

Recovery of akinetic mutism and injured prefronto-caudate tract following shunt operation for hydrocephalus and rehabilitation

A case report

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Abstract

Rationale: A 76-year-old female patient was diagnosed with an aneurysmal subarachnoid hemorrhage following rupture of a right posterior communicating artery aneurysm.

Patient concerns: She was treated surgically with clipping of the aneurysmal neck. Six months after onset, when starting rehabilitation at our hospital, she showed no spontaneous movement or speech.

Diagnoses: aneurysmal subarachnoid hemorrhage following rupture of a right posterior communicating artery aneurysm.

Interventions: During 2 months' rehabilitation, her AM did not improve significantly. As there was no apparent change, she underwent a ventriculo-peritoneal shunt operation for hydrocephalus 8 months after her stroke. After the surgery, she remained in the AM state, but participated in a comprehensive rehabilitative management program similar to that before shunt operation. During 1 month's intensive rehabilitation, her AM gradually improved. At 9 months after onset, she became able to perform some daily activities by herself including eating, washing, and dressing. In addition, she could speak with some fluency.

Outcomes: On 6-month DTT, the neural connectivity of the caudate nucleus (CN) to the medial prefrontal cortex (PFC, Brodmann area [BA]: 10 and 12) and orbito-frontal cortex (BA 11 and 13) was low in both hemispheres. However, the neural connectivity of the CN to the medial PFC increased on both sides on 9-month DTT. The integrity of the arcuate fasciculus (AF) was preserved in both hemispheres on both 6- and 9-month DTTs.

Lessons: Recovery of AM and injured PCTs was observed in a stroke patient.

Abbreviations: AF = arcuate fasciculus, AM = akinetic mutism, BA = Brodmann area, CN = caudate nucleus, DTI = diffusion tensor imaging, DTT = diffusion tensor tractography, PCT = pre-fronto-caudate tract, ROI = seed region of interest, WAB = Western Aphasia Battery.

Keywords: akinetic mutism, caudate nucleus, diffusion tensor tractography, pre-fronto-caudate tract, stroke

1. Introduction

Akinetic mutism (AM) is defined as a complete absence of spontaneous behavior and speech.^[1,2] It is a serious sequela of brain injury in terms of absolute need for a caregiver and loss of

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working ability.^[1,2] Bilateral injury of the fronto-subcortical circuit is a possible pathogenetic mechanism of AM: the pre-fronto-caudate tract (PCT) is most associated with the development of AM.^[3–6] Diffusion tensor tractography (DTT), derived from diffusion tensor imaging (DTI) data, allows reconstruction of the PCT. A few studies using DTT have diagnosed injury of the PCT in patients with brain injury.^[4–6] However, very little is known about recovery from AM and injured PCT.

In this study, we report on a stroke patient who recovered from AM and an injured PCT following a shunt operation for hydrocephalus and rehabilitation, demonstrated by serial DTTs.

2. Case report

A 76-year-old female patient was diagnosed with an aneurysmal SAH, a rupture of a right posterior communicating artery aneurysm. She underwent craniotomy and aneurysmal neck clipping at the neurosurgery department of a university hospital (Fig. 1A). Six months after onset, she was admitted to the rehabilitation department of another university hospital. She did not move or speak spontaneously; her language function was uncheckable using the Korean-Western Aphasia Battery (K-WAB).^[7] A brain MRI showed leukomalactic lesions in both

