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Letter to the Editor

Shunt-Responsive Idiopathic Normal Pressure Hydrocephalus Patient With Parkinson's Disease-Compatible Findings on Dopamine Transporter Scans

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Dear Editor,

Idiopathic normal pressure hydrocephalus (INPH) involves non-obstructive enlargement of the cerebral ventricles along with symptoms of gait disturbance, cognitive impairment, and urinary dysfunction.[1] While INPH is treatable, the diagnosis of INPH can be difficult due to various degrees of the classic clinical symptoms and an overlap of symptoms with other common diseases such as Parkinson's disease (PD).[2] Fluorinated N-3-fluoropropyl-2β-carbomethoxy-3β-(4-iodophenyl)-nortropane (F-18 FP-CIT) positron emission tomography (PET) imaging has been widely used for evaluation of parkinsonian syndrome as dopaminergic system imaging.[3] F-18 FP-CIT PET is a helpful tool for imaging dopamine transporters (DATs).[3] There exists histopathological evidence of an association between the level of compromised DAT binding and the amount of loss of nigrostriatal nerve terminals.[3] Similar to observations in PD, in various forms of atypical parkinsonian syndrome a significant loss of striatal DAT binding is also observable.[3] As neurodegeneration of presynaptic nigrostriatal nerves is generally nonexistent in INPH, patients with this disease demonstrate normal DAT binding values and therefore can be easily distinguished from patients with neurodegenerative parkinsonian syndrome.[3] We present an INPH patient with abnormal F-18 FP-CIT PET scans.

A 66-year-old woman presented to our hospital for evaluation of progressive impairment of balance, gait, and memory. At the presentation, she could not walk independently and needed firm physical assistance. She presented with rest tremor bilaterally in the arms. Brain magnetic resonance imaging (MRI) showed communicating hydrocephalus with an Evans' ratio of 0.34 and a disproportionately enlarged subarachnoid space hydrocephalus (DESH) sign (**Fig. 1A**). She was diagnosed with INPH in accordance with the consensus criteria of Relkin et al.[2] F-18 FP-CIT PET imaging showed typical preferential DAT loss in the dorsal posterior putamen (**Fig. 1B**). A lumbar tap was performed to take out 40 mL of cerebrospinal fluid (CSF). She showed evident improvement in her gait and was judged as a responder according to the criteria proposed by Ishikawa et al.[1] (**Supplementary Table 1**). After considered discussion, she underwent shunt surgery. One month after ventriculoperitoneal shunt placement, she demonstrated a considerable improvement in gait, and her Korean Mini-Mental State

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Conflict of Interest

The authors have no financial conflicts of interest.

Author Contributions

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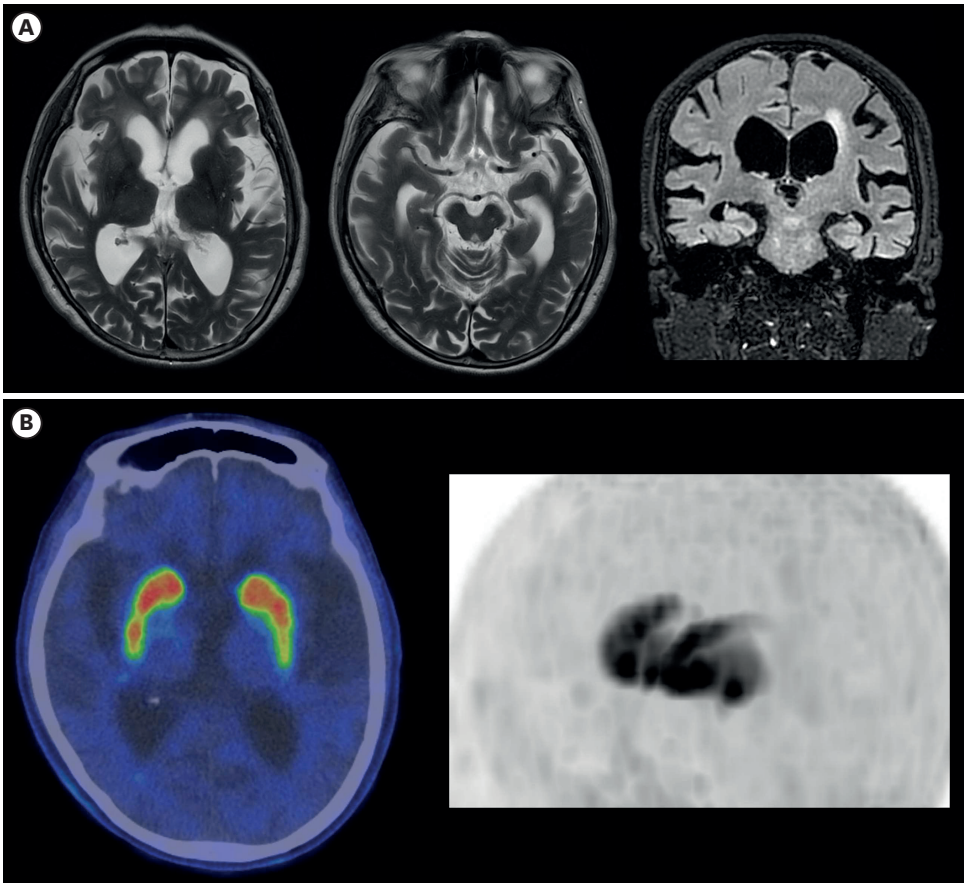

