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Case Report

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Ipsilateral Motor Evoked Potentials in a Preschool-age Child With Traumatic Brain Injury: A Case Report

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HIGHLIGHTS

- The upper age limit at which post-neonatal cerebral palsy manifests remains unclear.
- We report ipsilateral motor evoked potential (iMEP) caused by brain injury at age 4.
- This case is the oldest in which brain injury occurred and iMEP was maintained.
- iMEP is a valuable indicator of motor system plasticity in the developing brain.

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Ipsilateral Motor Evoked Potentials in a Preschool-age Child With Traumatic Brain Injury: A Case Report

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ABSTRACT

To the best of our knowledge, the upper age limit at which post-neonatal cerebral palsy (CP) can manifest remains uncertain. This uncertainty is attributed to the lack of objective parameters for assessing the developing brain. In a previous study, we reported that an ipsilateral corticospinal projection associated with brain injury, as manifested in the paretic hand of a CP patient, had never been observed in individuals aged > 2 years. In this case report, we present an instance of ipsilateral motor evoked potential (iMEP) in a girl whose traumatic brain injury occurred at the age of 4 years. This case is the oldest in which brain injury occurred and iMEP was maintained. In conclusion, iMEP can be a valuable indicator of motor system plasticity in the developing brain.

Keywords: Motor Evoked Potentials; Traumatic Brain Injury; Transcranial Magnetic Stimulation; Brain Plasticity; Case Report

INTRODUCTION

While there is a general consensus that the lower age limit of postneonatal cerebral palsy (CP) is the 28th day after birth, the upper age limit of post-neonatal CP is uncertain [1,2]. In a recently reported postneonatal CP surveillance, the upper age limit of CP was at the age of 2 years [3]. The uncertainty of this upper age limit is attributed to the lack of objective parameters associated with the developing brain.

In previous studies on CP using transcranial magnetic stimulation (TMS), short latency ipsilateral motor evoked potential (iMEP) from the unaffected hemisphere manifesting in the intrinsic muscles of the paretic hand was not reported for lesions acquired after the age of 2 years or in adults post-stroke. Rather, iMEP was only observed in cases of prenatal and perinatal brain lesions [4]. Thus, iMEP in the hand's intrinsic muscles could be elicited under specific pathologic conditions during early development. Therefore, we suggest that iMEP could be considered as an electrophysiological marker of brain plasticity and that it might be used as an objective parameter for determining the upper age limit of early brain lesions.

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Conflict of Interest
The authors have no potential conflicts of interest to disclose.

Author Contributions
Conceptualization: Kim SY, Park SH. Methodology: Kim SY, Park SH. Formal analysis: Kim SY, Park SH. Project administration: Kim DS, Kim GW, Won YH, Ko MH, Seo JH. Visualization: Roh CH, Park SH. Writing - original draft: Kim SY, Park SH. Writing - review & editing: Kim SY, Roh CH, Kim GW, Won YH, Ko MH, Seo JH, Park SH.

Here, we present a case of observed ipsilateral corticospinal projections in a girl whose traumatic brain injury (TBI) occurred at the age of 4 years. To our knowledge, this patient was the oldest in whom a brain injury was associated with a short latency iMEP. The consistency and stability of the iMEP were maintained over a long time period.

CASE DESCRIPTION

A 13-year-old female patient presented with right hemiparesis due to TBI at 4 years of age. TBI was caused by a traffic accident, and intracerebral hemorrhage (ICH) occurred in the patient's right frontal lobe and left basal ganglia. Prenatal and postnatal histories were uneventful. Developmental milestones were normal before the onset of TBI. She underwent conservative management for traumatic ICH and received rehabilitation therapy over a short period before being discharged to her home. About 9 years after discharge, she revisited our hospital for further rehabilitation treatment.

Neurological examination revealed weakness in the right wrist and hand; in the Medical Research Council scales, her strength was 3/5. The right wrist and hand had I+ and II spasticity on the Modified Ashworth Scale, respectively. The degree of motor disturbance in the right lower extremity was relatively mild, and walking was possible. The strength of the right ankle dorsiflexion and plantarflexion was 3/5, and the spasticity of the right ankle was grade I+. The grip strengths of the patient (Jamar Hydraulic Hand Dynamometer; Jamar Corp., Duluth, MN, USA) were 3.5 kg (right) and 19.5 kg (left). The dexterity was measured using a 9-hole pegboard (Sammons Preston, Mississauga, ON, Canada) and was defined as the time it took to insert and remove 9 headless pegs into the holes of a 5-inch square pegboard [5]. She needed 24 seconds for the left hand but could not perform the same procedure with the paretic right hand. The performance of mirror movements was not possible owing to spastic fisting of the right hand. While the left hand grasped and released voluntarily, the spasticity of the right hand increased. Informed consent was obtained from the patient to publish this case, and approval for this study was provided by the Institutional Review Board (IRB) of Jeonbuk National University Hospital (IRB No. 2021-07-027).

