Epileptogenic ganglioglioma with hypermetabolism on positron emission tomography with fluorine-18 fluorodeoxyglucose: a case report

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Summary

Positron emission tomography with fluorine-18 fluorodeoxyglucose ([18F]FDG-PET) usually shows ganglioglioma as hypometabolic, in the absence of malignant transformation or high grade. We report a case of benign ganglioglioma in the left precentral gyrus showing hypermetabolism on [18F]FDG-PET, which was associated with intractable focal motor seizures. An 11-year-old girl started having simple partial seizures refractory to anti-epileptogenic medication since 4 years of age. Magnetic resonance imaging revealed a tumorous lesion in the left precentral gyrus. Interictal [18F]FDG-PET showed high [18F]FDG uptake in the lesion. Gross total resection was carried out. The histological diagnosis was low-grade ganglioglioma. The hypermetabolism on [18F]FDG-PET may be associated with epileptogenic activity of the ganglioglioma.
Introduction

Ganglioglioma is rare, with an incidence of only 1.3% in large brain tumor series, but is the most common tumor entity in young patients suffering from chronic focal epilepsies [1, 2]. Most gangliogliomas occur in the temporal lobe, with only 8.2% to 12% in the frontal lobe [2] [3]. Ganglioglioma usually grows slowly, therefore few patients present with focal neurological deficit or evidence of increased intracranial pressure, whereas most patients have a long history of seizure disorder [3]. Ganglioglioma consists of large dysplastic neurons and neoplastic glial cells. Both cell populations may exhibit marked heterogeneity, with the morphologic spectrum of gangliogliomas ranging from predominantly neuronal to predominantly glial phenotype [1]. Malignant features are rare, and most gangliogliomas are low-grade tumors (World Health Organization [WHO] grade 1). Positron emission tomography with fluorine-18 fluorodeoxyglucose ([18F]FDG-PET) measures [18F]FDG uptake, which corresponds to the glucose metabolism and is related to malignancy. Most gangliogliomas show low uptake of [18F]FDG, in the absence of malignant transformation or high-grade ganglioglioma [3, 4].

We treated a girl with intractable partial seizures caused by a benign ganglioglioma which showed high uptake on [18F]FDG-PET.

Case report

An 11-year-old girl started to have right focal motor seizures involving the arm and hand since 4 years of age. Although she was treated with anti-epileptic agents immediately after onset, her seizures increased in frequency every year. Eventually she suffered from seizures every day and her school performance continued to decline. Magnetic resonance (MR) imaging revealed a tumorous lesion in the left precentral gyrus at age 10 years. She was admitted to our hospital for surgical treatment at age 11 years.

On admission, neurological examination revealed right hand impediment which had caused her to change her dominant hand to the left. The simple partial seizures occurred two or three times per day, starting from her right hand without impaired consciousness, and continued for 7-8 seconds. MR imaging revealed a single mass 2 cm in diameter in the left precentral gyrus, which was not enhanced with contrast medium (Fig. 1A, B). Interictal [18F]FDG-PET showed high [18F]FDG uptake in the lesion (Fig. 1C, D). Scalp electroencephalography (EEG) in the interictal period showed spikes and waves in the central and parietal areas of the left hemisphere (Fig. 2A). Although ictal EEG recording with video monitoring was tried, she could not endure being in a closed place for a long time. Based on these examinations, we assumed that the lesion in the precentral gyrus was the cause of her intractable seizures and planned to perform removal of the lesion.

A left frontoparietal craniotomy was performed under general anesthesia with nitrous oxide and propofol. The central sulcus was identified by the recording of phase reversal of somatosensory evoked potentials. The lesion was located in the precentral gyrus, and pro-
truded rostrally (Fig. 2B). Intraoperative electrocorticography (ECoG) showed epileptiform discharges on the protruded cortex and surroundings (Fig. 2C). Although we could not detect phase reversal of the paroxysm with bipolar recording, we surmised that the focus underlay the protruded cortex and spread the discharge to the cortex around the lesion. Gross total resection of the lesion as well as the overlying cortex was conducted under monitoring of the motor evoked potentials by monopolar cortical stimulation. There was no significant motor response following stimulation of the cortex overlying the tumor. Histological examination found a biphasic morphologic pattern consisting of dysplastic neurons and glial cells, with a tendency of a richer neuronal component. The histological diagnosis was ganglioglioma (WHO grade 1). No cortical dysplasia and no gliosis were ob-

Fig. 1: Axial T1-weighted MR image with contrast medium (A) and sagittal T2-weighted MR image (B) showing a 2-cm diameter mass in the left precentral gyrus. [18F]FDG PET scans (C, D) showing localized hypermetabolism corresponding to the lesion.
served in the cortex around the tumor.

After the operation, she showed improved right hand impediment and no new neurological deficits. She remained completely seizure-free for 2 years after the surgical treatment, with the use of the same doses of anti-epileptic agents as given preoperatively, but the medication was tapered gradually and then discontinued. She has remained seizure-free for 1 year after discontinuation.

Fig. 2 A: Preoperative interictal EEG demonstrating interictal spikes recorded at the left central and parietal electrodes. B: Intraoperative photograph showing the position of the ECoG electrodes at the left rolandic region. The black dotted line shows the inferred location of the tumor and the yellow dotted line represents the electrophysiologically determined central sulcus. Numbers of the electrodes correspond to the ECoG traces in Fig. 2C. C: Intraoperative ECoG traces showing epileptiform discharges recorded from the cortex overlying the tumor and the caudally adjacent cortical area.
Discussion

[18F]FDG-PET is very useful for the noninvasive monitoring of cerebral glucose metabolism and the evaluation of brain tumors. Glucose metabolism of gangliogliomas on [18F]FDG-PET has been described in several reports. Preoperative examination of 9 patients with ganglioglioma showed hypometabolism or isometabolism [3]. Thirty-four cases, almost all low-grade gangliogliomas, showed hypometabolism or isometabolism, except one tumor with hypermetabolic activity [5]. Another 13-year-old boy with a ganglioglioma within the left temporal lobe had hypermetabolic activity of the tumor [4]. In a recent study, prominent hypermetabolism on [18F]FDG-PET was noted in two of five cases of ganglioglioma, although the biologic features were benign and indolent [6]. Our findings are similar to these exceptional cases. They suggest that glioneuronal tumors with hypermetabolism may have intrinsic epileptogenic properties.

[18F]FDG-PET has also been used to detect seizure foci and evaluate surgical resection for the treatment of localization-related epilepsies [7]. Epileptogenic foci are hypometabolic in the interictal period but the glucose consumption increases sharply accompanying epileptic discharge during the ictal period. Such changes in glucose metabolism persist for 48 hours after the seizures [8]. Our patient had seizures two or three times a day, therefore the [18F]FDG-PET findings can be interpreted as extended hypermetabolism in the acute postictal period.

The pathogenic mechanisms underlying focal hyperexcitability of gangliogliomas have not yet been determined. Two major hypotheses can be considered. The neuronal component of the tumor may contribute to the epileptic activity, or tumor-associated epileptogenic changes may be present in the adjacent brain tissue [1] [9]. In this case, preoperative [18F]FDG-PET showed hypermetabolism within the tumor and intraoperative ECoG recorded interictal epileptiform discharges from the tumor and surroundings. Lesionectomy including the overlying cortex achieved total control of seizures without the need for medication. These observations might suggest that the tumor had epileptogenic activity.

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References