Fig. 1. (A) Preoperative brain magnetic resonance imaging. T2-weighted axial images show lateral ventricular enlargement with cerebrospinal fluid signal void in the cerebral aqueduct. Coronal fluid-attenuated inversion recovery image shows enlarged ventricles, tight high-convexity and medial surface subarachnoid spaces, and expanded Sylvian fissures. (B) Fluorinated N-3-fluoropropyl-2β-carbomethoxy-3β-(4-iodophenyl)-nortropane positron emission tomography images show typical preferential dopamine transporter loss in dorsal posterior putamen.

Examination (K-MMSE) score also improved to 14 (**Supplementary Table 1**). In terms of the subscores, the "Orientation" subscore and the "Recall" subscore of the K-MMSE improved. She was confirmed as shunt-responsive definite INPH.

The present case simultaneously demonstrated characteristic findings important in the diagnosis of INPH and PD. Relative to INPH, this case showed a DESH finding on MRI, improvement following the CSF tap test (CSFTT), and further improvement after shunt surgery. According to the Japanese INPH diagnostic criteria, improvement after CSFTT and a DESH finding raise the diagnosis accuracy of INPH from possible to probable, and improvement after shunt surgery raises the accuracy from probable to definite.[4] Relative to PD, this case clearly showed rest tremor with characteristic F-18 FP-CIT PET findings observed in PD patients. According to the PD diagnostic criteria, rest tremor is one of the core symptoms of PD, and F-18 FP-CIT PET abnormal findings are also important in support of the diagnosis.[5] Therefore, we judged that the patient had definite INPH with a very high possibility of comorbid PD.

Resting tremor is a cardinal symptom of PD. It occurs predominantly in the upper limbs. In contrast to PD, tremor at rest is known to be unusual among individuals with INPH.[6]

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Generally, lower body parkinsonism is a characteristic of INPH. Although it was reported that presynaptic dopaminergic depletion was not seen in patients with INPH, not every INPH patient showed normal DAT imaging in recent studies. One previous study reported that striatal dopaminergic depletion could be minimal in patients with INPH and these dopaminergic deficits had a higher incidence in the caudate nucleus than in the putamen, indicating a pattern different from PD.[7] In our patient, the comorbidity of PD was suggested from the upper limb resting tremor and PD-compatible findings on DAT scans. It seemed that comorbid PD modified the clinical symptoms in INPH by adding "upper" parkinsonism in the previously existing INPH-associated lower parkinsonism.

This is an interesting case of a shunt-responsive INPH patient demonstrating a clinically evident resting tremor of the upper limbs and severe gait impairment who showed PD-compatible findings on DAT scans. Clinicians should be aware that INPH and PD can occur together. In the differential diagnosis of elderly patients with prominent upper limb resting tremor who have abnormal DAT scans, we can consider a diagnosis of INPH after carefully examination. If shunt-responsive INPH is indicated by additional tests, shunt surgery should then be considered as a possible treatment option.

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SUPPLEMENTARY MATERIAL

Supplementary Table 1

Results of assessments during CSF tap test and in the postoperative follow-up

REFERENCES

1. Ishikawa M, Hashimoto M, Kuwana N, Mori E, Miyake H, Wachi A, et al. Guidelines for management of idiopathic normal pressure hydrocephalus. Neurol Med Chir (Tokyo) 2008;48 Suppl:S1-S23. PUBMED | CROSSREF
2. Relkin N, Marmarou A, Klinge P, Bergsneider M, Black PM. Diagnosing idiopathic normal-pressure hydrocephalus. Neurosurgery 2005;57:S4-S16. PUBMED | CROSSREF
3. Tatsch K, Poepperl G. Nigrostriatal dopamine terminal imaging with dopamine transporter SPECT: an update. J Nucl Med 2013;54:1331-1338. PUBMED | CROSSREF
4. Mori E, Ishikawa M, Kato T, Kazui H, Miyake H, Miyajima M, et al. Guidelines for management of idiopathic normal pressure hydrocephalus: second edition. Neurol Med Chir (Tokyo) 2012;52:775-809. PUBMED | CROSSREF
5. Postuma RB, Berg D, Stern M, Poewe W, Olanow CW, Oertel W, et al. MDS clinical diagnostic criteria for Parkinson's disease. Mov Disord 2015;30:1591-1601. PUBMED | CROSSREF
6. Molde K, Söderström L, Laurell K. Parkinsonian symptoms in normal pressure hydrocephalus: a population-based study. J Neurol 2017;264:2141-2148. PUBMED | CROSSREF
7. Lee JY, Park SB, Lee M, Ju H, Im K, Kwon KY. Detailed visual assessment of striatal dopaminergic depletion in patients with idiopathic normal pressure hydrocephalus: unremarkable or not? BMC Neurol 2020;20:277. PUBMED | CROSSREF