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Case Report

Surgical resection of epileptogenic cortical dysplasia in precentral gyrus ☆

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ABSTRACT

We present the case of a patient with epilepsy arising from cortical dysplasia in his precentral gyrus. The lesion was resected based on the results of video-ECoG (electrocorticograph) monitoring and electrical cortical stimulation. The patient has been seizure-free for 1 year since operation, and no neurological deficits have been observed. We discuss possible mechanisms of cortical reorganization in this patient and the features of focal cortical dysplasia (FCD) IIb in eloquent cortices.

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1. Introduction

Medically intractable seizures arising from sensory-motor cortices present a formidable challenge. The biggest risk for surgical therapy is postoperative sensorimotor deficits, which may be partially recovered in several months or years. In recent decades, electrical cortical stimulation has become the "gold standard" for functional mapping of pericentral areas. Our surgical epilepsy group reports a child suffering from intractable epilepsy with a lesion located at his precentral gyrus. After a complicated presurgical evaluation including video-ECoG monitoring and cortical mapping using electrical cortical stimulation (ECS), the patient's lesion was removed without any neurological complications observed after surgery.

2. Case report

The patient was a 7-year-old boy with intractable epilepsy since 2 months after birth. Despite the antiepileptic medications oxcarbazepine: 0.15 g, Q12h and lamotrigine: 37.5 mg, Q12h, the patient suffered from seizures as frequent as 8–10 times per day. No developmental delay was found with uneventful birth history. On physical examination, no abnormal findings were noted. MRI showed abnormal signal in the left anterior central gyrus, possibly a focal cortical dysplasia (FCD) (Fig. 1). Interictal EEG demonstrated spikes and slow waves in the left hemisphere (Fig. 2). Semiological studies showed seizures presenting as paroxysmal clonus of the patient's right hand and arm followed by secondary spreading to both sides.

We first performed intracranial electrode implantation for video-ECoG monitoring (Fig. 3). Abnormal spike activities and slow wave rhythms were localized at the cortices surrounding the lesion in the patient's left precentral gyrus (Fig. 4). After a 7-day recovery, ECS was conducted with the parameters including pulse width of 0.2 ms, frequency of 50 Hz, and intensity of 1 mA–15 mA, which demonstrated no function in the lesion area; the patient's right hand movement was functionally represented in the postcentral gyrus (Fig. 5).

Based on the results above, we resected the lesion in the precentral gyrus (Fig. 6), and abnormal cortical activities disappeared on the intraoperative ECoG. After surgery, the patient exhibited temporary weakness of the right upper limb which was resolved in 7 days. Antiepileptic medication remained the same as before surgery. Pathological findings were identified as FCD IIb. The patient has been followed up for 1 year, and he has experienced no more seizures since surgery.

3. Discussion

Focal cortical dysplasia (FCD) could be highly epileptogenic and frequently accompanied by medically intractable seizures. About 80% of patients with cortical dysplasia are seizure-free after epilepsy surgery, with much higher rates of becoming seizure-free with complete (80%) compared with incomplete (20%) resections [1]. However, for FCD in sensorimotor cortices, the likelihood of seizure-freedom after surgery is much lower because of incomplete resections due to the concern of postoperative neurological deficits, which is about 50–60% [2,3]. As the lesion was just located in the precentral area for this patient, we

