

# Severe ataxia due to injuries of neural tract detected by diffusion tensor tractography in a patient with pontine hemorrhage

## A case report

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### Abstract

**Rationale:** We examined injuries of the dentato-rubro-thalamic tract (DRTT), cortico-ponto-cerebellar tract (CPCT), dorsal spinocerebellar tract (SCT), and inferior cerebellar peduncle (ICP) in a patient with severe ataxia following bilateral tegmental pontine hemorrhage (PH), using diffusion tensor tractography (DTT).

**Patient concerns:** A 75-year-old female patient underwent conservative management for bilateral tegmental PH. She presented with moderate motor weakness, severe resting and intentional tremor on both hands, and severe truncal ataxia (Scale for Assessment and Rating of Ataxia [25 points/0–40 points: a higher score indicates a worse state]), and she was not able to sit independently.

**Diagnoses and outcomes:** On DTT taken at 2 weeks after initial presentation, both DRTTs and the left dorsal SCT were not reconstructed, whereas the CPCTs showed thinning of the entire pathways between the primary sensorimotor cortex and cerebellum in both hemispheres. The right ICP was discontinued at the transverse cerebellar branch of the ICP and thinning of the left ICP was observed in the vertical and transverse cerebellar branch of the ICP.

**Lessons:** Using DTT, concurrent injuries of the DRTT, CPCT, dorsal SCT, and ICP were demonstrated in a patient with severe ataxia following PH. Our result suggests the necessity of evaluation of these neural tracts in patients who develop ataxia after brain injury.

**Abbreviations:** CPCT = cortico-ponto-cerebellar tract, DRTT = dentato-rubro-thalamic tract, DTI = diffusion tensor imaging, DTT = diffusion tensor tractography, FAC = functional ambulation category, ICP = inferior cerebellar peduncle, PH = pontine hemorrhage, ROI = regions of interest, SARA = Scale for Assessment and Rating of Ataxia, SCT = *spinocerebellar tract*.

**Keywords:** ataxia, cortico-ponto-cerebellar tract, dentato-rubro-thalamic tract, inferior cerebellar peduncle, pontine hemorrhage, spinocerebellar tract

## 1. Introduction

Approximately 5% to 10% of intracerebral hemorrhages are pontine hemorrhage (PH), accompanied by neurological symp-

