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Dramatic unilateral decrease in uptake via the dopamine transporter: Imaging in a patient with hemiparkinsonism following the lacunar stroke in substantia nigra

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Abstract

Vascular parkinsonism (VaP) is typically defined as having predominant lower body involvement, postural instability, less prominent rest tremor and little or no response to treatment with levodopa. In this study, we report a patient with VaP with clear demonstration of a dramatic unilateral decrease of radiotracer uptake in a ^{18}F-FP-CIT-PET study. A 62-year-old right-handed woman was referred to the neurology department due to rest tremor and rigidity in the right hand, which began after undergoing resection surgery for a left acoustic neuroma 7 years prior. Brain MRI, taken at 1 year after surgery showed an ischemic stroke lesion in the left medial pons and the left substantia nigra. ^{18}F-FP-CIT-PET revealed a marked reduction of radiotracer uptake in left striatum compared to that of the right. We treated the patient with 100 mg of levodopa, 200 mg of entacarpone and 25 mg of carbidopa. There was an improvement in bradykinesia and tremor, but the symptoms persisted, and there was no deterioration during 6 months of observation. After acoustic neuroma surgery, ischemic complications are uncommon, and even a small lesion in the nigrostriatal pathway can cause a hemiparkinsonism. If a patient experience sudden onset hemiparkinsonism, they should be carefully examined for lesions in the nigrostriatal pathways. Under these conditions, the ^{18}F-FP-CIT-PET scan can enable visualization of a unilateral decrease and is a useful tool for diagnosis and differentiation from idiopathic Parkinson's disease.

Keywords: Parkinson disease, secondary; parkinsonian disorders; secondary vascular parkinsonism; lacunar stroke; substantia nigra; positron emission on tomography; FP-CIT

INTRODUCTION

Vascular parkinsonism (VaP) was first described in 1929 by Critchley as "atherosclerotic parkinsonism"[1] and is usually defined by predominant lower body involvement, postural instability, less prominent rest tremor and little or no response to levodopa. VaP has been mostly associated with unilateral and bilateral infarctions in the striatum, lentiform nucleus, or cerebellar peduncle.[2-4] Most of the lesions observed have been multiple lacunar infarctions affecting the basal ganglia and hemispheric white matter but sparing the substantia nigra (SN).[5] There have been few cases reporting ischemic stroke in SN causing parkinsonism.[4,6-10] The ischemic lesion in SN produces parkinsonism by interfering with normal dopaminergic production or transmission. This dysfunction of the nigrostriatal dopaminergic pathway is accompanied by a reduction of striatal radiotracer uptake in PET studies.[11] In this study, we report a patient with VaP, determined by a dramatic unilateral decrease of radiotracer uptake in ^{18}F-FP-CIT-PET study.

CASE REPORT

A 62-year-old right-handed woman was referred to the neurology department due to rest tremor and rigidity in the right hand, which began after undergoing resection surgery for a left acoustic neuroma using translabyrinthine approach 7 years

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prior. Since this surgery, she had complained of tremor and weakness in her right upper and lower extremities. The patient had no history of drug ingestion, including dopa-blocking agents, head trauma, toxin exposure, infection, or metabolic disease. Neurological examination revealed rest tremor in the right hand. Bradykinesia and rigidity were observed in the right upper and lower extremities. Postural instability or short-stepped gait was not observed. The unified Parkinson's disease rating scale (UPDRS)[12] score was 18 and the Hoehn and Yahr stage was 1.[13] Although she complained of weakness on her right side, all motor power showed Medical Research Council (MRC) grade V. The patient underwent ^{18}F-FP-CIT- PET for dopamine transporter imaging, which revealed a marked reduction of radiotracer uptake in the left striatum compared to that of right (Figure 1).

We reviewed her past medical charts and brain imaging studies. Seven years ago, she was diagnosed with the left acoustic neuroma and underwent translabyrinthine approach tumor removal (Figure 2). Brain MRI taken on the day after the surgery revealed a lacunar infarction in the left medial pons and a bilateral subdural hemorrhage. She had right-sided weakness (MRC 4) after surgery, but the motor weakness improved after conservative management over 6 months. Fine-motor difficulty and right side rest tremor persisted. Initially, these symptoms were considered to be a complication of the ischemic stroke. As the symptoms persisted, an MRI was performed 1 year after surgery and a small encephalomalacia in left SN was confirmed (Figure 3). To check the current status, she received a follow-up MRI, which showed no interval change compared with the previous image (Figure 4). We treated her with 100 mg of levodopa, 200 mg of entacarpone and 25 mg of carbidopa. There was an improvement in bradykinesia and tremor, but the symptoms persisted, and there was no deterioration during 6 months of observation.

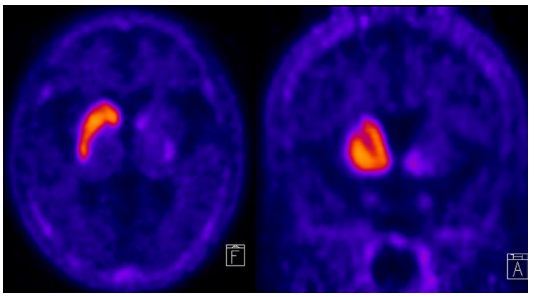

Figure 1. [^{18}F] FP-CIT PET showed a marked reduction of radiotracer uptake in left striatum with normal uptake in the right striatum.