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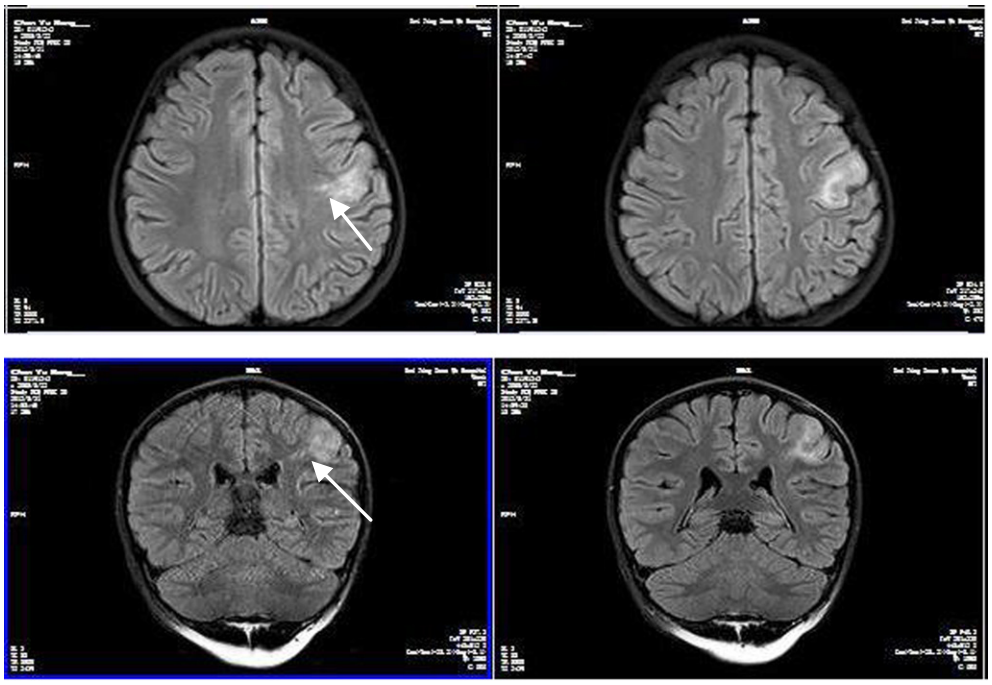

Fig. 1. T1, T2, FLAIR MRI for this patient. The lesion was characterized by high signal intensity, located in precentral gyrus. The white matter signal alteration tapers toward the ventricle, reflecting the involvement of radial glial–neuronal units (white arrow), which is called as "transmantle sign", first described by Barkovich in 1997, and almost exclusively found in FCD IIb [24].

were most concerned about whether the lesion was epileptogenic and the findings of functional cortical mapping.

Extensive abnormal spikes were seen in our case in the left hemisphere on EEG. Intracranial ECoG showed that interictal abnormal discharges were focused, surrounding the lesion, in the precentral area (red spot in Fig. 5), consistent with Honda and Marusic's reports about abnormal discharges for FCD [4,5]. The key point of discussion is focused mainly on what role balloon cells play, which are the hallmark for FCD IIb [6]. Marusic suggested that epileptogenicity

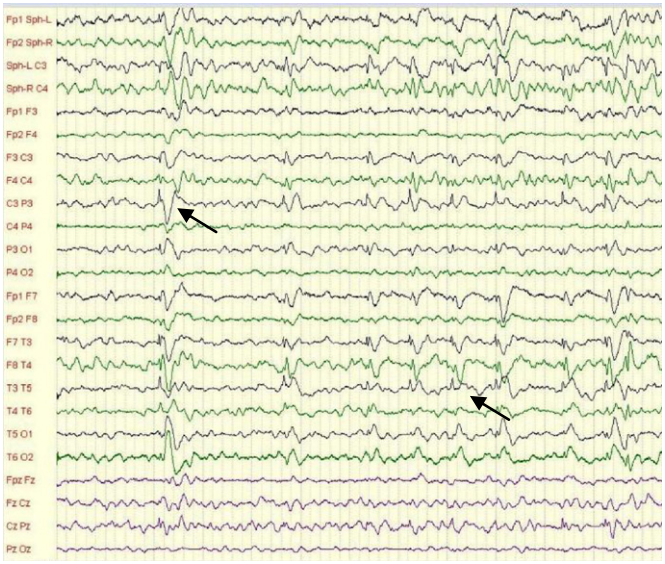

Fig. 2. Interictal epileptic discharges in this case. Spikes and slow waves were seen in the left hemisphere (black arrow). No abnormal rhythms were seen before the seizure.

was mainly identified in the immediately adjacent cortical areas [5]. Sarkis reported 3 cases of FCD IIb in the perirolandic area with drug-refractory epilepsy or drug-resistant epilepsy in which seizures were aggravated after incomplete resection, suggesting a possible inhibitory role of balloon cells in the setting of FCD IIb [7]. The possibility that FCD's have "intrinsic epileptogenicity" is supported by the demonstration of extremely active epileptiform discharges by steroelectroencephalography [8]. In our case, the patient became seizure-free for 1 year after the resection of his lesion with clear boundary leaving adjacent cortices intact. This supports the intrinsic epileptogenicity of FCD IIb in the precentral gyrus. The proposals from Marusic and Sarkis may need further evaluation by more cases.