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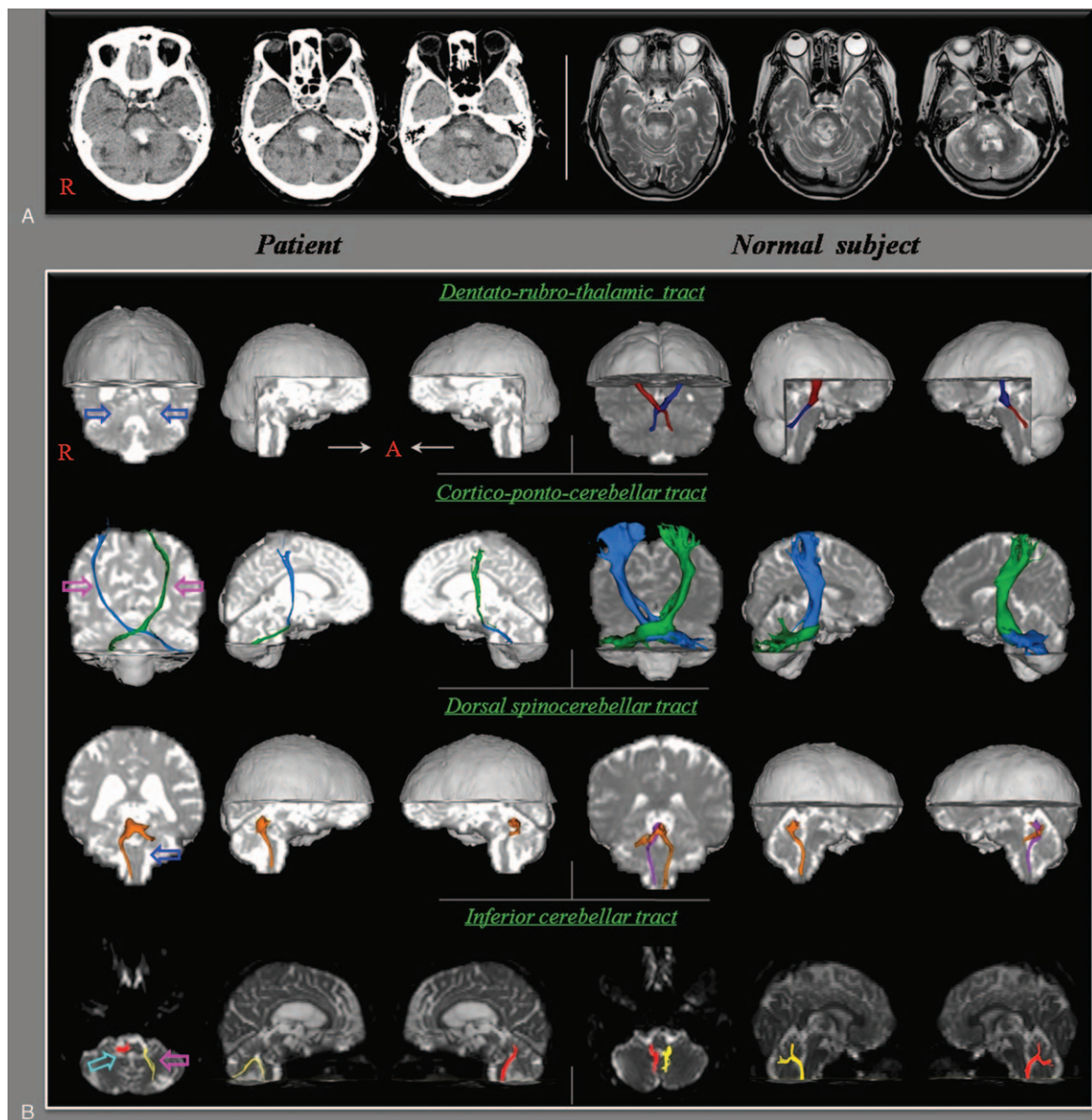
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toms including ataxia, dysarthria, abnormality of eye movements, and vertigo.<sup>[1–3]</sup> Ataxia frequently occurs after PH because neural tracts related to ataxia including the dentato-rubro-thalamic tract (DRTT), cortico-ponto-cerebellar tract (CPCT), ventral and dorsal spinocerebellar tracts (SCTs), and inferior cerebellar peduncle (ICP) pass the pons.<sup>[4,5]</sup> Given this, the diagnosis of ataxia should include the examination of those neural tracts. However, research on these neural tracts is difficult in the live human brain as neural tracts are small, long, multi-synaptic, or decussating fibers to the opposite hemisphere.<sup>[4]</sup> Recently developed diffusion tensor tractography (DTT), derived from diffusion tensor imaging (DTI), enables 3-dimensional visualization and estimation of most such neural tracts.<sup>[6–9]</sup> Many studies have reported on injuries or recovery of neural tracts related to ataxia in patients with brain injury using DTT; however, no study in patients with PH has been reported so far.<sup>[10–21]</sup>

In this study, using DTT, we examined injuries of the DRTT, CPCT, dorsal SCT, and ICP in a patient with severe ataxia following bilateral tegmental PH.

