A case of unilateral dystonia appeared ten years after a blow to the head

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Received 5 January 2017; received in received from 2 March 2017; accepted 6 March 2017

Abstract

We report on a 45-year-old male patient who presented with unilateral secondary dystonia. He had a history of a blow to the head at four years old. Right unilateral dystonia developed ten years later. A cyst-like change was found in the left globus pallidus medial segment by head CT and MRI. A blood flow decrease was found in the cerebral blood flow SPECT mainly on the left parietal lobe. As the mechanism of pathogenesis of the dystonia of this case, the contribution of the organic functional disorder of brain cortex - basal ganglia loop was suggested.

Key Words:

A blow to the head, dystonia, basal ganglia, globus pallidus, SPECT

Case Report

The patient was a 45-year-old, right-handed man. From about 40 years old, hypertension and hyperuricaemia had been treated. The family history was negative. The parent was not consanguineous marriage. He received a blow on the head from playground equipment at kindergarten at the age of four. He remained lucid, but vomiting and abnormal vision were found then. After home rest of approximately 40 days, he came to the kindergarten at a return garden. He broke his left hand joint in a traffic accident at the age of 14. A twisting-like involuntary movement of the right upper extremity appeared and became slowly aggravated. When he ran at a walk, his right foot first toe was numb, and he bent it. At the age of 15, he had a diagnosis of "right upper extremity dystonia, right leg paresis" in a certain neurosurgery. Abnormal findings were found in the left cerebral hemisphere by head computed tomography. However, the medical treatment did not have an effect. The movement disorder of top right corner lower limbs progressed from about 20 years old. When 45 years old in 2014, he visited the Tokushima National Hospital.

A general physical examination did not show any abnormality. Without a cardiac murmur, abnormal findings were not found in the chest and abdomen. As for the neurologic findings, he was lucid, and the intelligence was normal. MMSE was normal (30/30 points). Aphasia, apraxia, or agnosia were not found. Abnormal posture of the trunk was not found. The cranial nerve system was normal. The appendicular muscle tone was normal with the motor system, but the top right corner lower limbs had muscle weakness. The grip was 16 kg for the right side, 42 kg for the left. Voluntary movement of the right finger and the right leg was slow, and it was unskillful. When walking, the following involuntary movements were found. Twisting and adduction of the right upper extremity, and the athetosis-like exercise of the right finger and the flexure of the right toes appeared. The effect and the circadian rhythm were not found early in the morning. The sense

trick was not clear. The appendicular coordinated movement was normal. The deep tendon reflexes were normal, and the pathologic reflex was negative. In the right half of the body, palmesthesia decreased slightly, a vibratory sensation decreased slightly. There was no clear bladder rectal disorder. As for the test result, the peripheral blood and the blood sedimentation were normal. The blood chemical examination results were as follows, AST 22IU/l, ALT 27IU/l, γ-GTP 21IU/l, LDH 140IU/l, CPK 65IU/l, uric acid 5.4 mg/dl, BUN 8.1 mg/dl, Cr 0.74 mg/dl, Na 141mEq/l, K 3.6mEq/l, Cl 104 mEq/l, T-cho 189 mg/dl, TG 186 mg/dl, HDL-cho 43 mg/dl, blood glucose 106 mg/dl, HbA1c 5.7%, CRP 0.1 mg/dl, serum iron 74 μg/dl, serum ferritin 73.4ng/ml, serum copper 75 μg/dl, serum ceruloplasmin 20 mg/dl, lactic acid 10.4 mg/dl, pyruvic acid 0.61 mg/dl. The serum protein fraction was normal, and RA and the antinuclear antibody were negative. Electrocardiogram and chest XP did not show abnormal findings. The electroencephalogram was in the normal range without laterality. In a head CT (**Figure 1**) and head MRI (**Figures 2 and 3**), a cyst-like change was found in the left globus pallidus medial segment. Transformation was found in the temporal bones, and soft tissue expansion of the left lateral ventricle and the left maegashira. The depiction of the M1-M2 segment transitional region of bilateral MCA was poor. In the cerebral blood flow SPECT (**Figure 4**), a blood flow decrease was found in the left vertex, temple, occipital lobe and basal ganglia.

Discussion

As differential diagnoses of this case, cerebrovascular disorder, the aceruloplasminemia, Wilson's disease, and the mitochondrial abnormality symptom were denied. This case had a history of seriously illness following a blow to the head at the age of four, and top right corner lower limbs dystonia developed ten years later. An approved left globus pallidus medial segment lesion was regarded as a responsibility lesion of the unilateral secondary dystonia by CT, MRI. The following mechanisms are considered as showing the onset of dystonia mechanism. In other words, the activity of the globus pallidus medial segment and the substantia nigra pars reticulata decreases, and suppression of the thalamus and brain cortex decreases, and cerebro-cortical excitability is enhanced, and hyperkinesia occurs [1]. In this example, due to a decrease in output from the left globus pallidus medial segment, the inhibitory projection decreased for the left thalamus, and, as a result, the left thalamus might have become overactive. The dysfunction of the left thalamus - cortex projection increased, and it was judged that secondary dystonia had developed. Unilateral dystonia after head trauma is found in 0.9% of patients with head trauma [2]. After head trauma, the average latency period before unilateral dystonia develops is considered to be 20 months [3]. However, patients who received the head trauma before seven years old can be compared to patients who receive head trauma in adulthood and have a long latency period before unilateral dystonia develops [4]. In the past report, the shortest was 9th for the time to dystonic expression, and the most head was six years. As for this case, dystonia developed ten years later. There was much putamen in the lesion in the past report, but this case was globus pallidus medial segment. In this example, a blood flow decrease of the left hemisphere was confirmed. When we have a history of a blow to the head, it is necessary to be careful about basal ganglia lesions and cerebral blood flow disorders.

