

CASE STUDY

Development of involuntary movements after ventriculoperitoneal shunting for normal pressure hydrocephalus in a patient with chronic-phase thalamic haemorrhage

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Abstract

Background: Delayed-onset involuntary movements have been described after thalamic stroke.

Methods: We treated a patient with involuntary movements that increased after ventriculoperitoneal shunting (VPS) for normal pressure hydrocephalus (NPH) following thalamic haemorrhage. One and one-half years after right thalamic and intraventricular haemorrhage, NPH suggested clinical evaluation and neuroimaging studies in a 56-year-old man.

Results: Hemidystonia and pseudochoreoathetosis were evident in the left arm, leg and trunk. Proprioceptive impairment and mild cerebellar dysfunction affected the left upper and lower extremity. Yet the patient could walk unassisted and carry out activities of daily living (ADL) rated as 90 points according to the Barthel Index (BI). Lumbar puncture lessened both gait disturbance and cognitive impairment. After VPS, cognition and urinary continence improved, but involuntary movements worsened, precluding unaided ambulation and decreasing the BI score to 65 points. Computed tomography after VPS showed resolution of NPH, while single-photon emission computed tomography showed increased cerebral blood flow after VPS.

Conclusion: Increased cerebral blood flow after VPS is suspected to have promoted development of abnormal neuronal circuitry.

Keywords: *Normal pressure hydrocephalus, ventriculoperitoneal shunting, involuntary movement, stroke, thalamus*

Introduction

Involuntary movements after thalamic stroke have been described as showing various forms such as dystonia, pseudochoreoathetosis and myoclonus. Their emergence has ranged from 2 days to 10 years after onset of thalamic stroke [1–6]. Mechanisms according for delayed-onset may include development of abnormal neuronal circuitry or gradual changes in neuronal synaptic activities [1–3]. The authors know of no previous reports describing progressive involuntary movements following ventriculoperitoneal shunting

(VPS) for treatment of normal pressure hydrocephalus (NPH) occurring after thalamic stroke.

This case study describes a patient with right thalamic haemorrhage who developed hemidystonia and pseudochoreoathetosis after VPS for NPH.

Case presentation

In a 56-year-old man with sudden onset of left hemiparesis, computed tomography (CT) scan showed right thalamic and intraventricular haemorrhage. After conservative treatment, rehabilitation was initiated 1 week after stroke onset. At 107 days

Table I. Changing in clinical variables in relation to interventions.

	Admission	Discharge	Readmission	Post-STT	Pre-VPS	Post-VPS
Days from onset	107	187	526	534	553	594
MMSE	26	26	25	29	27	30
Reaction time (ms)	NT	NT	257	249	213	193
TMT-A (s)	NT	NT	116	67	72	62
Left BRS	IV/V/V	IV/V/V	IV/V/V	IV/V/V	IV/V/V	IV/V/V
Left MFS (%)	47	63	63	63	63	56
MWS (m min ⁻¹)	37	106	93	99	90	15
Barthel Index	60	90	90	90	90	65
Involuntary movements	No	Slight	Moderate	Mild to moderate	Moderate	Severe

STT, spinal tap test; VPS, ventriculoperitoneal shunting; MMSE, Mini-Mental State Examination; TMT-A, Trail-making Test-A; BRS, Brunnstrom Recovery Stage in proximal upper extremity/distal upper extremity/lower extremity; MFS, Manual Function Test; MWS, maximum walking speed.

