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Compensatory Hyperactivity of the Ipsilesional Red Nucleus in a Patient With Somatosensory Cortex Damage: A Case Report

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HIGHLIGHTS

- We reported a patient with damage to the primary somatosensory cortex.
- We observed hyperactivity in the ipsilesional red nucleus in this patient.
- This might be related to the motor recovery of the patient.

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Compensatory Hyperactivity of the Ipsilesional Red Nucleus in a Patient With Somatosensory Cortex Damage: A Case Report

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ABSTRACT

This case study describes a patient who experienced motor recovery and involuntary movements following damage to the right primary somatosensory cortex caused by an intracranial hemorrhage. The patient initially suffered from paralysis in her left arm and leg, but exhibited significant motor recovery later, accompanied by multiple episodes of ballistic movement during the recovery process. A diffusion tensor imaging analysis was performed to investigate changes in sensorimotor-related brain areas in the patient. The patient had higher fractional anisotropy and lower mean diffusivity values in the ipsilesional red nucleus (RN) than age-matched controls. We assume that hyperactivity of the ipsilesional RN might play a role in motor recovery after damage to the primary somatosensory cortex, potentially through its involvement in sensorimotor integration. Our findings demonstrated the potential for adaptive changes in the ipsilesional RN following damage to the primary somatosensory cortex.

Keywords: Somatosensory Cortex; Intracranial Hemorrhages; Primary Motor Cortex; Red Nucleus

INTRODUCTION

The primary somatosensory cortex (S1), associated with sensory information processing, is interconnected with motor regions and can influence movement [1,2]. Motor output is abnormal or inaccurate when sensory input is not properly processed. Damage to the S1 may result in neurological conditions characterized by motor dysfunction [3-5]. Herein, we present the case of a patient who experienced motor recovery and involuntary movements following damage to the right S1 caused by an intracranial hemorrhage (ICH). This study used diffusion tensor imaging (DTI) to investigate the changes in sensorimotor-related brain areas affected by alterations in the S1. The patient has provided informed consent for the publication of this case.

Funding

None.

Conflict of Interest

The authors have no potential conflicts of interest to disclose.

Author Contributions

Conceptualization: Yoo WK, Lee J; Data curation: Lee E, Kim GJ; Formal analysis: Ohn SH, Jung KI, Bashir S; Writing - original draft: Lee J; Writing - review & editing: Lee J, Yoo WK.

CASE DESCRIPTION

A 20-year-old right-handed woman with no underlying disease visited the emergency room because of a sudden onset of mental change. Upon arrival, brain computed tomography revealed acute ICH in the right frontoparietal cortex with midline shifting (**Fig. 1A**). She received surgical treatment on March 31, 2023 and was transferred to the Department of Rehabilitation Medicine on April 17, 2023. Initially, she experienced paralysis in her left arm and leg, although voluntary movement started to return first in her leg. She gradually regained movement in her arm by co-contracting flexor and extensor muscles, accompanied by increased muscle tone. At 5 weeks postoperatively, she developed sudden involuntary movements characterized by high amplitude, arrhythmic, and rapid movements in both upper and lower extremities. She demonstrated flinging and swinging movements owing to sudden contractions of the proximal muscles of her arms. These movements included abduction, adduction, internal and external rotation of the arms, and extension of the forearms. They were accompanied by restlessness in her legs and grimacing facial expressions. She continuously repeated flexion and extension of her hip and knee joints. These movements persisted for 5 min and were managed with an injection of lorazepam. This symptom occurred twice, with a 2-day interval between occurrences. No epileptiform discharges were detected by electroencephalography. The patient reported experiencing similar symptoms twice after being discharged from our hospital on May 12, 2023. She was readmitted to our hospital on June 20, 2023, and since her readmission, no recurrences of the same symptoms were observed. After undergoing 3 months of rehabilitation therapy following brain hemorrhage, the Fugl-Meyer Assessments for upper extremity and lower extremity scale scores improved from 2 to 46 and from 3 to 16, respectively. In the box and block test, the patient was unable to move blocks with the affected hand, but was able to move 11 blocks after regaining hand function. Her modified Barthel index score improved from 11 to 57. She required maximum assistance to walk 10 m before undergoing rehabilitation therapy; however, she was able to walk independently within 24 seconds after rehabilitation therapy. We performed magnetic resonance imaging (MRI) and DTI twice. The initial brain MRI, which was conducted 17 days after the onset of symptoms, revealed ICH in the right S1, with no lesions in the bilateral thalamus and red nucleus (RN). A follow-up brain MRI was performed 3 months after the onset of symptoms (**Fig. 1B and C**). We extracted the

