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Case Report

Successful surgery in lesional epilepsy secondary to posterior quandrant ulegyria coexisting with benign childhood focal epilepsy: A case report

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ABSTRACT

The present study reports, for the first time, a rare case of benign childhood focal epilepsy(BCFE) coexisting with lesional epilepsy secondary to parietooccipital ulegyria. The patient underwent right parietooccipital lobe disconnection plus tailored resection of temporooccipitoparietal junction cortex under electrocorticography (ECoG) monitoring. Post-operatively, there was no impairment of neurological function and the patient only experiences a few breakthrough benign partial seizures during sleep.

1. Introduction

Benign focal epileptiform discharges of childhood (BFEDCs)is a form of genetically determined, age related electroencephalographic abnormity, and about 8.8% of these children who experience clinical epileptic seizures were diagnosed as benign childhood focal epilepsy (BCFE) [1]. Patients with BCFE are characterized by low seizure frequency, easy medication control and self-healing in puberty. About 64% of the BCFE patients suffer from attention-deficit and hyperactivity disorder, severely impairing their behavior, learning ability and normal lives. A few paper have reported that a subset of patients with BFEDCs or BCFE could suffer from lesional focal epilepsy simultaneously, which can be the cause of "intractable" BCFE [2–5]. Here, we report a child with BCFE coexisting with intractable focal epilepsy secondary to parietooccipital ulegyria. The patient underwent right parietooccipital lobe disconnection plus tailored resection of temporooccipitoparietal junction cortex. Encouraginly, the patient experienced only three breakthrough seizures of BCFE with oxcarbazepine (OXC) monotherapy for two years postoperatively.

2. Case report

We present a case report of coexisting intractable focal epilepsy secondary to parietooccipital ulegyria and BCFE with the parents' permission. This 7-year-old right-handed boy initially suffered from refractory epilepsy at the age of 4. His habitual seizure consisted of an aura of blurred vision, followed by blinks, eyes deviating to the left, stiffness in the left upper limb and loss of consciousness, lasting for 30 s–1 min in total. Although the patient was given sodium valproate, Levetiracetam, OXC, clonazepam, topiramate and lamotrigine, as monotherapy or combinations, the seizures still occurred 6–7 times per day. The patient suffered from "hypoxic ischemic encephalopathy" on the postnatal 3rd day. His uncle and father suffered from epilepsy in childhood and self-healed in puberty. The patient's general and neurological examinations were normal except for the left homonymous hemianopia. Wechsler Intelligence Scale for Children revealed a moderate defect in the patient, as verbal intelligence quotient (IQ) 86, performance IQ 77, and full scale IQ 80, respectively.

The high-resolution 3-T magnetic resonance imaging (MRI) showed abnormalities in the bilateral parietooccipital lobe, sug-

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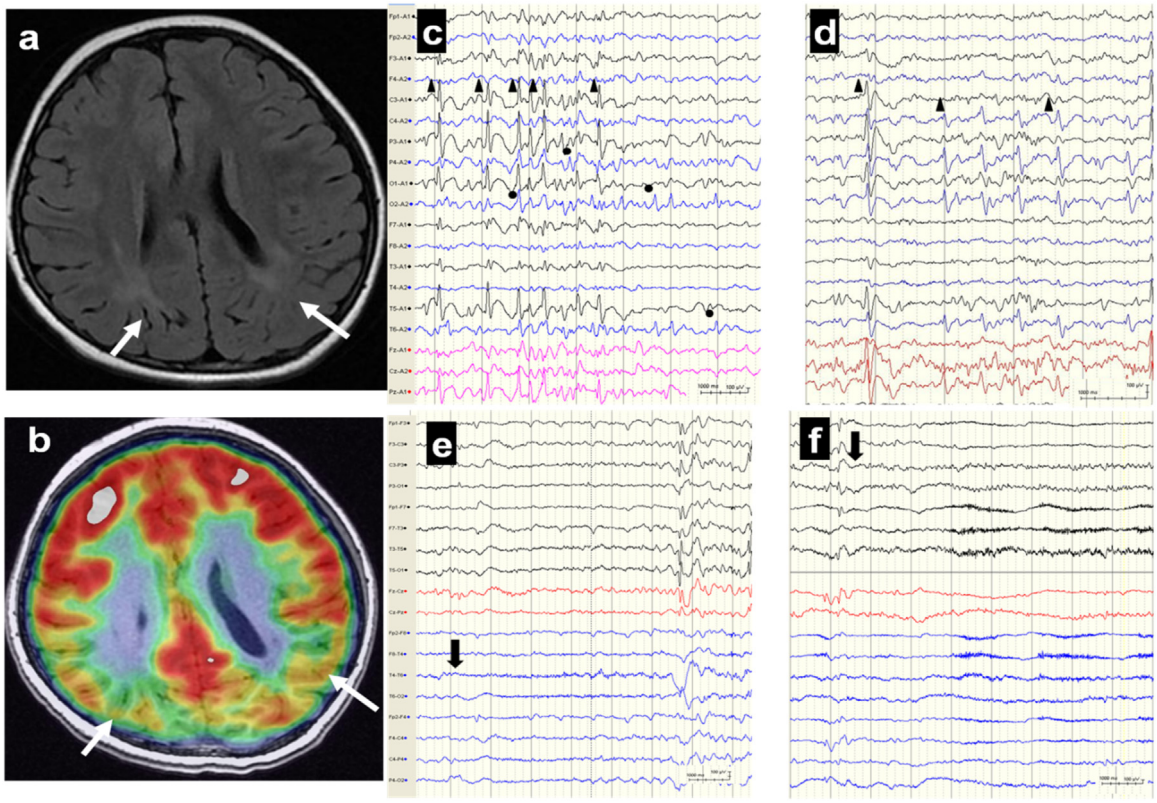

