Painful Focal Sensory Seizure Arising from the Primary Somatosensory Cortex

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Abstract

A 31-year-old, right-handed woman had frequent focal painful seizures involving the right hand without any movement. EEG demonstrated an ictal activity arising from the left centroparietal region. No cerebral structural abnormality was seen on MRI. Ictal single photon emission CT showed markedly increased activity in the left perirolandic cortex, which remained active following the ictal symptoms when the EEG seizure pattern had completely disappeared. It is concluded that the painful seizures in the present patient originated from the primary somatosensory cortex. The prolonged increase of regional blood flow in the perirolandic area may reflect the possibility of persistent subclinical epileptogenicity. (Internal Medicine 42: 875-879, 2003)

Key words: sensory seizure, primary sensory cortex, EEG, SPECT, epileptogenicity

Introduction

Seizures manifesting pure sensory symptoms (sensory seizure) are rarely encountered in adults, while they are rather frequently seen in children (1, 2). Since sensory seizures have no objective ictal signs and since non-motor simple partial seizures are less accompanied by scalp-recorded EEG changes, not infrequently the subjective description of the ictal semiology is the important clue that can lead to proper clinical diagnosis and appropriate treatment. Especially when the patients with pure sensory seizures have accompanied psychiatric complaints, the chances for misdiagnosis or delayed diagnosis are high (3). Penfield and Jasper (4) attributed the possible origins of somatosensory seizures to the primary and second somatosensory cortices, and supplementary motor areas. Seizures arising from the postcentral gyrus, i.e., primary somatosensory area (SI), are less frequently seen as compared with seizures from temporal and frontal lobes (5). In addition, extratemporal lobe epilepsy unassociated with a structural lesion less frequently shows focal hypometabolism of fluoro-deoxyglucose in positron emission tomographic (PET) studies (5-7), as opposed to temporal lobe epilepsy (TLE) in which interictal temporal hypometabolism occurs in about 85% of cases.

We had an opportunity to study a patient who had focal somatosensory seizure without any motor symptoms during the attack, and we report the results of physiological evaluation of ictal and interictal brain activity by using electroencephalogram (EEG), single photon emission computed tomography (SPECT) and PET.

Case Report

A 31-year-old, right-handed Japanese woman was first admitted to a local hospital because of progressive severe fatigue with low grade fever and nasal discharge. She had been previously well. She had non-consanguineous, healthy parents, and her family history was negative for epilepsy or neurodegenerative disorders. Four days after admission, focal clonic convulsions of the right forearm associated with loss of consciousness occurred, followed by generalized tonic clonic seizure. The seizures were well controlled by intravenous administration of phenytoin sodium (PHT). There was no evidence of inflammation in the peripheral blood or cerebrospinal fluid, and cranial MRI with gadolinium DTPA showed no structural abnormalities of the brain.

Eight months later, following poor compliance of oral PHT for the previous several weeks, focal motor seizures...
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with secondary generalization appeared. It was well treated by intravenous PHT again, but she started complaining of attacks of shooting pain in the right upper extremity each lasting several minutes. Later it was accompanied by a continuous tingling sensation of the right hand. On admission to Kyoto University Hospital, she frequently complained of attacks of the pain in the right forearm which were associated with a puffy sensation. In addition, she had frequent intermittent auditory verbal hallucination, each lasting about 30 seconds. The auditory hallucination occurred independent of the pain. Consciousness was preserved throughout the attacks.

When admitted, the patient was well, cooperative and oriented, but slightly slow in responding to the examiner’s instructions. Neurological examination revealed severe sensory loss in all modalities in the right hand and forearm, and clumsiness in fine movements of the right fingers. Occasionally she noticed spontaneous bizarre sensation in the right hand and forearm. During the attack, she also felt as if her right hand changed in configuration. At times, the right forearm slowly moved like choreoathetosis, although no jerky or twitching movements were observed. The seizures were easily elicited by hyperventilation, and easily stopped within 20 seconds by rubbing the right hand with her own left hand. Muscle power was preserved without asymmetry, and deep tendon reflexes were normal.

Laboratory examinations including hematological, biochemical and serological tests were all within normal range. No inflammatory activity was found. Serological titer for herpes simplex virus in both blood and cerebrospinal fluid was repeatedly normal. Scalp EEG was recorded by using a digital EEG equipment with the sampling rate of 200 Hz,

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**Figure 1.** Interictal (A) and ictal (B–D) EEGs in the present patient. A: Interictal EEG showed bilateral irregular slow waves, with more in the left hemisphere, and occasional focal slow waves at the left fronto-central area. B: At the onset of focal sensory seizure (indicated by the arrow), the posterior dominant rhythm became poorly organized on the left, and low amplitude fast activity appeared at the left central area. C: Eighty seconds after the onset, the faster activity extended to the frontal and parietal region, and became larger in amplitude and slower in frequency. D: Two minutes after the onset, the paroxysmal activity suddenly disappeared (indicated by the arrow), and the background rhythm reappeared.
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high frequency filter of 60 Hz, and time constant of 0.3 second. Interictal EEG demonstrated a poorly organized background activity intermixed with irregular slow waves bilaterally more on the left hemisphere. There were occasional focal slow waves at the left fronto-central area (Fig. 1A). There were no definite interictal epileptiform discharges. At the beginning of the ictal phase, low amplitude quasi-rhythmic 20Hz fast activity occurred at the left central area, which became larger in amplitude and slower in frequency (Fig. 1B). It then spread to the bilateral frontal and parietal areas, much more on the left side (Fig. 1C). About 2 minutes afterwards, the ictal activity suddenly disappeared (Fig. 1D). Seven episodes with the same EEG findings occurred in 30 minutes, and four of them coincided with the abnormal sensation of the right hand.