Conventional magnetic resonance imaging (MRI)

Conventional MRI of the brain was performed when our patient was 13 and 16 years old. There were no significant changes observed between the 2 time points. The resulting images showed that a contusion was observed in the left lentiform nucleus and the contralateral right orbitofrontal area.

The right arm-dominated hemiparesis corresponded with the putamen and internal capsular lesions in the left hemisphere. The left crus cerebri was smaller than the right, which indicated corticospinal atrophy in the left hemisphere (**Fig. 1**).

Diffusion tensor imaging (DTI)

A DTI study was conducted when the patient was 16 years old. The equipment, program, technique, and imaging parameters of DTI were the same as those in our previous papers [6,7]. The corticospinal tract (CST) was reconstructed to visualize the motor fibers by selecting regions of interest (ROIs). The seed ROI was drawn at the CST portion of the anterior mid-pons. To assess the CST, the number of fibers, fractional anisotropy (FA), and apparent diffusion coefficient (ADC) values at the mid-pons were measured (**Table 1**). The

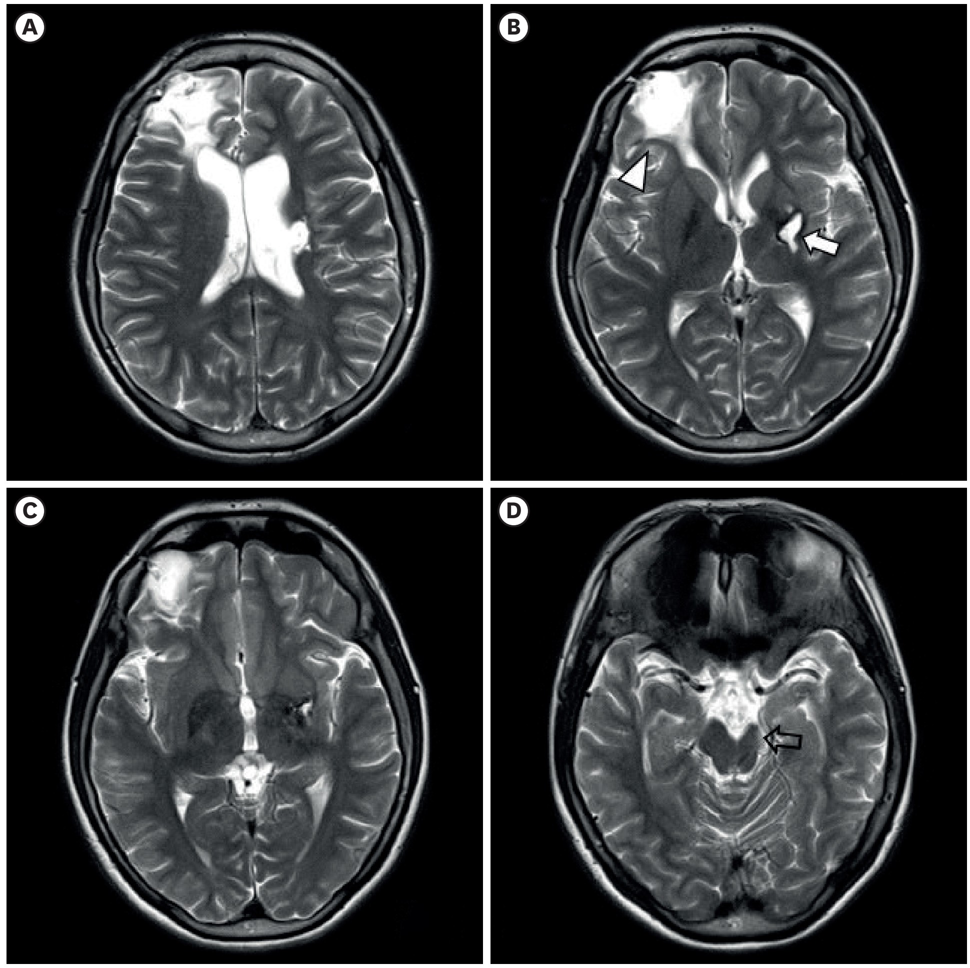

Fig. 1. The axial T2-weighted images at the level of the (A) corona radiata, (B, C) basal ganglia, and (D) midbrain, showing the contusion in the left lentiform nucleus (arrow on B) and contralateral right orbitofrontal area (arrowhead on B). The left crus cerebri was smaller than the right, which was indicated by corticospinal atrophy in the left hemisphere, compatible with right side hemiplegia (void arrow on D).

