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Putaminal hyperperfusion in dystonic posturing on subtracted SPECT: A case report

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Dystonic posturing (DP) is one of the most reliable lateralizing symptoms for mesial temporal lobe epilepsy, although the mechanism remains unclear. We demonstrated a hyperperfusion area in the right putamen on subtracted postictal SPECT by using the automatic registration technique in one patient with ictal DP of the left hand. The putamen may play a key role in DP, similar to other diseases with dystonia.

Key words: dystonic posturing, temporal lobe epilepsy, subtracted SPECT, putaminal hyperperfusion

INTRODUCTION

DYSTONIC POSTURING (DP) is one of the ictal symptoms of mesial temporal lobe epilepsy (MTLE). It is characterized by sustained peculiar posturing of one upper extremity with a rotational component. DP is recognized as the most reliable symptom to determine lateralization for MTLE.[1,2] The basal ganglia or supplementary motor area, or both, were thought to play a key role in this phenomenon. Newton et al.[3] described a hyperperfusion area of the basal ganglia on ictal SPECT in patients who manifested DP. But the exact location remains unknown.

Subtraction of ictal from interictal SPECT co-registered with the MRI technique has been performed recently in localization-related epilepsy, and it is possible to clearly demonstrate the epileptogenic focus on MRI.[4–8] Semi-automatic systems were usually available for these studies, but fully automatic registration relied on anatomical correlation, and few clinical studies were reported. In this paper we report on one patient with ictal DP correlated with a hyperperfusion area on fusion images of MRI and subtracted postictal SPECT, by using the automatic registration technique.

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CASE REPORT

A 36-year-old-man was admitted to our hospital for epilepsy surgery. He had had medically intractable complex partial seizures for more than sixteen years. The SPECT study was done during the pre-surgical evaluation. Bilateral temporal epileptic discharges were recorded under routine interictal EEG. Long-term video-EEG monitoring (BMSI 5000, Nicolet, Madison, WI) was performed for ictal symptoms and ictal EEG. The ictal symptoms of the seizure began with head deviation to the right side, followed by DP of the left hand, automatisms of the mouth, arms and legs, with salivation during a confused state. DP started twenty seconds after ictal EEG onset. Ictal EEG showed rhythmic 6 Hz theta waves predominantly over the right temporal region and on the sphenoidal lead during complex partial seizures.

For the postictal SPECT, 750 MBq (20 mCi) Technetium-99m-ethyl cysteinate dimmer (^{99m}Tc-ECD) was injected immediately after the ictal symptoms disappeared. SPECT images were acquired two hours after the injection with a high-resolution three-head gamma camera (GCA9300, Toshiba, Tokyo, Japan). The data were acquired in a 128 × 128 matrix with 64 views. Postictal SPECT showed hyperperfusion areas in the right temporal lobe and basal ganglia. Interictal SPECT was performed one month later, and the SPECT revealed hypoperfusion areas in the bilateral temporal lobes. MRI was performed throughout the brain with 2 mm thin axial slices (T1-weighted image, 1.0 T Siemens Magnetom,