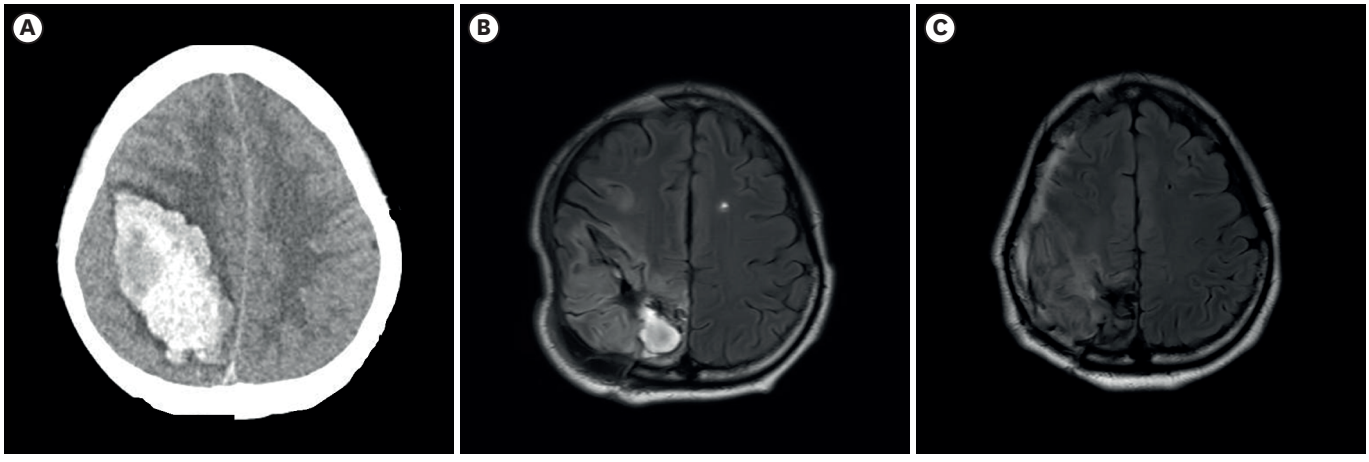

Fig. 1. Brain images of the patient. (A) Initial brain computed tomography revealed ICH in the right frontoparietal lobe with midline shifting. (B) Brain MRI which was conducted 17 days after the onset of symptoms revealed ICH in the right S1. (C) Brain MRI which was performed 3 months after the onset of symptoms revealed resolution of hemorrhage and encephalomalacic changes in the right S1.
ICH, intracranial hemorrhage; MRI, magnetic resonance imaging; S1, primary somatosensory cortex.

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diffusion metrics from the regions of interest (ROIs) at 2 time points: initial and follow-up assessments. These diffusion metrics were compared with those of age-matched controls (4 controls, average age 20.8 ± 2.4, M:F = 2:2).

DTI was acquired using a diffusion-weighted, echo-planar imaging sequence (repetition time = 5,000 ms; echo time = 100 ms; slice thickness = 2.2 mm; no gap; in-plane resolution = 2.4 × 2.4 mm, 32 independent diffusion gradient directions using b = 1,000 s/mm^2). DTI data were analyzed using the FMRIB Diffusion Toolbox from the FMRIB Software Library (Oxford, UK). The ROIs were selected from the hand areas of the primary motor cortex (M1), leg areas of M1, RN, S1, and thalamus. The ROIs of the hand areas of M1, leg areas of M1, and RN were manually drawn. The ROIs of the hand areas of the M1 were located along the precentral gyrus based on the anatomical location of the hand knob and the leg areas of M1 were located close to the midline within interior sections folding into the medial longitudinal fissure. The ROIs of the RN corresponded to the dark area in the midbrain on the T2 weighted image. The ROIs of the S1 and thalamus were based on 3D T1 images using the FreeSurfer software version 5.1.0. All the ROIs were acquired from the standard space and projected onto individual spaces using an inverse transformation matrix. We extracted the fractional anisotropy (FA) and mean diffusivity (MD) from the ROIs of both the patient and controls. We used a modified t-test for comparisons. It treats the control sample as a reference and establishes a confidence interval based on the deviation of scores from the norm [6,7]. Statistical analyses were conducted using the Singlims_ES.exe program. Significance was set at $p < 0.05$. This study was approved by the Institutional Review Board (IRB) of the Hallym University Sacred Heart Hospital (IRB No. 2023-08-023).