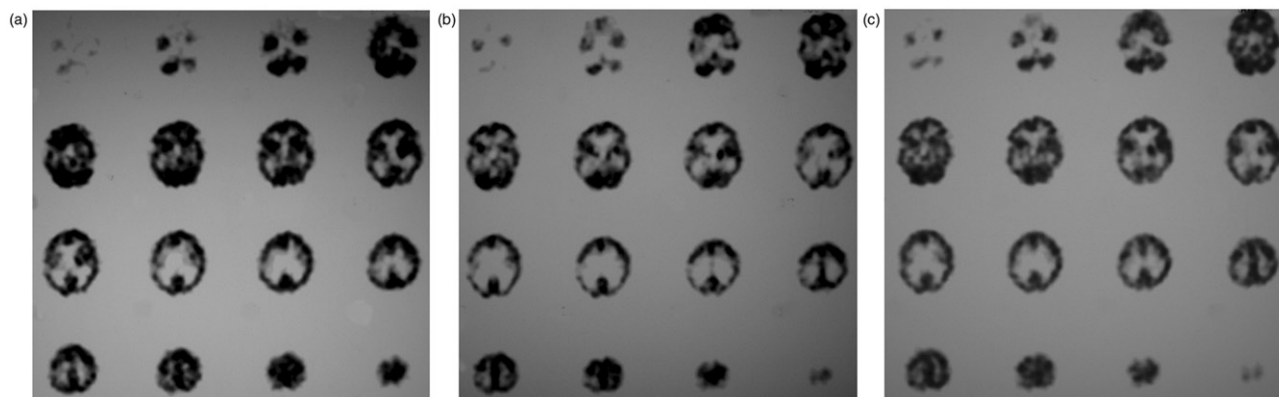


Figure 1. Cerebral blood flow (CBF) demonstrated by single-photon emission computed tomography (SPECT, using ¹²³I-iomazenil). (a) Decreased CBF in the right thalamus and bilaterally in the white matter at 126 days from stroke onset. (b) Further reduction in CBF in all lesions at 549 days from stroke onset. (c) Increased CBF after ventriculoperitoneal shunting at 594 days from stroke onset.

after stroke onset, he was transferred to the university hospital to undergo additional rehabilitation. Results of clinical evaluations are shown in Table I.

The patient showed mild-to-moderate left hemiparesis, marked sensory disturbance and mild cerebellar ataxia, but no involuntary movements. Cerebral blood flow (CBF) demonstrated by single-photon emission computed tomography (SPECT, using ¹²³I-iomazenil) was decreased in the right thalamus and bilaterally in the white matter (Figure 1(a)). Magnetic resonance imaging (MRI) at 121 days from stroke onset suggested NPH. A spinal tap test (STT) resulted in no differences in the cognitive disorder or gait disturbance. He could walk and use the left arm more effectively, but no change in hemiparesis was evident when he was discharged from the university hospital after 80 days of rehabilitation (Table I); mild hemidystonia and pseudochooreoathetosis were present in his left arm and leg.

During monthly follow-up in the university hospital outpatient clinic, the patient gradually developed mixed involuntary movements, inattention and urinary dysfunction. He took nifedipine (20 mg), candesartan cilexetil (4 mg), loxoprofen sodium (180 mg) and teprenone (150 mg) a day. In addition, he had taken clonazepam (1 mg) a day from 157–450 days and trihexyphenidyl hydrochloride (4 mg) a day from 387–415 days from stroke onset. As MRI findings suggested NPH (Figure 2), he was re-admitted for evaluation NPH 526 days from stroke onset (Table I).

Hemidystonia and pseudochooreoathetosis had worsened in the left arm, leg and apparent in the trunk. He could carry out activities of daily living (ADL) rated as 90 points according to the Barthel Index (BI), scored as dependent for 'bathing' and occasional accident for 'bladder'. SPECT showed further reduction in CBF in all lesions compared with CBF demonstrated at the first admission (Figure 1(b)). He again underwent STT, with

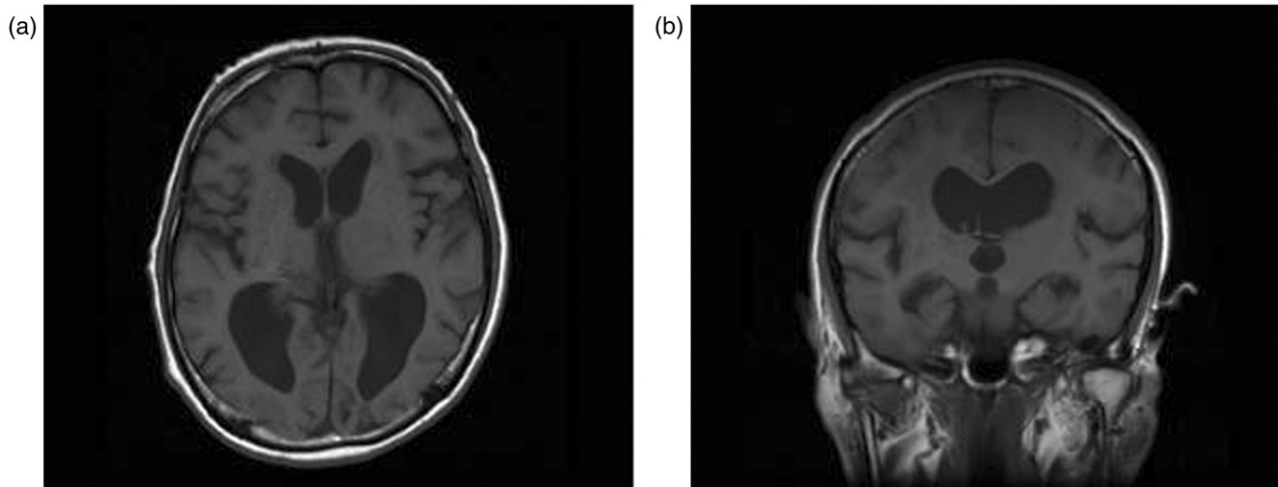


Figure 2. T_1 -weighted brain magnetic resonance imaging (MRI) findings at re-admission 502 days after right thalamic haemorrhage. (a) Post-haemorrhagic changes are seen in the right thalamus, while scattered infarcts are in the white matter and basal ganglia. (b) Ventricular size and contour are consistent with normal pressure hydrocephalus.