Fig. 1. Presurgical MRI, PET-MRI, and s-VEEG. (a) Axial T2-FLAIR shows bilateral parietooccipital "mushroom" like gyri prominently on the right side (arrow). (b) PET-MRI reveals hypometabolism at parietooccipital lobe, prominently on the right side (arrow). (c,d) Interictal EEG show two different kinds of interictal discharges (black circles and black triangles). (e,f) The EEG onset of habitual (e) and un-habitual (f) seizures (thick arrows).

gesting ulegyria secondary to hypoxic-ischemia encephalopathy (HIE) impairment, prominently on the right side (Fig. 1a). The fluorodeoxyglucose positron emission tomography imaging (FDG-PET) fusioned with MRI revealed hypo-metabolism in the bilateral parietooccipital ulegyria and the surrounding cortex, especially on the right side (Fig. 1b). Scalp Video Electroencephalography (s-VEEG) was performed for 22.5 h without antiepileptic drugs. With interictal EEG two forms of discharges were observed: one was frequent sharp waves with high amplitude in the bilateral parietooccipital region, prominently on the right side (Fig. 1c); the other form was high frequency, stereotyped, and biphasic sharp waves with low amplitude slow wave in the bilateral posterior quadrant region, prominently on the left side. And the second form of discharge appeared to have dipolar distribution with maximal negativity at the parietal leads and positivity at the frontal leads (Fig. 1d). The two kinds of discharges occurred independently. Eleven clinical seizures, including 9 habitual seizures were recorded. EEG data of habitual seizures showed low-amplitude fast waves in bilateral parietooccipital and posterior temporal region, predominantly on the right side (Fig. 1e). For the other two un-habitual seizures that are characterized by nausea, vomiting and without the loss of consciousness, the ictal EEG showed low-amplitude fast waves starting from the left centroparietal region (Fig. 1f). Additionally, dozens of subclinical discharges were recorded, displaying the same onset pattern as habitual seizures.

Taking together, according to the history of perinatal hypoxia, manifestations of imaging, s-VEEG, semiology of habitual and un-habitual seizures, the patient was diagnosed as lesional epilepsy secondary to the parietooccipital ulegyria coexisting with BCFE.

Epileptogenic zone of intractable lesional epilepsy was supposed to be at the right posterior quandrant cortex secondary to the parietooccipital ulegyria. Operation was performed under electrocorticography (ECoG) monitoring. The ECoG revealed frequent sharp waves at the right parietooccipital ulegyria and the surrounding cortex. Subclinical discharges were recorded twice, which were characterized by fast activities starting from the frontal edge of parietooccipital ulegyria. Firstly, disconnection of right parietooccipital lobe was done. Since there were still frequent sharp waves at frontal edge of disconnection boundary, tailored resection of temporooccipitoparietal junction cortex was then performed (Fig. 2a & b). In addition, relatively independent bipolar sharp waves were observed in the central cortex, which were supposed to be BFEDCs.

During the following 2 years, with OXC 0.6 g/d monotherapy, 3 times of seizures characterized by the right facial twitching and salivation, without loss of consciousness, occurred during sleeping. Scalp EEG of 22 months postoperatively showed reduced frequency of BFEDCs in the bilateral parietooccipital region (Fig. 2c). According to the typical nocturnal seizure semiology and feature of interictal EEG, we assumed these seizures as breakthrough seizures of BCFE.

3. Discussion

In this case, the BFEDCs were distributed in the bilateral parietooccipital region. Although the typical BFEDCs distributed mainly in the Rolandic area or occipital region, they were also occasionally observed in the parietal occipital area.This kind of discharges could be transferred to the Rolandic region, even to the frontal region with the maturation of the brain [2]. The BFEDCs recorded by intracranial EEG have rarely been reported. Here, for the first time, we recorded typical BFEDCs in the central cortex by intra-operative ECoG, relatively independent to the frequent sharp waves at surrounding cortex of parietooccipital ulegyria. Symptomatic partial