References

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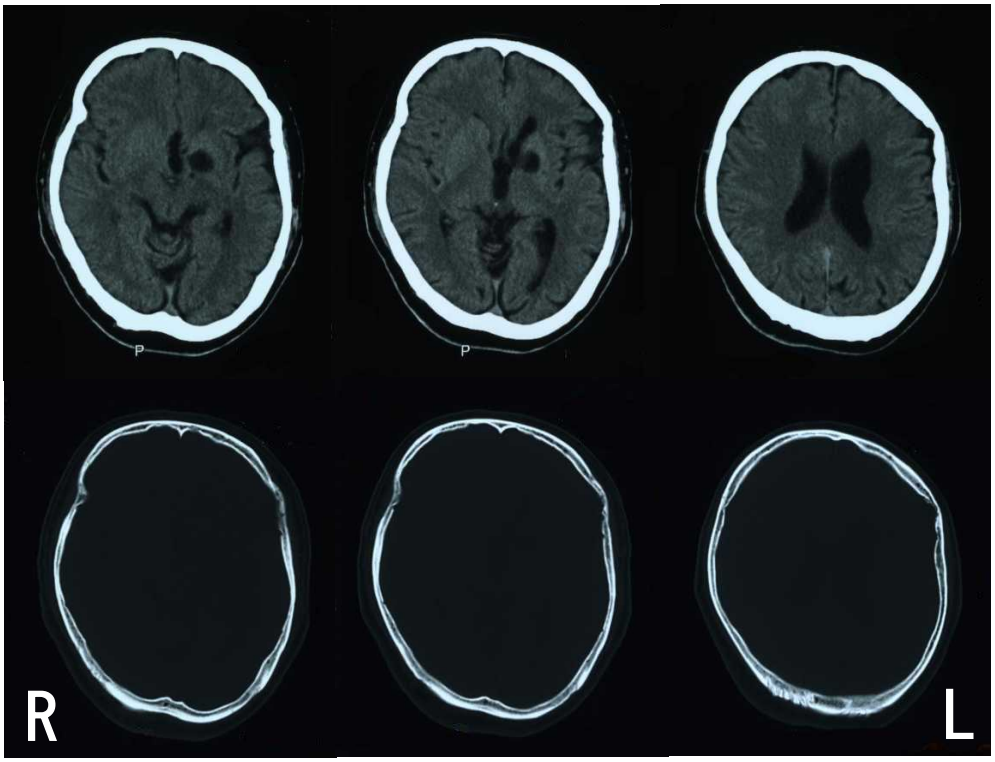

Figure 1. Brain CT of the present patient.