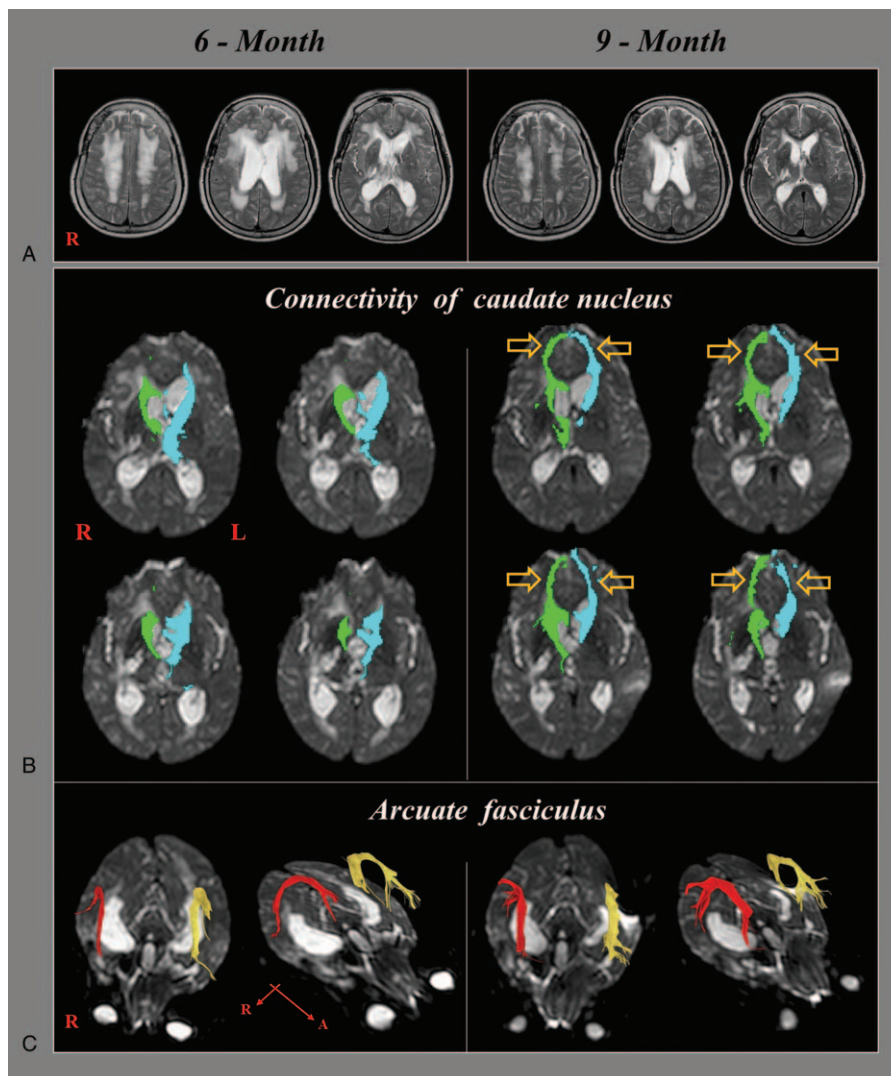


Figure 1. (A) T2-weighted brain MR images at 6 months after onset show leukomalactic lesions in both fronto-parieto-occipital areas, right thalamus and hydrocephalus and relief of hydrocephalus at 9 months after onset. (B) On 6-month diffusion tensor tractography, the neural connectivity of the CN to the medial prefrontal cortex (Broadmann area: 10 and 12) and orbitofrontal cortex (Broadmann area 11 and 13) is decreased in both hemispheres. However, the neural connectivity of the CN to the medial prefrontal cortex is increased on both sides (arrows) on 9-month DTT. The integrity of AF is preserved in both hemispheres on both 6- and 9-month DTTs. AF=arcuate fasciculus, CN=caudate nucleus, DTT=diffusion tensor tractography, MR=magnetic resonance.

fronto-parieto-occipital areas, right thalamus, and hydrocephalus (Fig. 1A). The patient underwent a comprehensive rehabilitative management program, including physical and occupational therapy, and dopaminergic drugs to improve AM (pramipexole, amantadine, ropinirole, and levodopa).^[8–10] During 2 months' rehabilitation, her AM did not significantly improve. She underwent a ventriculo-peritoneal shunt operation for hydrocephalus 8 months after the subarachnoid hemorrhage occurred. After the shunt surgery, she remained in the AM state. She participated in a comprehensive rehabilitative management program similar to that before her shunt operation. During 1 month's intensive rehabilitation, her AM gradually improved. Nine months after onset, she became able to perform some daily activities by herself including eating, washing, and dressing. In addition, she could speak with some fluency; her language ability was 81% (aphasia quotient, full score; 100%) on the WAB.^[7] The patient's husband provided signed, informed consent, and our institutional review board approved the study protocol.

