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Case Report

Complex behavioral automatism arising from insular cortex

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Abstract

We describe two cases of complex partial seizures with ictal violent movements arising from the insular cortex. The first patient, a 14-year-old girl, presented with hyperkinetic behavior such as rolling, thrashing, and pedaling, and the second case, a 38-year-old woman, had been suffering from frequent daytime hyperkinetic seizures characterized by bizarre vocalization, jumping, and violent bimanual movements. Both patients showed a slight high signal change in the right posterior ventral insular cortex in fluid-attenuated inversion recovery (FLAIR) studies involving magnetic resonance imaging, and extensive subdural electroencephalographic monitoring revealed EEG seizure onset from the temporal lobe. The posterior ventral insular and lateral temporal cortices were resected, resulting in complete seizure freedom in both cases. The histological diagnoses were focal cortical dysplasia in the first case and gliosis in the second case. There may exist a group of patients with complex partial seizures with ictal violent automatism that can be ameliorated by the resection of epileptogenic lesions in the insular cortex. Careful inspection of the insular cortex is necessary to diagnose this type of epileptic seizure.

Keywords: Hyperkinetic seizure; Violent movement; Epilepsy surgery; Insular cortex; Temporal lobe

1. Introduction

The relationship between violence and epilepsy has been discussed as a social problem for a long time, at least since the 19th century [1]. Interictal violence has been reported as a possible result of an increased prevalence of violent behavior in patients with epilepsy [1]. Ictal violence is related to legal issues, such as diminished legal responsibility and insanity defenses [1–6], and this condition includes psychomotor seizures and motor automatism [1].

Complex behavioral automatism with ictal violent movements has been given various names including violent automatism [7], bimanual–bipedal automatism [8], and bicycling movement [9], based on the characteristics, although these have not been characterized with standard nomenclature.

The frontal lobe [8,10] is known as the origin of complex partial seizures with motor automatism, whereas the insula is a rare source for this [11].

We report here on two patients with complex behavior automatism with violent movements, diagnosed as seizures originating in the insula, that were completely eliminated by resection of the posterior ventral insular and lateral temporal cortices.

2. Case reports

2.1. Case 1

This right-handed female patient had a history of complex partial seizures associated with blinking of the right eye from the age of 2, and with falling attacks and tonic posturing from the age of 8. At 8 years of age, moreover, daily nocturnal seizures associated with hyperkinetic behaviors, such as rolling, thrashing, and pedaling, began

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to occur. During an attack, she did not bear on the subject of directed aggression. She was introduced to our institute at the age of 14.

Her seizure frequency had increased during the last few years despite treatment with multiple antiepileptic medications. She had no neurological deficits except for a moderate cognitive decline, and her Full Scale IQ (FIQ) score on the Wechsler Adult Intelligence Scale—Revised (WAIS-R) was 49. Her interictal mental condition was normal, and there were no psychiatric or personality changes.

A fluid-attenuated inversion recovery (FLAIR) study involving MRI showed a slight signal change in the right posterior ventral insular cortex (Fig. 1), and interictal 2-deoxy-2[^{18}F]fluoro-D-glucose (FDG) positron emission tomography (PET) revealed decreased glucose uptake by the bilateral anterior cingulate gyri and the right temporal lobe.

A scalp ictal EEG recording revealed bilateral diffuse desynchronization, and the right temporal lobe showed dipoles of epileptic spikes by magnetoencephalography (MEG). Also, extensive subdural EEG monitoring covering the medial, lateral, and basal aspects of the right frontotemporal cortices demonstrated EEG seizure onset from the basolateral aspect of the temporal lobe.

The posterior ventral insular and lateral temporal cortices were resected under intraoperative electrocorticography, with preservation of the medial temporal lobe structures. Subsequently, a histological diagnosis of focal cortical dysplasia was made. Since surgery, the patient has been seizure-free for more than 24 months as a high school student without significant neurological deficits.

