apparent epileptic spikes, but High-Frequency Oscillation (HFO, 80 Hz) was frequently observed, mainly in the bilateral fronto-parietal lobes. Frequent unconscious fits with occasional foot stomping were observed; therefore, we diagnosed her with frontal lobe epilepsy. She was started on levetiracetam 1000 mg/day, and the clinical seizure was well controlled. Since surgical removal of the large tumor of our case may cause blood flow damage in the posterior half of the interhemispheric fissure, conservative treatment was indicated. Although there are previous reports of lipomas in the corpus callosum, there are no reports of epilepsy cases with HFO recordings until now. However, rich vascularization of the large corpus callosum lipoma cannot neglect the future risk of tumor enlargement, malignant transformation or seizure recurrence, periodic follow up is indicated for further work up including hospitalization for possible surgical indication. We report a case of large corpus callosum lipoma in which HFO was useful to diagnose frontal lobe epilepsy.

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Introduction

Intracranial lipomas are rare tumors that account for less than 0.5% of all primary brain tumors. They often occur near the midline of the brain, and the corpus callosum is one of the main sites of occurrence [1-3]. Most intracranial lipomas are asymptomatic and discovered incidentally during diagnostic imaging. In symptomatic cases, epileptic seizures are a commonly reported symptom [3-5], but there have been no case reports of Electroencephalogram (EEG) recordings in corpus callosum lipomas. EEG is an important test in the diagnosis of epilepsy, and epileptic spikes have been the most common epileptogenic biomarkers. However, recently, High-Frequency Oscillations (HFOs) were expected to become useful as well as epileptic spikes as biomarkers used to identify epileptogenic zones, not only by intracranial EEG recordings but also by conventional scalp recordings. In this report, we present a case of large corpus callosum lipoma in which HFOs led to the diagnosis of epilepsy.

Case Presentation

The patient was a 15-year-old female with no history of heat cramps in infancy, no epilepsy cases in her parents or relatives, and no problems with growth or intellectual disabilities. She was actually preparing entrance examination for public high school. Her first seizure occurred when she was 14 years old. During a conversation with a friend at lunchtime in a school classroom, she suddenly lost consciousness and fell backward. The second seizure occurred during the opening ceremony of a sports game when others saw her to be in a foot stomping manifestation during unconscious fits. Since there were no symptoms of headache or nausea after regaining consciousness during daytime in her school, she was not taken to the emergency room and did not seek medical attention. However, she continued to have occasional attacks of brief impairment of consciousness, which occurred one to three times a month with occasional foot stomping; therefore, she visited our hospital.