The patient initial had significantly lower FA and higher MD values in the right S1 compared to the control group. The FA and MD values in the right S1 slightly increased at the follow-up DTI (**Supplementary Tables 1** and **2**). Compared with the control group, the patient initially had significantly lower FA and higher MD values in the right areas of M1. In the follow-up DTI, an increase in the FA value and a decrease in the MD value were observed on the right side of M1. The patient had lower MD values in the left-hand areas of M1 than the control group at both the initial and follow-up assessments (**Fig. 2A**). The patient initially had significantly higher FA and lower MD values in the right RN than the control group. After 3 months, the FA value decreased, and the MD value in the right RN increased. No significant differences were observed in the left RN (**Fig. 2B**). No significant differences in either the FA or MD values of the bilateral leg areas of M1, left S1, and bilateral thalamus were also noted (**Supplementary Tables 1** and **2**).

DISCUSSION

This case study demonstrated changes in diffusion metrics, not only within sensorimotor networks but also in the RN, associated with the recovery of motor function following severe damage to the somatosensory cortex. Impairment of S1 function may have implications for the activity and motor control of M1 [8,9] as well as the ability to learn new motor tasks [10,11].

Despite limitations in motor recovery resulting from impaired sensorimotor integration, the patient exhibited significant improvement in motor function. This improvement was accompanied by hyperactivity in ipsilesional RN, as indicated by the increased FA and decreased

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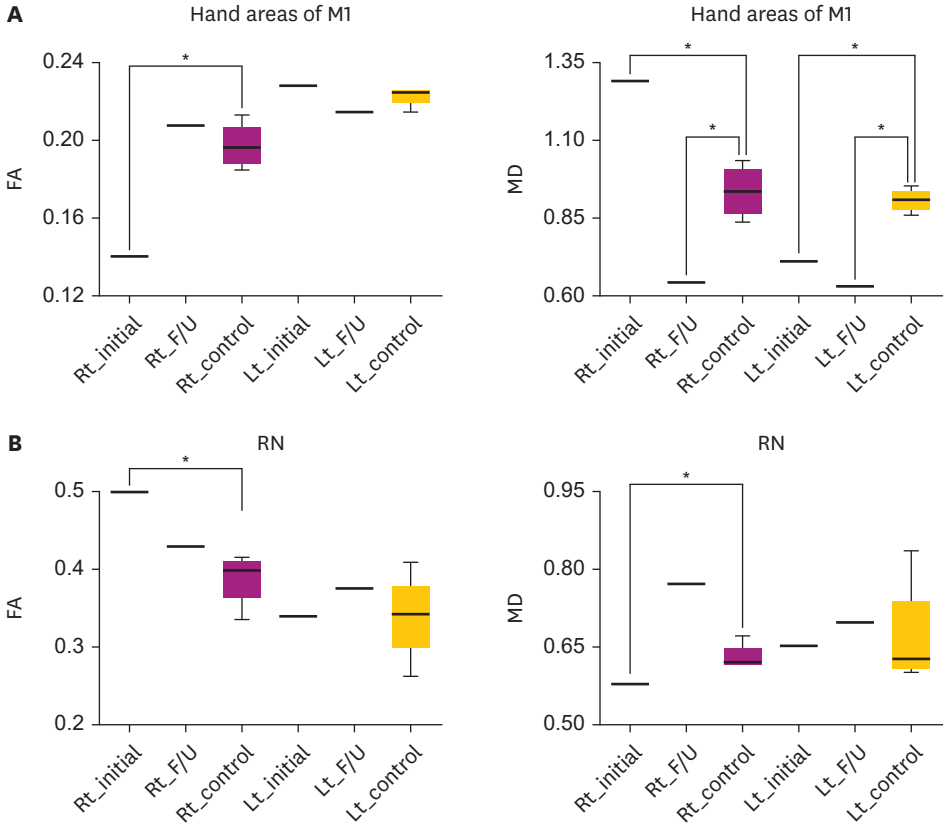