Somatosensory evoked potentials (SEPs) were recorded after admission. The amplitude of P25 component, measured from the preceding N20 peak, at the centro-parietal area following median nerve electrical stimulation at the wrist was decreased on the left: -0.23 μV at CP3 to the right stimulation and -1.43 μV at CP4 to the left stimulation. However, there was no difference in latency between the two sides. After increasing the dosage of PHT and adding carbamazepine, both sensory attacks and auditory hallucinations became infrequent, and disappeared completely in one month after the second admission. Since then she has been free from those attacks.

Neither brain CT nor Gd-DTPA enhanced MRI obtained by 1.5T machine showed abnormality. Brain SPECT was performed to evaluate cerebral blood flow (CBF) on two occasions. The first study was done by intravenous injection of

Figure 2. 99mTc-ECD SPECT obtained during the focal sensory seizure and a follow-up study. Top (ictal): flow tracer was rapidly injected at the clinical seizure onset. A markedly increased regional uptake was seen in the whole left parietal area slightly extending to the central area. Bottom: 5 months later when the seizures completely disappeared, persistent high perfusion activity was still present in the left posterior parietal area.

Discussion

Seizures manifesting only paroxysmal local pain (sensory seizure, or painful seizure if painful) are not common (8, 9). In 1983, Young and Blume (9) reported 24 patients with painful seizures among their 858 epileptic patients and classified them into three groups: (i) unilateral group with unilateral pain in the face, arm, leg and trunk; (ii) cephalic group with pain restricted to the head; and (iii) abdominal group with central abdominal pain. They postulated that unilateral pain could be derived from the ictal activity in the contralateral rolandic area (primary somatosensory cortex, SI). The second somatosensory area (SII) is also conceivable as a source if the pain involves all four limbs with face spared (8, 10, 11). In the present patient, Gd-DTPA did not enhance any brain lesion at MRI, and cranial CT did not suggest any structural abnormality in the brain, but all functional studies (EEG, evoked potentials and SPECT) clearly indicated the abnormalities in the left central, superior temporal to parietal cortices. All these findings are consistent with the
clinical manifestations mainly consisting of somesthetic syndromes. Therefore, it is concluded that this patient most likely had sensory seizures arising from the left SI. The question as to whether pain perception is mainly processed in the hand area of SI or SII, or in both areas, remains to be solved (12, 13). The recent study by means of MEG and subdural recording for pain related evoked potentials suggested that SI is also actively involved in pain perception (14). The present study may support that SI is actively involved in pain perception. The etiology of the seizures in the present patient still remains unsolved especially as to whether it was inflammatory or not. In patients with focal seizure, brain SPECT and PET often reveal regional abnormalities, and the regional CBF in the focus increases ictally and decreases interictally (15–17). In the present patient, ictal SPECT revealed increased regional CBF broadly in the left centro-parietal to superior temporal area, and no significant changes in the frontal cortex or the right sensorimotor cortices (Fig. 2). The high perfusion in the left posterior parietal area was still detected by SPECT and PET 5 months later, though to a lesser degree as compared with the ictal state. The present finding of ictal SPECT is consistent with the previous report (18), but it is unsolved as to why the follow-up SPECT obtained 5 months later still showed focal high perfusion in spite of complete absence of seizures. There are two possible explanations for this finding. One is a persistent inflammatory process possibly existing in the posterior parietal area. However, it is rather unlikely because there was no definite evidence in the laboratory findings to suggest an inflammatory process including CSF. The other is that the left posterior parietal region could be epileptogenically active possibly due to a persistent cortical abnormality such as focal cortic dysplasia. Currently, localized neuronal migratory disorder is considered to be one of the most important causes of medically intractable partial epilepsy of neocortical origin (19–21). T2-weighted or fluid-attenuated inversion recovery (FLAIR) images often, but not always, identify these cortical abnormalities (19, 22). Kuzniecky and Powers (22) reported two patients with intractable partial seizure caused by focal cortic dysplasia, in whom focal increase of CBF in the ictal state was demonstrated by Tc-hexamethylpropyleneamineoxime (HMPAO) SPECT, but MRI did not demonstrate any cortical abnormality. Chugani (23) also showed two patients with intractable infantile spasms caused by focal cortical dysplasia, and FDG-PET demonstrated localized hypermetabolism in one of them, presumably like the present patient. In the present patient, the absence of interictal spikes may be explained by the small size of the epileptogenic focus. According to the description by Palmini et al (19), 10 out of 34 patients with cortical dysplasia did not show any ictal activity over the scalp, but it was observed only by direct cortical recording. Thus, it is conceivable that, in the present patient, the subclinical, congenital epileptogenic activity might involve the postrolandic gyrus. Follow-up SEP recording might have been helpful to compare with the findings of the blood flow study, but it was not undertaken due to the patient’s condition.

In conclusion, it was also suggested that partial sensory seizures, especially painful seizures, are rarely observed, but when it is clinically suspected, proper diagnostic approaches including EEG are essential.

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