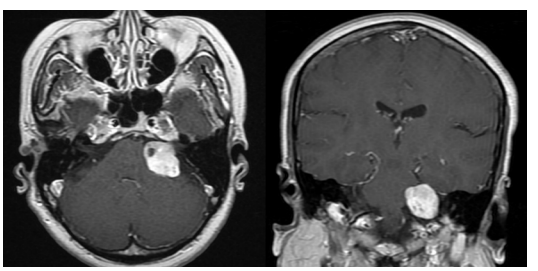

Figure 2. Magnetic resonance imaging (MRI) revealed a 2.1 x 2.9 cm sized acoustic neuroma in the left cerebellopontine angle.

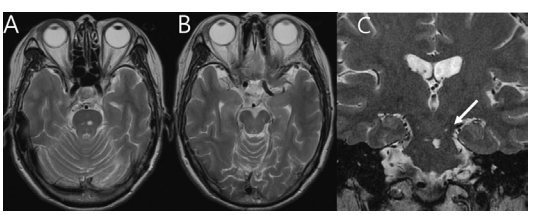

Figure 3. Magnetic resonance imaging (MRI) 1 year after surgery showed small encephalomalacia in the left medial pons (Fig. 3A) and the left substantia nigra (Fig. 3B) with the coronal image (Fig. 3C)

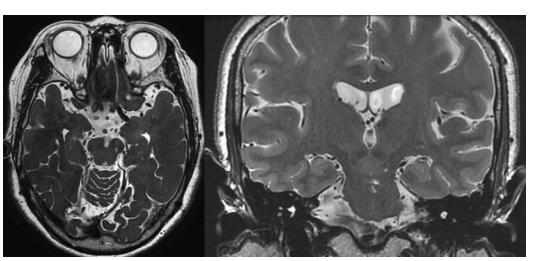

Figure 4. Magnetic resonance imaging (MRI) 7 years after surgery showed no interval change compared with previous images.

DISCUSSION

This case showed hemiparkinsonism following acoustic neuroma surgery. Initially the diagnosis was difficult to be certain due to accompanying weakness. Furthermore, there was no new lesions associated with parkinsonism on the first MRI. The left SN infarction which detected at 1 year after the surgery induced damage to the nigrostriatal pathways and hemiparkinsonism on the contralateral side occurred. Although we could not confirm the SN lesion in the brain MRI performed immediately after the surgery, this was

probably because the SN lesion was too small to be detected by routine MRI.

An updated diagnostic criteria suggested by expert working group divided VaP into 3 subtypes, acute, insidious and mixed neurodegenerative parkinsonism and cerebrovascular disease.[14] According to this classification, our case belongs to the acute VaP and it has been known to have good response to levodopa treatment, less prominent lower body symptoms and asymmetry of neurological involvement. These features are seen in our patient.

Unilateral reduction of presynaptic dopaminergic transporter uptake is an uncommon finding in Parkinson's disease (PD). Introduction of PET helps early diagnosis of PD patients who has hemiparkinsonism or even in subclinical PD.[15] Early stage PD patients with unilateral symptoms show a reduction of the striatal radiotracer uptake not only contralateral to the affected side but also ipsilateral.[11] The radiotracer uptake of the striatum decrease with disease progression bilaterally and asymmetrically. As seen in our patient, extreme reduction of uptake in the unilateral striatum suggests a secondary parkinsonism, rather than PD. Previously reported cases with SN infarctions using PET or single-photon emission computed tomography also showed reductions in striatal radiotracer uptake, which is similar to our patient.[6-8] However, previous cases did not show extreme differences between left and right striatum.

After acoustic neuroma surgery, ischemic complications are uncommon. Two previous studies reported 0.5%~0.86% of patients had a post-operative brainstem infarct.[16] The brainstem infarcts mostly occur as a result of tumor direct adherence to the brainstem and its vessels.[17] Acoustic neuroma had a high incidence of involvement of various arteries, such as superior cerebellar artery (79%), anterior inferior cerebellar artery (AICA) trunk (91.5%), AICA branches (100%), posterior inferior cerebellar artery (PICA) trunk (59.5%), PICA branches (79%), and the vertebral artery (93.5%).[18] In our patient, there was no evidence of involvement of the arteries or consequential brainstem infarct. These vessels are not directly associated with the SN lesion because the SN is primarily supplied by branches arising from the posterior communicating artery.[21] Thus, we presume that embolic infarction of vertebral artery or a basilar artery during surgery may have resulted in the SN lesion.

In conclusion, we report a case of hemiparkinsonism due to a lacunar infarction in the SN with marked reduction of radiotracer uptake in the left striatum on 18F-FP-CIT-PET. Our patient illustrate that even a small lesion in nigrostriatal pathway can cause hemiparkinsonism. If a patient experience sudden onset hemiparkinsonism, he or she should be carefully examined for lesions in the nigrostriatal pathways. Under these conditions, the 18F-FP-CIT-PET scan can help in the visualization of unilateral decrease, and is a useful tool for diagnosis and differentiation from PD.

DISCLOSURE

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Conflicts of interest: None

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