DTI data were acquired 6 months and 9 months after onset using a 6-channel head coil on a 1.5T Philips Gyroscan Intera (Philips, Ltd., Best, The Netherlands) with 32 diffusion gradients by single-shot echo-planar imaging. Imaging parameters were as follows: acquisition matrix=96 × 96; reconstructed to matrix=192 × 192; field of view=240 × 240 mm²; TR=10,398 ms; TE=72 ms; parallel imaging reduction factor=2; echo-planar imaging factor=59; $b=1000$ s/mm²; and a slice thickness of 2.5 mm. Head motion effect and image distortion due to eddy current were corrected by affine multiscale two-dimensional registration.

2.1. Reconstruction of the caudate nucleus

Fiber tracking used probabilistic tractography, applying the default tractography option in the Oxford Centre for Functional Magnetic Resonance Imaging of the Brain (FMRIB) Diffusion Software (5000 streamline samples, 0.5 mm step lengths, curvature thresholds=0.2). To reconstruct the caudate nucleus

(CN), the seed region of interest (ROI) was placed on the CN, which was isolated by adjacent structures.^[11] The threshold of 10 streamlines was applied for fiber tracking.

2.2. Reconstruction of the arcuate fasciculus

Fiber tracking used the fiber assignment continuous tracking algorithm implemented within the DTI task card software (Philips Extended MR Work Space 2.6.). To reconstruct the arcuate fasciculus (AF), the seed ROI was placed on the deep white matter of the posterior parietal portion of the superior longitudinal fascicle. The second ROI was placed on the posterior temporal lobe.^[12] The termination criteria used for fiber tracking were $FA < 0.15$, $angle < 27^\circ$.

The 6-month DTT showed that the neural connectivity of the CN to the medial prefrontal cortex (PFC, Brodmann area [BA]: 10 and 12) and orbitofrontal cortex (BA 11 and 13) was low in both hemispheres. However, the neural connectivity of the CN to the medial PFC increased on both sides on 9-month DTT. The integrity of AF was preserved in both hemispheres on both 6- and 9-month DTTs.

3. Discussion

In the current study, we cared for a stroke patient who recovered from AM following a shunt operation for hydrocephalus and rehabilitation. Before the shunt operation (done 8 months after the SAH), the patient showed typical clinical features of AM. On 6-month DTT, we found that the neural connectivity of the CN to the medial PFC, associated with motivation, was low in both hemispheres.^[1,3,13-16] This indicated an injury of the PCT in both hemispheres, and these injuries appeared to be mainly ascribed to hydrocephalus. Although the shunt operation was recommended for the patient, her family insisted upon rehabilitation for fear of surgical complications. At 8 months after onset, she was obliged to undergo a shunt operation for hydrocephalus, as there was no significant recovery of AM despite intensive rehabilitation. The patient remained in the AM state after the shunt operation, but her condition improved significantly during 1 month's rehabilitation following the surgery. She developed some spontaneity in activities of daily living and language. On 9-month DTT, the neural connectivity of the CN to the medial PFC was restored. This suggested recovery of the injured PCTs. Recovery of the injured PCTs in both hemispheres contributed to clinical recovery of AM. We think that the relief of hydrocephalus by shunt operation was the primary reason for the recovery of the injured PCTs, and rehabilitation contributed additionally to recovery from AM in this patient.

In conclusion, recovery from AM and injured PCTs were demonstrated in a stroke patient. Therefore, our results stress the importance of evaluation of the PCT in patients with AM after brain injury and rehabilitation. To the best of our knowledge, this is the first study to demonstrate recovery of an injured PCT in a stroke patient. However, the limitations of DTT should be considered: crossing fiber and partial volume effects may produce false negative results.^[17]

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