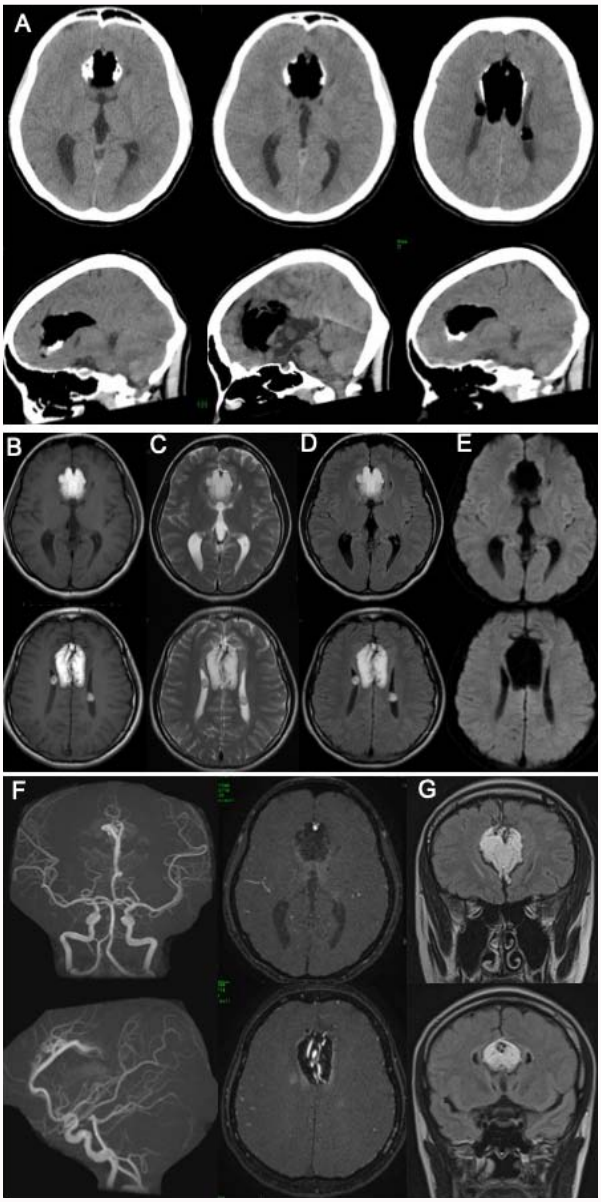

Figure 1: CT: A sizeable fatty tumor, measuring 50 mm × 35 mm × 35 mm, was located deep in the bilateral interhemispheric fissures of both frontal lobe, and peri-tumoral calcifications were observed (A). MRI: T1-weighted images (B), T2-weighted images (C), and FLAIR (D, G) show high intensity. Diffusion-weighted images (E) show low intensity. Multiple small lipomas were observed in the bilateral lateral ventricles.MRA (F) shows that the anterior cerebral artery of A2 portion was bifurcated at A3 with Azygos formation. A2 and A3 were entrapped in the tumor.

Neurological findings at the time of initial examination did not reveal any abnormal findings including intellectual performance. Head CT showed a 50 mm × 35 mm × 35 mm large, fatty intracranial tumor located deep in the interhemispheric fissures of bilateral frontal lobe. Calcification was also observed around the tumors. On 1.5T MRI the tumor showed high intensity on T1- and T2-weighted images, high intensity on Fluid-Attenuated Inversion Recovery (FLAIR), and low intensity on diffusion-weighted images with the multiple small tumors in the bilateral ventricles; thus, they were diagnosed as multiple lipomas. The medial aspect of the bilateral frontal lobes is strongly compressed bilaterally by the lipoma. Anterior part of the corpus callosum was almost replaced by the big lipoma. Fortunately, bilateral foramen of Monro was still intact and there observed no dilated lateral ventricles. Associated cortical dysplasia or migration disorder was not observed in the present case. Disconnection syndrome of corpus callosum was also not observed in this case.MRA showed that the anterior cerebral artery of A2 portion was bifurcated at A3 with Azygos formation and that A2 and A3 were entrapped in the center of tumor. Moreover, rich blood flow into the tumor was also observed (Figure 1). The frequent impairment of consciousness was suspected to be due to epileptic seizures, and conventional digital EEG was performed. The conventional digital EEG was a Neurofax EEG-1218 made by Nihon Kohden, with a high-cut filter at 120 Hz and a time constant of 0.3 s. Although there was no apparent epileptic spike in EEG recording, frequent HFOs of over 80 Hz were observed mainly in the bilateral fronto-parietal lobes; Duration of HFOs were less than 1sec (Figure 2). We suspected epileptic HFO and diagnosed the patient with frontal lobe epilepsy because of frequent clinical seizures of short loss of consciousness with occasional foot stomping. Valproic acid was considered the first choice, but as the patient was close to the gestational age, levetiracetam 1000 mg/day was started. Serum level of levetiracetam was (22.70 μg/ml (12-46 μg/ml)). Follow-up EEGs were obtained several times afterwards, and HFOs were continuously observed in all of them. However, the clinical epileptic seizures disappeared, and the clinical seizure was well controlled after the start of the medication. This case was considered to have abundant blood flow from the bilateral A2 on MRA, and since surgical removal of the tumor threatened to result in blood flow injury in the posterior half of the interhemispheric fissure with no enlargement of lateral ventricles until now. As she was facing toward the entrance examination of public high school, further examinations which require hospitalization were not indicated by request from her family. However, rich vascularization of the large lipoma cannot neglect the future risk of tumor size development, worsen seizure development or malignant transformation of the lipoma, we explained necessity of periodic follow up at outpatient clinic to her family such as further work up including hospitalization for surgical indication if necessary. Consequently, conservative treatment was continued with regular outpatient consultations and examinations to monitor the possible progress of epilepsy and corpus callosum lipoma.

