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SHORT COMMUNICATION

# Intrinsic epileptogenicity of gangliogliomas may be independent from co-occurring focal cortical dysplasia

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**Summary** Gangliogliomas are a frequent cause of drug-resistant epilepsies in children. It remains unknown, however, whether gangliogliomas are intrinsically epileptogenic or if associated lesions contribute to their high epileptogenicity, i.e. associated focal cortical dysplasia (FCD). We report on a child operated twice for drug-resistant focal seizures symptomatic of a right temporal lobe lesion. Histological examination of the first, incomplete lesionectomy revealed tumor-associated FCD Type IIIb. The child was not seizure-free, and surface as well as intracerebral recordings were obtained during a second presurgical assessment. Histopathological examination of the second operation revealed a ganglioglioma. Intralesional EEG recordings from the ganglioglioma documented rhythmic bursts of fast activity suggesting that the high epileptogenicity of gangliogliomas is related to intrinsic epileptogenic activity.
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## Introduction

Gangliogliomas are the most frequent tumor entity in young patients with intractable focal epilepsy (Blümcke and Wiestler, 2002). Pathophysiological mechanisms underlying epileptogenicity in gangliogliomas have not been clarified yet. According to one hypothesis, the neuronal component of the tumor itself might contribute to epileptic activity. The expression of neuroactive molecules such as glutamatergic neurotransmitter receptors by dysplastic neurons (Aronica et al., 2001) supports the possibility of a hyperexcitable neuronal tumor component functionally integrated into excitatory circuitries (Blümcke and Wiestler, 2002). Another possibility is that tumor-associated epileptogenic changes in the adjacent brain might be primarily involved in seizure onset, as suggested by the observation that the epileptogenic zone may be larger than the tumor mass (Elger, 2002). Differentially expressed neurotransmitter receptors can be frequently encountered in perilesional brain tissue, further indicating its potential epileptogenicity (Wolf et al., 1996).

While specific electrographic features have been widely described in focal cortical dysplasia (FCD) using scalp and intracranial recordings (Chassoux et al., 2000; Boonyapisit et al., 2003; Widdess-Walsh et al., 2006), only few studies have characterized the EEG correlates of glioneuronal tumors. Ferrier et al. (2006) reported that continuous spiking was the only pattern to be seen significantly more often during acute electrocorticography (ECoG) in FCD versus gangliogliomas, while bursts and recruiting discharges were observed in either lesion. Continuous ECoG epileptiform discharges were recorded by Palmini et al. (1995) in 67% of patients with FCD versus only 2.5% of those with other epileptogenic lesions, mainly tumors. Kirschstein et al. (2003) published a series of eight patients, in which event-related potentials were directly recorded from focal lesions including glioneuronal tumors. Detection of larger intralesional amplitudes were compatible with the hypothesis that cortical lesions interact with synaptic pathways related to cognitive functions.

We describe a child with a right temporal ganglioglioma chronically explored by intracranial electrodes and provide new insights into the pathophysiological mechanisms underlying epileptogenicity in gangliogliomas.

## Case history

We observed a 3 year old girl, who had uneventful family and personal history. The child developed normally until the age of 6 months when weekly seizures appeared, characterized by motionless staring with perioral cyanosis. At the age of 17 months, spasms also appeared. Brain MRI showed a right temporal lesion that was suspected as tumor (Fig. 1A). Complete lesionectomy was planned but the occurrence of a subdural hematoma during the operation prevented the neurosurgeons from completing the procedure (Fig. 1B). Histology revealed FCD, which should be classified according to the new ILAE classification system as Type IIIb (Blümcke et al., 2010b). The child remained seizure free for 8 months; then two types of seizures appeared, having daily frequency (a) spasms on awakening and (b) staring with arrest and rightward head version. The interictal EEG showed subcontinuous right posterior spikes and spike and wave complexes, sometimes preceded by brief low voltage fast discharges. The seizures originated from the right posterior region with rapid contralateral spread. Neuropsychology revealed a normal cognitive level with mildly impaired language skills and attention span. Repeat MRI demonstrated no modification of the remnant lesion.

To better define the extent of the resection, we performed invasive recordings using two subdural grids exploring the right temporo–parieto–occipital (T0 in Fig. 2a and b) and fronto-anterior temporal (L in Fig. 2a and b) regions and by a depth temporal electrode implanted in between (T in Fig. 2a and b).

At the depth electrode and on the contacts of the grid ''L'', exploring the temporal lobe (i.e. L1–4 and L9–12), we recorded very low voltage background activity with subcontinous rhythmic bursts of low voltage fast activity intermingled with spikes. Brief discharges of fast activity were observed, asynchronously, on the most anterior line of the ''T0'' grid (contacts T034–38). The remaining ''T0'' lines showed subcontinuous slow activity, intermingled with spikes on the inferior parietal leads (T07–8 and T015–16) and on the temporal contacts T012–14; T018–21; T026–29 (Fig. 2). The ictal EEG during spasms showed very fast low voltage discharges on the depth electrode and on the same ''L'' contacts capturing the rhythmic bursts, spreading towards the temporal T0 34–38 leads and, less frequently, towards the basal region of the temporal lobe (i.e. T02–3, T010–11, T018–19 and T026–27). The ictal onset zone was considered to involve the temporal lobe from the area immediately posterior to the border of the first surgery to the most anterior line of the ''T0'' grid, while the irritative zone, defined by the presence of both the subcontinous rhythmic bursts of low voltage fast activity and the slow waves intermingled with spikes, extended towards the temporo-occipital junction and the inferior parietal region (Fig. 2a and b). Surgery was tailored according to anatomical and neurophysiological data (Figs. 2a and 3): the area of resection included the temporal lobe towards the temporo–occipital junction i.e. the epileptogenic zone and part of the irritative zone, sparing the inferior parietal lobe whose ictal involvement was inconsistent. Second surgical procedure was successful. The child is on carbamazepine and seizure free 2 years after surgery.

## Histopathological review

All paraffin-embedded tissue blocks were submitted to the Department of Neuropathology, University Hospital Erlangen, Germany for histopathological review. Sections were cut at 4 μm with a microtome (Microm, Heidelberg; Germany), stretched in water at 40 °C and mounted on slides coated with silane (Langenbrinck; Emmendingen, Germany). The slides were air-dried in an incubator at 37 °C overnight, deparaffinised in descending alcohol concentration and stained with hematoxylin and eosin (H&E) as well as cresyl violet. Immunohistochemical reactions were performed as previously described (Blümcke et al., 2010a) using the following antibodies: CD34 (1:100, Dako), GFAP (1:800, Dako), MAP2C (clone C; 1:100, supplied by Dr. B. Riederer, Lausanne, Switzerland), NeuN (1:1000, Millipore),

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