30 ml of cerebrospinal fluid withdrawn via lumbar puncture between the fourth and fifth lumbar spines. Results for the Mini-Mental State Examination (MMSE), reaction time and Trail-making Test-A (TMT-A), as well as gait, were significantly improved at 1 day but showed deterioration at 4 days after STT (Table I). Involuntary movements transiently appeared to decrease after STT. The STT findings were considered highly suggestive of reversible NPH manifestations and VPS was carried out with a Codman-Hakim programmable valve at 555 days from stroke onset. Initial shunt pressure was 20 cm H_2O , this was decreased twice weekly in increments of 2 cm H_2O based on clinical evaluations and CT findings. Unexpectedly, mixed involuntary movements appeared and worsened from day-to-day.

At 39 days after VPS, at a shunt pressure of 12 cm H_2O , CT showed reduced ventricular size. SPECT indicated that CBF had increased more than present at pre-VPS (Figure 1(c)). The MMSE, reaction time and TMT-A results improved significantly and urinary dysfunction pollakiuria diminished. In spite of this; unexpectedly worsened left-sided hemidystonia and pseudochoreoathetosis resulted in an overall decrease in performance. His scores of the BI fell to 65 points. The ADL items 'dressing', 'toilet use', 'transfers', 'mobility' and 'stairs' worsened and were scored as 5, 5, 10, 5 and 5, respectively.

Thus, while NPH was certainly alleviated by VPS, mixed involuntary movements developed after VPS. Subsequent therapeutic trials of medications including clonazepam, baclofen and fluvoxamine showed little affect against these movements. At 75 days after VPS, the patient transferred to another

hospital to be treated for the mixed involuntary movements.

Discussion

When the patient developed NPH after right thalamic and intraventricular haemorrhage, STT showed lessening of both global cognitive impairment and gait disturbance. VPS was effective against global cognitive impairment and urinary dysfunction and induced normalization of ventricular size as shown by CT and increased CBF as shown by SPECT. However, gait and left arm performance worsened because of unexpected development of mixed involuntary movements. Thus, a large discrepancy existed between changes in mixed involuntary movements following STT vs VPS. The authors know of no previous studies describing a relationship between NPH and involuntary movements after thalamic stroke.

Delayed-onset involuntary movements after thalamic stroke have been linked with proprioceptive sensory deficits, cerebellar dysfunction and unbalanced recovery from motor dysfunction or spasticity [1–6]. The movements were related to lesions in the ventrolateral, ventroposterolateral and paramedian nuclei of the thalamus [1–3, 5–7], which contain portions of the proprioceptive, cerebellar, pyramidal and sub-thalamic pathways. Disturbances of these afferent and efferent pathways would contribute to development of involuntary movements over time. This patient would have potential for involuntary movements based upon neurologic and

neuroimaging findings. However, progression of involuntary movements after VPS for NPH was unexpected.

Neuroimaging studies have shown decreases in CBF decreased in white matter, thalamus and basal ganglia in patients with NPH [8, 9]. CBF decreases resulting from NPH may have inhibited development of abnormal neuronal circuitry, while increased CBF resulting from shunt drainage of NPH may have activated such abnormal pathways.

Difference in effects on NPH between STT and VPS might be based on a difference in changes of CBF. Dumarey et al. [10] reported that gait improvement after STT in patients with NPH is associated with a bilateral regional CBF increase in the dorsolateral frontal cortex and an increase in the left mesial temporal cortex. This patient could walk more rapidly and smoothly after STT, because of partial alleviation of NPH. On the other hand, the greater CBF increase resulting from VPS might have permitted development of neuronal activity patterns that disrupted gait by producing involuntary movements.

Conclusion

Delayed-onset involuntary movements worsened after VPS for NPH in a patient with thalamic haemorrhage, which was not predicted by the response to STT. Increased CBF after VPS may have promoted development of abnormal neuronal circuitry. Clinicians will need to consider such possibilities in preventing and managing delayed-onset mixed involuntary movements after thalamic stroke.

References

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