Discussion

Intracranial lipomas are rare intracranial tumors, accounting for less than 0.5% of all primary brain tumors. They occur near the midline of the brain and often in the cerebral vault. The most common locations are the corpus callosum, quadrigeminal cistern, suprasellar cistern, cerebellopontine angle cistern, and sylvian cistern [1–3]. Most intracranial lipomas, including corpus callosum lipomas, are asymptomatic and are often discovered incidentally on imaging of other brain diseases. In symptomatic cases, epilepsy has been reported [3–5], but there have been no case reports of EEG recordings in corpus callosum lipomas. In this case, epileptic seizures with brief impairment of consciousness with occasional foot stomping position were observed, and we diagnosed the patient with frontal lobe epilepsy because of the symptoms of frequent clinical seizures. A 1.5T MRI scan showed intracranial lipomas in the deep part of the bilateral frontal interhemispheric fissures. There was no apparent spike-wave on scalp EEG at the first outpatient visit. It has also been reported that frontal lobe epilepsy is more common in patients with complex seizure symptoms and is less likely to show abnormal epileptic spikes on EEG. In our case HFOs were observed in the bilateral fronto-parietal lobes of the brain. In normal conventional

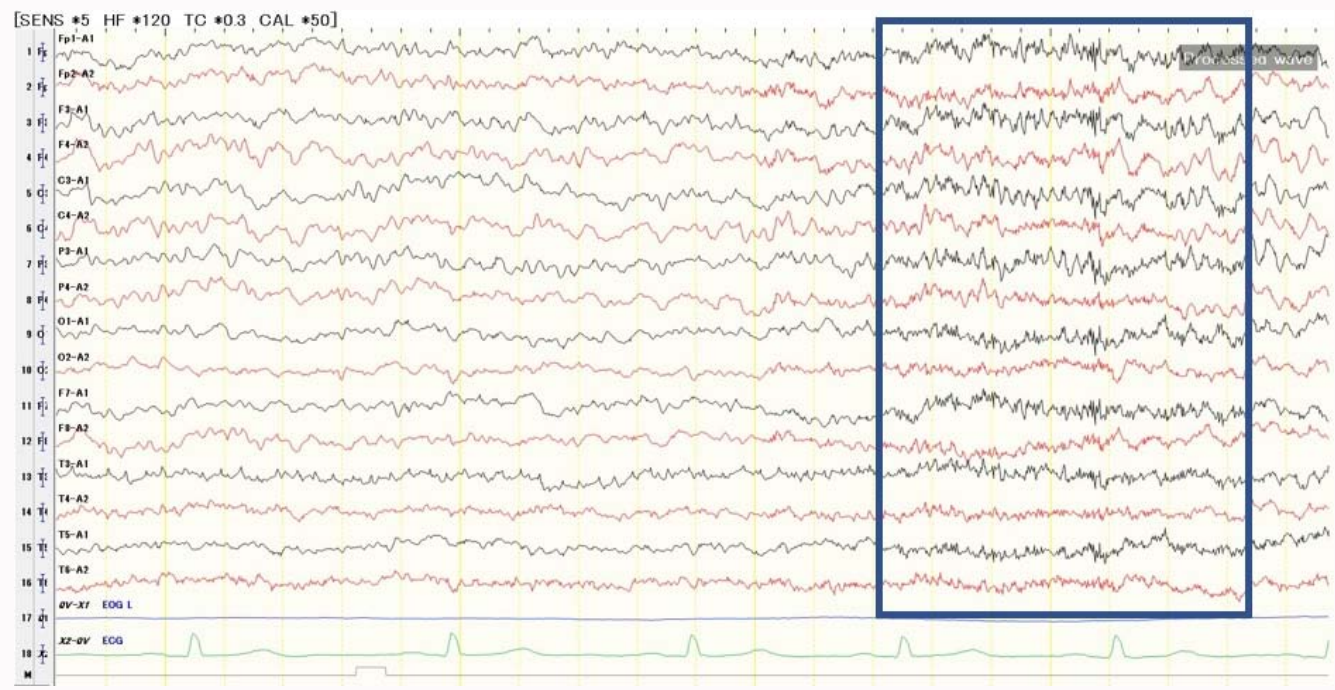

Figure 2: An HFO of over 80 Hz was observed mainly in the bilateral fronto-parietal lobes on conventional digital EEG.