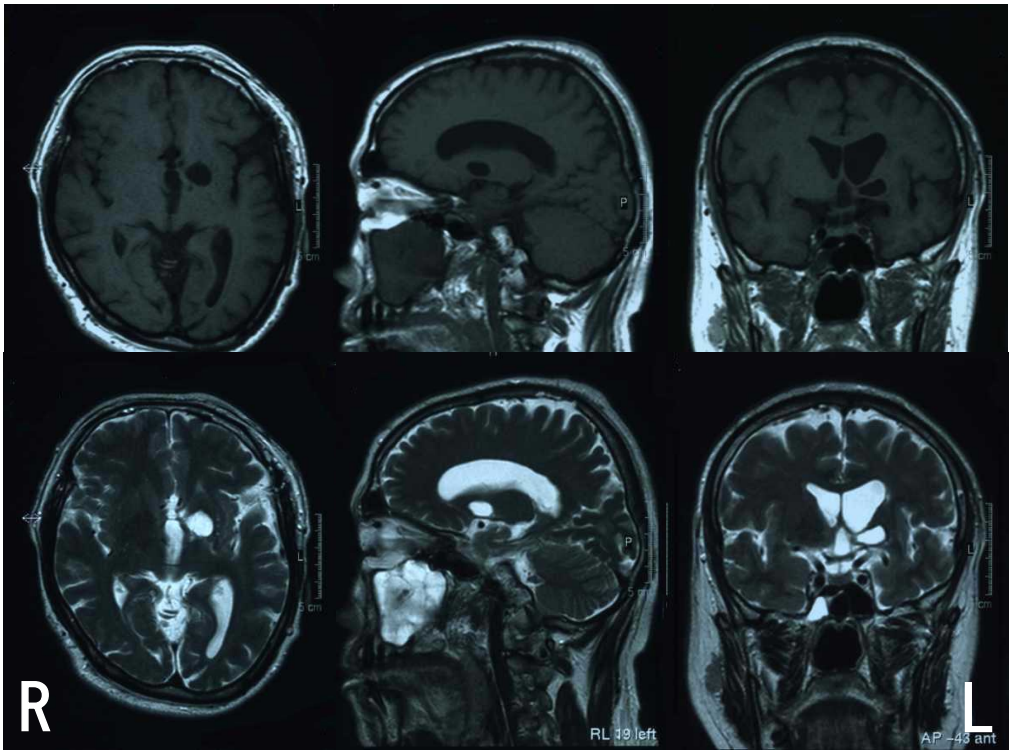

Figure 2. Brain MRI of the patient. Upper panels, T1-weighted images; lower panels, T2-weighted images.

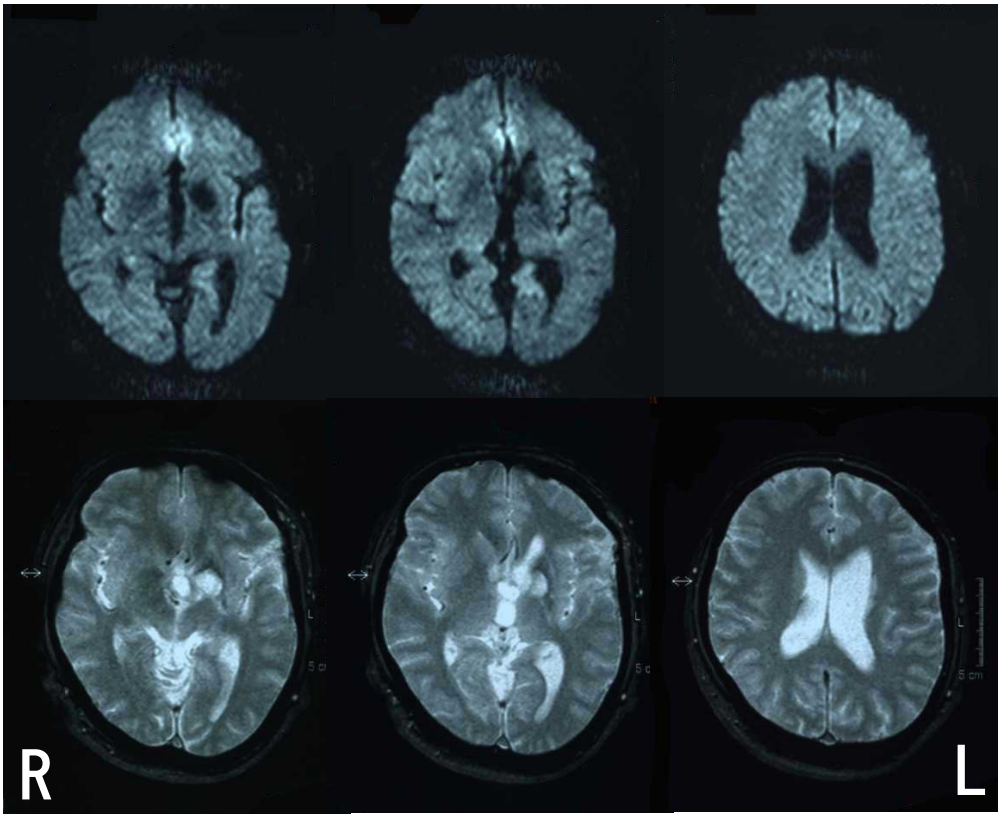

Figure 3. Brain MRI of the patient. Upper panels, Diffusion-weighted images; lower panels, T2 star-weighted images.

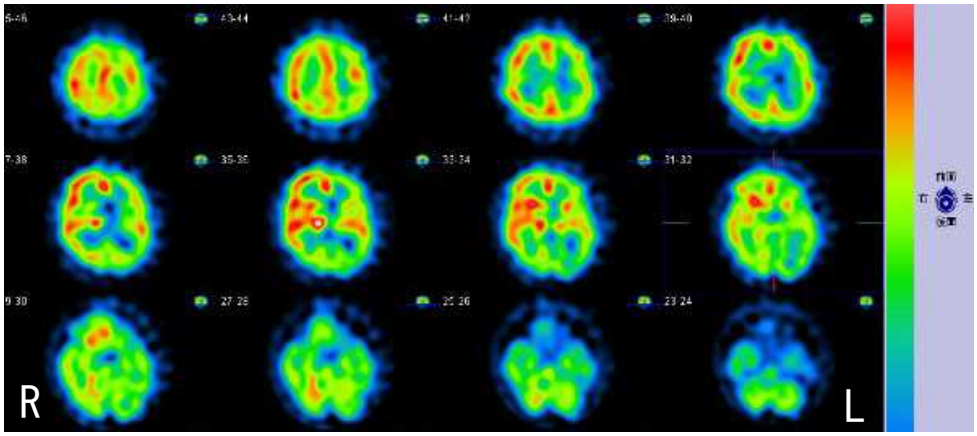

Figure 4. SPECT of the patient using ^{123}I-IMP