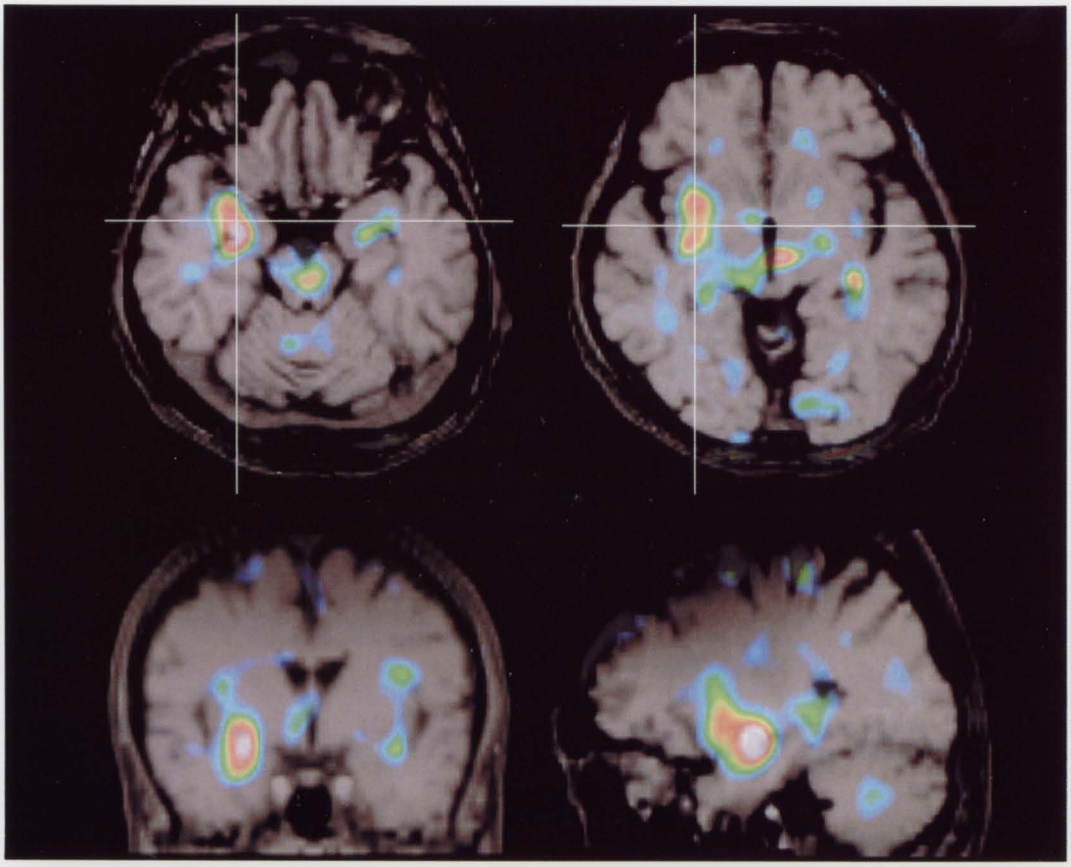

Fig. 1 Subtracted postictal-interictal SPECT. Hyperperfusion areas are demonstrated in the right amygdala, hippocampus, insula and ventral putamen. Upper images indicated axial views, and left side of the lower image indicated coronal view, and right side of the lower image indicated sagittal view.

Erlangen, Germany), and it showed right hippocampal atrophy. Each SPECT image was adequately registered to MRI with established software (Automatic Registration Tool (ART), Toshiba, Tokyo, Japan) according with Ardekani's method.[9] Then, the subtracted postictal SPECT was then calculated. The fusion images of MRI and subtracted postictal SPECT showed an increase in regional blood flow in the right side of the hippocampus, amygdala, insula and ventral putamen (Fig. 1). After presurgical evaluation, a right anterior temporal lobectomy was done. Hippocampal sclerosis was found in the operated specimen. He has been free of seizures for one year.

DISCUSSION

DP is useful in determining lateralization in cases of MTLE, but the exact location inducing ictal DP was unknown. It was presumably caused by activated basal ganglia or a supplementary motor area. Newton et al.[3] compared ictal SPECT with and without DP, and reported that the basal ganglia played an important role in this phenomenon, although it was unclear which nucleus was most important in the symptom. We demonstrated a remarkable increase in the putamen within the basal ganglia. This finding strongly supports the theory that abnormal discharges in the unilateral putamen are responsible for DP of the contralateral side. The putamen appeared to play a very important role in DP in patients with MTLE, similar to other diseases with dystonia.[10,11]

Head deviation to the ipsilateral side of the focus was found on electrical stimulation of the hippocampus, and salivation also followed stimulation of the deep fissure of Sylvians.[12,13] In our patient epileptic discharges started from the right mesial temporal structure, and presumably spread to the insula and putamen on the same side. The latter activations are thought to cause autonomic symptoms and unilateral DP, respectively. Hyperperfusion areas on the SPECT images in this case are in reasonable accord with the symptoms.

Timing of the tracer injection is controversial. At the beginning of a seizure, early injection of the tracer can detect the initial onset zone,[6] but postictal SPECT with an automatic registration technique succeeded in detecting the epileptic foci.[8,14] Epileptic discharges propagated to the symptomatic zone, and caused the ictal symptoms. The regional blood flow, associated with ictal symptoms,

often continues to increase even in the early postictal state.

Ichise et al.[15] reported that relative distribution of ^{99m}Tc-ECD in the basal ganglia remained stable for 120 minutes. In this case, the SPECT images were acquired within this limit, so that the hyperperfusion in the putamen is thought to reflect a true change in focal blood flow. The fact that the increase was observed only in the right putamen further supports a symptom related change, although ^{99m}Tc-ECD SPECT is generally scanned within one hour.

Further study with this method will be needed to determine the mechanisms of ictal seminology.

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