## 2. Case report

A 75-year-old female patient underwent conservative management for bilateral tegmental PH at the department of neurosurgery of a university hospital (Fig. 1A). At 2 weeks after her



**Figure 1.** (A) Brain CT images show hematomas in the bilateral tegmental pons, and T2-weighted brain images at 2 weeks after initial injury show leukomalactic lesions in the bilateral tegmental pons. (B) Results of diffusion tensor tractography. The dentato-rubro-thalamic tracts are not reconstructed in both hemispheres (blue arrows), whereas the cortico-ponto-cerebellar tracts show thinning of the entire pathways between the primary sensorimotor cortex and cerebellum in both hemispheres (purple arrows), and the dorsal spinocerebellar tract is not reconstructed in the left hemisphere (blue arrow). The right inferior cerebellar peduncle (ICP) is discontinued at the transverse cerebellar branch of the ICP (sky-blue arrow) and thinning of the left ICP is observed in the vertical and transverse cerebellar branch of the ICP (purple arrow). CT = computed tomography, ICP = inferior cerebellar peduncle.

neurologic injury, she was transferred to the rehabilitation department of the same university hospital to undergo rehabilitation. Her neurological state was examined by a physiatrist and the patient showed normal reflex in biceps, triceps, knee, and ankle of both sides. She had no spasticity, flaccid, or ocular symptom including diplopia, ptosis, and nystagmus. However, she presented with moderate motor weakness (manual muscle test—upper/lower extremities: right side—fair/fair grade, left side—fair/fair grade), severe resting and intentional tremor on both hands, and severe truncal ataxia due to the bilateral tegmental PH. Brain MR images taken 2 weeks after her initial PH showed leukomalactic lesions in the bilateral tegmental pons (Fig. 1A). The Scale for Assessment and Rating of Ataxia (SARA, 0–40 points: a higher score indicates a worse state) and the Functional

Ambulation Category (FAC, 5 points: a lower score indicates a worse state) were measured.<sup>[22,23]</sup> The patient scored 25 points on the SARA and 1 point on the FAC, and she was unable to sit independently. The patient provided written informed consent, and the study protocol was approved by the Yeungnam University Hospital Institutional Research Board.

DTI data were acquired at 2 weeks after her initial injury using a 1.5T Philips Gyroscan Intera (Philips, Ltd, Best, the Netherlands) with 32 gradients. Imaging parameters were as follows: acquisition matrix =  $96 \times 96$ ; reconstructed to matrix =  $192 \times 192$ ; field of view =  $240 \times 240 \text{ mm}^2$ ; repetition time = 10,398 ms; echo time = 72 ms;  $b = 1000 \text{ s/mm}^2$ ; and a slice thickness of 2.5 mm. Scanning was performed from the cortex to the middle of the second cervical vertebral body. Prior to the fiber

tracking, eddy current correction was applied for correction of the head motion effect and image distortion using the Oxford Centre for Functional Magnetic Resonance Imaging of the Brain (FMRIB) Software Library.<sup>[24]</sup> For fiber tracking, we used the FMRIB Software Library based on probability tracking for DRTT CPCT, and dorsal SCT with a result threshold of 2 streamlines and DTI-Studio software based on deterministic tracking for ICP with a fractional anisotropy of 0.2 and a tract turning-angle of < 60 degrees.<sup>[6–9,24,25]</sup> DRTT, CPCT, dorsal SCT, and ICP were analyzed for evaluation of state of neural tracts in the brain using regions of interest (ROIs) as follows<sup>[6–9]</sup>: (1) DRTT: ROI 1—dentate nucleus on the coronal image, ROI 2—junction of the superior cerebellar peduncle on the coronal image, and ROI 3—contralateral red nucleus of the upper midbrain on the axial image<sup>[6]</sup>; (2) CPCT: ROI 1—primary sensorimotor cortex on the axial image, ROI 2—anterior pons on the axial image, and ROI 3—contralateral cerebellar hemisphere on the coronal image<sup>[7]</sup>; (3) dorsal SCT: ROI 1—dorsolateral funiculus of the upper cervical cord on the axial image, ROI 2—the restiform body on the axial image, and ROI 3—the vermis of the anterior lobe in the cerebellum on the axial image<sup>[8]</sup>; (4) ICP: ROI 1—the restiform body on the axial image, and ROI 2—the caudal part of the superior cerebellar peduncle on the axial image.<sup>[9]</sup>

On 2-week DTT, both DRTTs were not reconstructed, whereas the CPCTs showed thinning of the entire pathways between the primary sensorimotor cortex and cerebellum in both hemispheres, and the dorsal SCT was not reconstructed in the left hemisphere (Fig. 1B). The right ICP was discontinued at the transverse cerebellar branch of the ICP and thinning of the left ICP was observed in the vertical and transverse cerebellar branch of the ICP (Fig. 1B).