DTI axial color map showed that the CST from the left cerebral hemisphere was shrunken and disrupted, indicating a possible injury to the CST. When comparing the left and right CST at the midbrain, there was significant asymmetry of the cross-sectional area (**Fig. 2**).

TMS

The equipment, stimulation technique, and analysis method of TMS were identical to those reported in our previous papers [6-8]. TMS was performed on patient at ages 13, 16, and 18 years (**Table 2**, **Fig. 3**). The optimal stimulation position was (−4, 0) for the left cortex and (4, 0) for the right cortex according to the 10–20 system, while the threshold for the contralateral MEPs was 45% and that for the iMEPs was 60%.

Table 1. Number of fibers, FA, and ADC values of the corticospinal tract

Variable	No. of fibers		FA		ADC	
	Right	Left	Right	Left	Right	Left
CST	404	167	0.63 ± 0.18	0.53 ± 0.03	0.74 ± 0.41	0.86 ± 0.45

Values are presented as means ± standard deviations.
FA, fractional anisotropy; ADC, apparent diffusion coefficient; CST, corticospinal tract.

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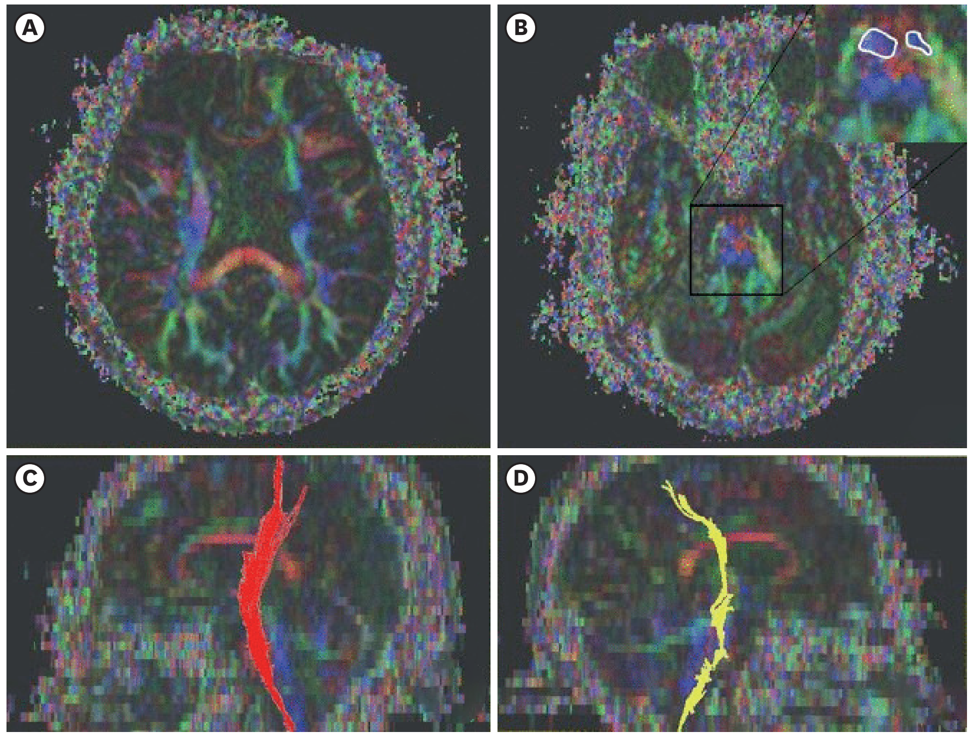

Fig. 2. Axial color-coded fractional anisotropy map demonstrating well-defined CST (blue) at the level of the cerebral cortex (A) and pons (B). The cross-sectional area of the CST at the level of the pons estimated with diffusion tensor imaging. (B). Our patient featured symmetry indices of 0.21. Right CST in the sagittal section (C). Left CST in the sagittal section (D).
CST, corticospinal tract.