It is controversial whether FCD in precentral gyrus has a functional role in movement. Chassoux considered that FCD in the precentral area participated in normal function [9]. However, Marusic and Janszky disagreed with this hypothesis [5,10]. Their results suggested that dysplastic lesions that are characterized by the presence of balloon cells did not show evidence of electrically inducible functions despite their location in anatomical areas such as precentral gyrus, just as in our case.

Besides ECS, fMRI and TMS are also used to identify cortical functional states [11,12]. In other studies of rolandic epilepsy surgery, neurological deficits recovered in 2 weeks to 6 months [2,7,13–15]. In our case, before resective operation, we performed ECS, suggesting negative results, and there were no obvious movement deficits after the resection of the lesion. Here, we propose the possibility of "cortical reorganization" or "cortical plasticity", which is a key process for a patient's recovery from stroke or perinatal lesion. Early research has found that FCD with balloon cells is a neuronal proliferational disorder, whereas the FCD without balloon cells is a cortical organization disturbance [10,16]. That means FCD with balloon cells emerged in an earlier stage of cortical development, which provides a greater chance for cortical or fasciculus reorganization [17,18]. As our ECS suggests that the part of movement function was shifted anatomically to postcentral gyrus, it is hypothesized that in epilepsy with FCD IIb in the precentral area, functional shifts or cortical reorganization is

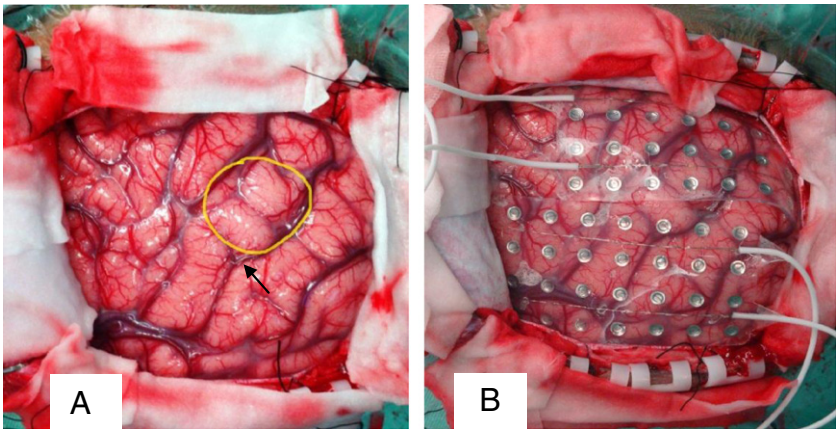

Fig. 3. A—lesion seen in operation. The lesion had a slightly different color and solid texture compared with normal cortices (yellow circle), vein was slightly pushed backward (black arrow). B—electrode grid implanted surrounding the lesion and covering the whole central area.

possible if the patient has no movement disabilities before surgery. After the lesion resection, there is a substantial possibility for seizure freedom without obvious deficits. Other work and case reports have supported our hypothesis [19–21].

The other possibility is the overlapping of functional states in the precentral gyrus or the so-called mechanism of compensation. The cortical areas which represent hand movement are larger than other areas [22]. Moreover, Teskey found that repeated seizure activities resulted in a doubling of the caudal forelimb motor area [23].

Curing rolandic epilepsy is considered possible without leaving neurologic deficits based on the experience in this case. However, it requires careful presurgical evaluation and the approach of wake surgery if conditions permit. The mechanisms need further studies.

Statement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines. We state that human studies have been approved by Xuanwu Hospital Ethics Committee and have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki. This work is original.