### 3. Discussion

In this case report, concurrent injuries of the DRTT, CPCT, dorsal SCT, and ICP were demonstrated in a patient with severe ataxia following bilateral tegmental PH. Marx et al's<sup>[5]</sup> study described the association of 5 neural tracts (DRTT, CPCT, ventral and dorsal SCTs, and ICP) with ataxia in 49 patients with lesions of the brainstem using 3-dimensional mapping. Hence, the DRTT, CPCT, dorsal SCT, and ICP, except for the ventral SCT, for which the reconstruction method by DTT has not been devised, were reconstructed. Our results can be summarized as follows: (1) both DRTTs and the left dorsal SCT were not reconstructed, indicating severe injury; (2) partial injury of both CPCTs (thinning) and both ICPs (right: discontinuation, left: thinning) was observed. The dorsal SCT and ICP mainly receives the proprioceptive information and carries it to the cerebellum, and then the DRTT and CPCT transfer the information on movement between the cerebellum and cerebrum via pons.<sup>[4,5,26,27]</sup> Therefore, it appears that the severe ataxia in this patient was at least in part attributable to injuries of the DRTT, CPCT, dorsal SCT, and ICP.

Injuries of neural tracts, diagnosed with DTT, related to ataxia are common with brain pathologies including stroke, brain tumor, and traumatic brain injury.<sup>[10–15]</sup> Hong et al<sup>[10]</sup> and Kwon and Jang<sup>[11]</sup> reported injury of the ICP in patients with ataxia following traumatic brain injury.<sup>[10,11]</sup> In 2015, Marek et al<sup>[12]</sup> found injury of the cerebello-thalamic portion of the DRTT in 6 patients (demyelination: 2 patients, ischemia: 2 patients, hemorrhage: 1 patient and neoplasm: 1 patient) with cerebellar dysfunction including ataxia and tremor.

Schulz et al<sup>[14]</sup> reported association of injuries of the DRTT and CPCT with motor output including fine motor skill in 26 patients with chronic ischemic stroke. During the same year, Jang and Kwon<sup>[15]</sup> reported that a patient's tremor and ataxia that started 2 weeks after mild traumatic brain injury was caused by injury of the DRTT in the right hemisphere following mild traumatic brain injury. In this study, severe ataxia in a patient with PH was ascribed to concurrent injuries of the DRTT, CPCT, dorsal SCT, and ICP. Therefore, to the best of our knowledge, this is the first study to demonstrate concurrent injuries of the DRTT, CPCT, dorsal SCT, and ICP in a patient with brain injury. Because it is a single case report, this study is limited. In addition, other limitations of this study should be considered. First, we could not examine associations between the severity of ataxia and each neural tract. Second, we could not evaluate the ventral SCT due to a limitation of the current DTT technique. Third, use of DTT could lead to both false positive and negative results due to multiple fiber orientations in a voxel.<sup>[28]</sup> Therefore, we suggest that further studies including large numbers of patients and on the prognosis (regeneration or degeneration) or feature of injured neural tracts related to ataxia should be encouraged.

In conclusion, using DTT, concurrent injuries of the DRTT, CPCT, dorsal SCT, and ICP were demonstrated in a patient who developed severe ataxia following PH. Our result suggests the necessity of evaluation of these neural tracts in patients who develop ataxia after brain injury and can provide how to evaluate the patients with ataxia in terms of neural tracts for clinicians and researchers in treatment or studies of the ataxia using DTT.

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