Fig. 2. Diffusion metrics of bilateral hand areas of the M1 and RN. (A) The patient initially had significantly lower FA and higher MD values in the right-hand areas of the M1 compared to the control group. In the F/U diffusion tensor imaging, an increase in FA value and a decrease in MD value were observed in the right-hand areas of the M1. (B) The patient initially had significantly higher FA and lower MD values in the right RN than the control group. After 3 months, the FA value decreased, and the MD value in the right RN increased.
M1, primary motor cortex; RN, red nucleus; FA, fractional anisotropy; MD, mean diffusivity; F/U, follow-up; Rt, right; Lt, left.
*$p < 0.05$.

MD, which was observed within 3 weeks after onset. This hyperactivity normalized when her hand function recovered to a level at which she could perform activities of daily living.

Many studies have reported compensatory increases in the activity of the RN due to damage to corticospinal tract (CST). The mean FA of the RN in the affected hemisphere was higher than in the unaffected hemisphere in DTI performed within 3 weeks after the onset of cerebral infarction [12], and an increase in FA in ipsilesional RN was observed at 3 months after the onset of subcortical stroke [13]. In patient with severe damage to the CST, increased neural connectivity [14] and integrity [15] of RN in the unaffected hemisphere was observed in the chronic stage. In this case study, we report the hyperactivity in ipsilesional RN following damage to S1, and to the best of our knowledge, this unique finding has not been previously reported. These findings are in line with our previous study [15] in which patients with severe CST damage, potentially concurrent S1 damage, had a higher FA value in the RN in the affected hemisphere than controls, although this was not statistically significant. The evidence of RN neurons being activated in response to sensory stimulation in a somatotopically organized way have been reported in cat and primates [16,17]. Therefore, feedback control of the rubrospinal tract efferent through proprioceptive-afferent sensory input to the interposed nucleus of the paravermal cerebellum and magnocellular RN [18-20]

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may be the underlying pathway that induces sensorimotor integration of the motor cortex instead of the damaged S1 in this case.

The patient exhibited involuntary movements in the subacute phase. We consider these movements to be ballistic movements characterized by repetitive and stereotype flinging movements of arms and legs. Several case reports have suggested that lesions in the parietal lobe can induce hemiballism [21-24]. The hypothesis for this phenomenon includes the interruption of neural circuits from the frontal or parietal cortex to the caudate nucleus and putamen [25], disorganization of the basal ganglion–thalamocortical circuit [23], or disorganized sensorimotor integration in the frontoparietal lobes with decreased excitatory output to the subthalamic nucleus [22]. However, these previous studies have reported the occurrence of such movement in the acute phase. Our findings demonstrated that RN hyperactivity may have preceded these movements and subsequent DTI showed that hyperactivity in RN subsided after symptoms disappeared. It indicates the possibility that these movements might be attributed to the RN, and cannot be excluded a form of rubral tremor. Rubral tremor is known to be due to a lesion of the RN [26], but more recently it has been suggested that it may be due to lesions of dopaminergic nigro-striatal projections and cerebellar dento-thalamic fibers [27-30]. However, since the patient's symptoms differ from the typical rubral tremor, cautious interpretation is necessary regarding the possibility. The onset and resolution of these abnormal movements in our patient may be due to the lesion in S1 and a result of improvement or it might also potentially be attributed to the underlying hyperactivity of the RN, which may play a role in restoring damaged sensorimotor integration. In conclusion, this case study delineates the possibility that adaptive changes in the RN response to somatosensory cortex damage may induce sensorimotor integration.

SUPPLEMENTARY MATERIALS

Supplementary Table 1
Fractional anisotropy values of the patient (initial and F/U) and controls

Click here to view

Supplementary Table 2
Mean diffusivity values of the patient (initial and F/U) and controls

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