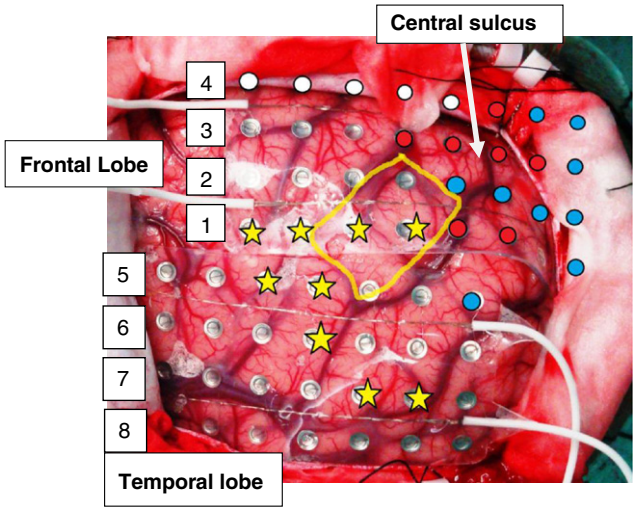

Fig. 5. Electrode implantation and result of ECS: No movement functions for the lesion area in the precentral gyrus (yellow circle). The cortex which represents hand movement was shown in red spot. Numbness was felt in the hand and arm when the cortex was electrically stimulated in the blue spot. According to Fig. 4, epileptic discharges were focused in the adjacent cortex surrounding the lesion with higher amplitude (yellow star).

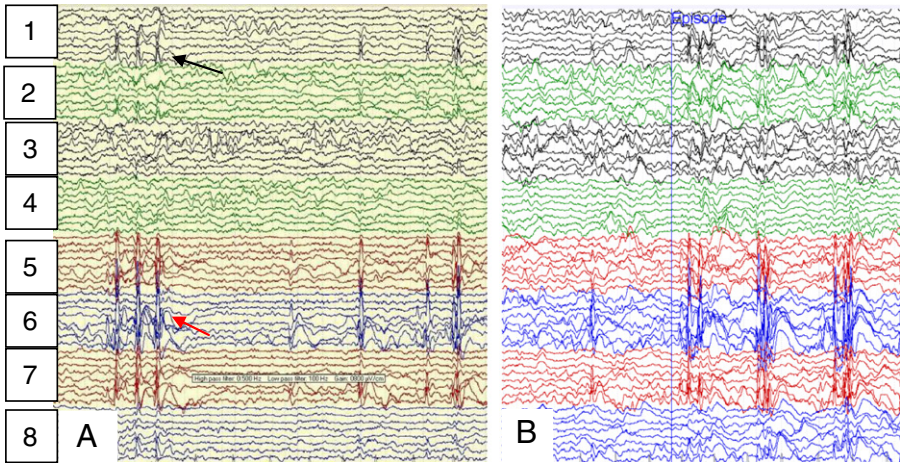

Fig. 4. A—interictal discharges from implanted intracranial ECoG. Some spikes were detected inside the lesion area (black arrow), but the amplitude of spikes was higher in the adjacent areas of the lesion (red arrow). B—no ripple fast rhythm or obvious discharges except isolated spikes before seizure, performed as left hand clonus (the upright blue line marked seizure beginning observed by video as right hand clonus).

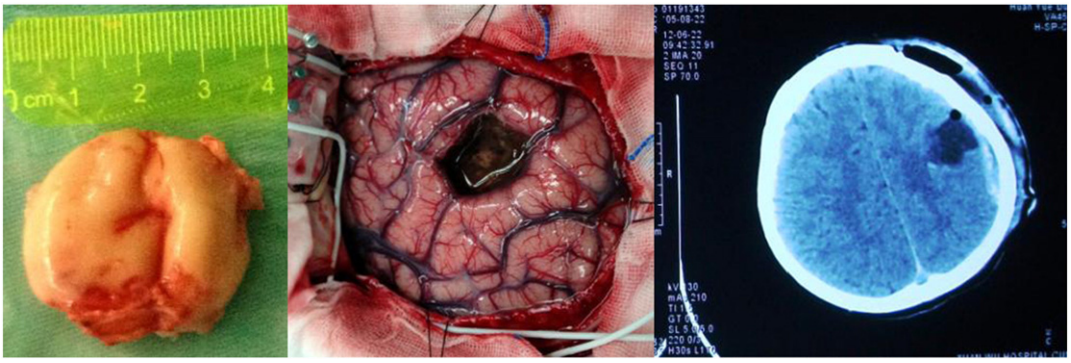

Fig. 6. Lesion resected in the second operation. The lesion is about 3.5 cm∗3.0 cm∗2 cm with solid texture.