2.2. Case 2

This right-handed female patient began to experience brief complex partial seizures associated with motionless staring for several seconds at 15 years of age. At 25, her seizures changed to motor seizures with violent behavioral automatism. The frequency of the seizures kept increasing despite her use of multiple antiepileptic medications. She was introduced to our institute for the treatment of frequent hyperkinetic seizures occurring in the daytime with a clustering tendency at the age of 38.

She had no neurological deficits except for a mild cognitive decline; her FIQ score on the WAIS-R was 71. Her seizures started with abrupt bimanual to-and-fro movement without aura, followed by pelvic thrashing, pedaling, and

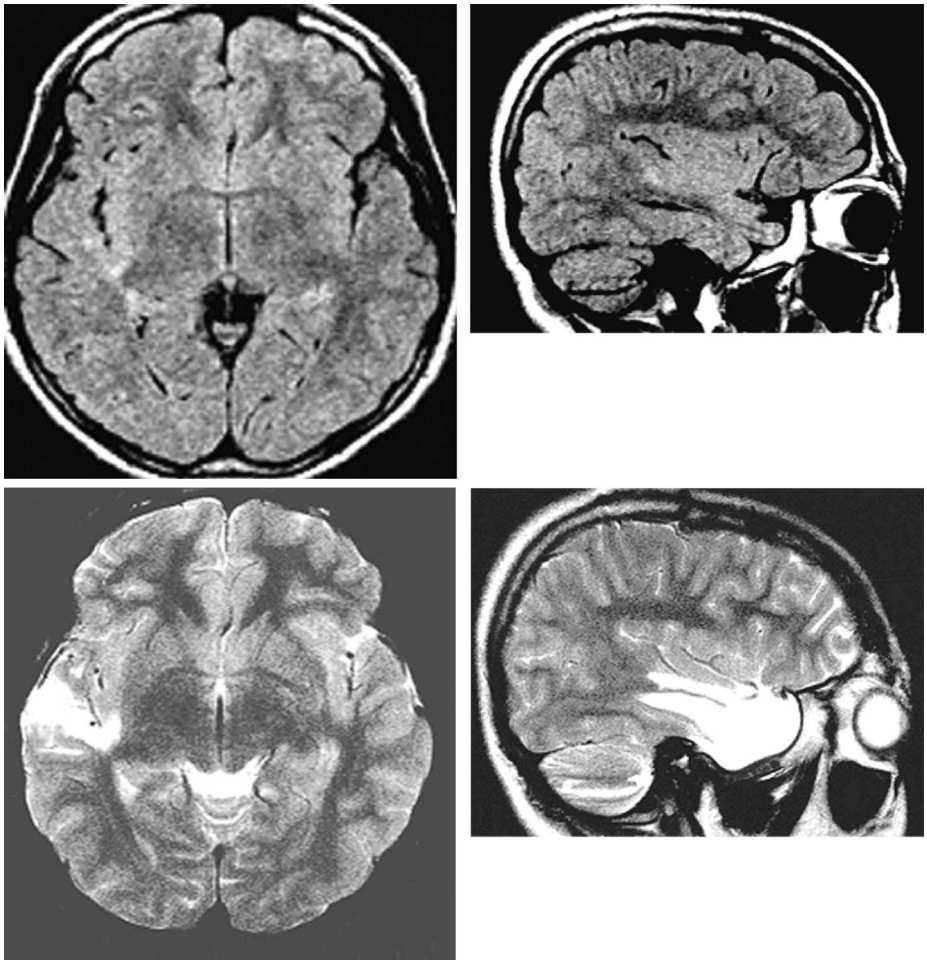

Fig. 1. Axial (left) and sagittal (right), FLAIR (top), and T_2-weighted (bottom) magnetic resonance images of case 1. Top: preoperative magnetic resonance images revealing a signal change in high intensity in the right posterior ventral insular cortex. Bottom: postoperative magnetic resonance images obtained showing resection of the posterior ventral insular and lateral temporal cortices.