digital EEG, the activity is mainly up to approximately 40 Hz (δ-β band) and sometimes ranges from 40 Hz to 80 Hz (gamma band). The EEG activity above the gamma band at 80 Hz is called HFO. Of the HFOs, the activities between 80 and 200 Hz are called ripples, and those above this range are called fast ripples [6,7]. HFO can be divided into "physiological activity" in cognitive processes and "pathological activity" found in the brains of patients with epilepsy, mainly MEG recording and intracranial EEG recordings. Pathological HFO is a biomarker that is expected to be useful in identifying epileptogenic regions as well as the conventional biomarker, epileptic spikes [8]. During interictal EEG recordings, HFO is usually intermittently recorded as a short activity. Recording with intracranial electrodes in patients with epilepsy has been reported, and it has been shown that HFO can be recorded during interictal periods not only with intracranial EEG but also with scalp EEG [7]. By using intracranial subdural or depth electrodes, which can cover a wide area of the brain, it has been reported that in the majority of patients, HFO is present not only at the seizure origin but also in the surrounding area [6,9]. In our case, the intracranial lipoma was located deep in the interhemispheric fissures of bilateral frontal lobe, and HFOs with a duration of less than 1s were frequently observed in the conventional digital EEG in the bilateral fronto-parietal lobes, suggesting that remote HFO waves from the widely spread zones epileptic zones in the bilateral frontal lobe interhemispheric fissures in contact with the tumor were recorded on the conventional digital EEG. Symptoms during seizures in frontal lobe epilepsy depend on the area of the cerebral cortex that is activated during seizures. There are focal tonic seizures, bilateral asymmetric tonic seizures, complex motor seizures, and a rare seizure type characterized by a brief loss of consciousness with foot stomping. The patient in this case was diagnosed with frontal lobe epilepsy due to the appearance of seizures with impaired consciousness and repetitive movements, such as stomping of the feet. Valproic acid was the first therapeutic choice, but since she was a 15-year-old female, levetiracetam 1000 mg/day was later started and continued. Since then, the patient has been followed up with several EEG examinations, all of which have shown HFO findings, mainly in the bilateral fronto-parietal lobes, but which have been well controlled with no clinical epileptic seizures since the start of medication. It is interesting to note that levetiracetam administration can suppress clinical epileptic seizures but HFOs were still recorded even after the treatment. MRA showed that the anterior cerebral artery A2 was bifurcated at A3 with Azygos formation and that A2 and A3 were entrapped in the tumor, suggesting a tumor with rich blood flow. Therefore, surgical removal of the tumor may result in blood flow injury in the posterior half of the interhemispheric fissure. Therefore, although many previous reports have been pessimistic about the indications for surgery [1,3,10], surgical removal of the tumor should be considered if seizures are not well controlled by antiepileptic drugs or interhemispheric lipoma develops to symptomatic lipoma. The patient had a large lipoma with multiple congenital anomalies such as bilateral intraventricular lipomas and Azygos formation in ACA, but normal development was observed except for frontal lobe epilepsy. She is currently under good control with antiepileptic medication, and preparing entrance examination for public high school, further advanced examinations require hospitalized examination were denied by patient and family. Our team continues to monitor her for tumor growth and epilepsy progress and follows up with regular outpatient examinations.

Conclusion

Considering the scarcity of literature covering the topic of seizures associated with corpus callosum lipomas, including seizure semiology, epilepsy course, and response to treatment, this case report could be a useful addition to existing knowledge. We also reported that the appearance of HFOs on scalp EEG should be noted in addition to epileptic spikes in cases of corpus callosum lipomas.

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