References

[1] Hauptman JS, Mathern GW. Surgical treatment of epilepsy associated with cortical dysplasia: 2012 update. Epilepsia 2012;53:98–104.
[2] Behdad A, Bertrand ME, Smyth MD. Epilepsy surgery in children with seizures arising from the rolandic cortex. Epilepsia 2009;50:1450–61.
[3] Oliveira RS, Santos MV, Machado H. Tailored resections for intractable rolandic cortex epilepsy in children: a single-center experience with 48 consecutive cases. Childs Nerv Syst 2011;27:779–85.
[4] Honda R, Saito Y, Suzuki K. Focal cortical myoclonus in rolandic cortical dysplasia presenting as hemifacial twitching. Brain Dev 2012;34(10):886–90.
[5] Marusic P, Najm IM, Luders H. Focal cortical dysplasias in eloquent cortex: functional characteristics and correlation with MRI and histopathologic changes. Epilepsia 2002;43:27–32.
[6] Blumcke I, Thom M, Armstrong DD. The clinicopathologic spectrum of focal cortical dysplasias: a consensus classification proposed by an ad hoc Task Force of the ILAE Diagnostic Methods Commission. Epilepsia 2011;52:158–74.
[7] Sarkis RA, Bingaman WE, Najm IM. Surgical outcome following resection of rolandic focal cortical dysplasia. Epilepsy Res 2010;90:240–7.
[8] Palmini A. Electrophysiology of the focal cortical dysplasias. Epilepsia 2010;51:23–6.
[9] Chassoux F, Chodkiewicz JP, Duport DC. Stereoelectroencephalography in focal cortical dysplasia: a 3D approach to delineating the dysplastic cortex. Brain 2000;123:1733–51.
[10] Janszky J, Tuxhorn I, Woermann FG. Functional organization of the brain with malformations of cortical development. Ann Neurol 2003;53:759–67.
[11] Barba C, Montanaro D, Guerrini R. Focal cortical dysplasia type IIb in the rolandic cortex: functional reorganization after early surgery documented by passive task functional MRI. Epilepsia 2012;53(8):141–5.
[12] Vitikainen AM, Lioumis P, Gaily E. Functional plasticity of the motor cortical structures demonstrated by navigated TMS in two patients with epilepsy. Brain Stimul 2012:1–6.
[13] Mikuni N, Ikeda A. Yoneko Hayase. Hashimoto N Surgical resection of an epileptogenic cortical dysplasia in the deep foot sensorimotor area Epilepsy Behav 2005;7(3):559–62.
[14] Pondal-Sordo M, Girvin JP, Wiebe S. Epilepsy surgery involving the sensory-motor cortex. Brain 2006;129:3307–14.
[15] Lerner JT, Hauptman JS, Mathern GW. Assessment and surgical outcomes for mild type I and severe type II cortical dysplasia: a critical review and the UCLA experience. Epilepsia 2009;50:1310–35.
[16] Barkovich AJ, Kuzniecky RI, Jackson GD. Classification system for malformations of cortical development—update 2001. Neurology 2001;57:2168–78.
[17] Staudt M. Reorganization after pre- and perinatal brain lesions. J Anat 2010;217:469–74.
[18] Goradia D, Chugani HT, Sood S. Reorganization of the right arcuate fasciculus following left arcuate fasciculus resection in children with intractable epilepsy. J Child Neurol 2011;26:1246–51.
[19] Lado FA, Legatt AD, Lasala PA. Alteration of the cortical motor map in a patient with intractable focal seizures. J Neurol Neurosurg Psychiatry 2002;72:812–5.
[20] Cui ZQ, Luan GM. A venous malformation accompanying focal cortical dysplasia resulting in a reorganization of language-eloquent areas. J Clin Neurosci 2011:404–6.
[21] Staudt M, Gerloff C, Grodd W. Searching for motor functions in dysgenic cortex: a clinical transcranial magnetic stimulation and functional magnetic resonance imaging study. J Neurosurg 2004;101:69–77.
[22] Sanes JN, Donoghue JP, Warach S. Shared neural substrates controlling hand movements in human motor cortex. Science 1995;268:1775–7.
[23] Teskey GC, Monfils MH, Vandenberg PM. Motor map expansion following repeated cortical and limbic seizures is related to synaptic potentiation. Cereb Cortex 2002;12:98–105.
[24] Barkovich AJ, Kuzniecky RI, Grant PE. Focal transmantle dysplasia: a specific malformation of cortical development. Neurology 1997;49:1148–52.