Table 2. Transcranial magnetic stimulation at the ages of 13, 16, and 18 years

Age (yr)	Stimulation/Recording	Latency (ms)		Amplitude (uV)	
		Right FDI	Left FDI	Right FDI	Left FDI
13	Left cortex	20.2 ± 0.2	Not evoked	248.0 ± 19.1	Not evoked
	Right cortex	22.4 ± 0.7	20.1 ± 0.1	149.3 ± 33.2	200.0 ± 29.4
16	Left cortex	22.5 ± 0.0	Not evoked	212.2 ± 32.8	Not evoked
	Right cortex	22.1 ± 0.4	20.6 ± 0.6	183.3 ± 19.2	104.0 ± 25.9
18	Left cortex	22.7 ± 0.3	Not evoked	167.8 ± 26.3	Not evoked
	Right cortex	22.5 ± 0.0	21.3 ± 0.7	246.9 ± 87.0	162.5 ± 12.5

Values are presented as means ± standard deviations.
FDI, first dorsal interosseous.

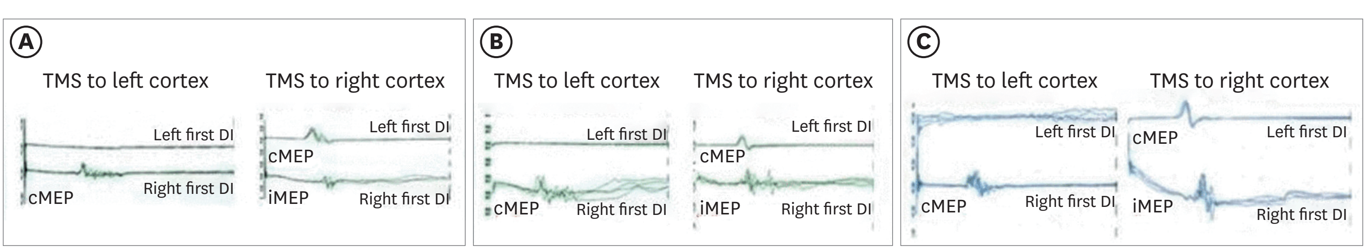

Fig. 3. TMS at the age of 13 (A), 16 (B), and 18 (C) years. The right and left first dorsal interosseous motor evoked potential responses to the transmagnetic stimulation of the left primary motor cortex and right primary motor cortex.
TMS, transcranial magnetic stimulation; cMEP, contralateral motor evoked potentials.

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DISCUSSION

We present a case of iMEP in a woman whose TBI occurred at the age of 4 years. To our knowledge, this patient was the oldest in whom a brain injury was associated with a short latency iMEP and in whom a consistent and stable iMEP was maintained over a long time period [9]. Therefore, iMEP can be a valuable indicator of motor system plasticity in the developing brain and present a new perspective on the upper age limit at which CP can occur [10].

Especially, iMEP in the intrinsic muscles of the hand may be a neurophysiological marker for developing brain plasticity and can be used as an objective parameter for determining the upper age limit of early brain lesions. In a previous MEP study of 20 patients with hemiplegia, iMEP was not observed in patients aged > 2 years [11]. However, in our case, short latency iMEP was observed at the age of 4 years in the relaxed paretic hand intrinsic muscles and elicited reproducibly. We assumed that iMEP observed in the developing brain has the following characteristics: (a) a short latency, (b) elicitation in the hand intrinsic muscles, and (c) elicitation during relaxation of the target muscle [6,7]. iMEP with these characteristics differs from long latency iMEP found in patients with stroke who had acquired unilateral cortical lesions in adulthood [12].

Previously, we reported long latency MEPs observed in paretic and non-paretic hands, occurring much later than those commonly reported [13]. In that paper, the average age of patients with congenital hemiplegia was 13.3 years, and iMEP was observed in both hands. One study has argued that the presence of ipsilateral corticospinal connections during the first decade of life appears to be a normal state in ontogeny [14]. iMEP was elicited through voluntary preconditioning of the target muscles and was observed more often in the proximal than in the distal muscles. Another study argued that in neonates, ipsilateral responses were obtained during spontaneous muscle contraction and were not elicited reproducibly by 18 months [15]. Interestingly, corticospinal ipsilateral projections were elicited reproducibly in our patient during relaxation in the hand intrinsic muscle, under conditions different from those reported in previous papers. iMEP during relaxation in the hand intrinsic muscle has not previously been observed in healthy children aged ≥ 2 years [8] or in patients with hemiplegia aged > 2 years [11]. Other brain damage, such as stroke, may cause ipsilateral projection, but other studies have reported that it was hard to elicit the motor response in affected muscles in patients with stroke during selective stimulation of the ipsilateral motor cortex [16].

DTI was conducted to visualize the CST and determine the FA and ADC values and the number of fibers. The DTI index reportedly correlates with the conventional MRI index in children with hemiplegia and provides a useful prognostic tool for anticipating upper-limb deficits [6]. Although the bilateral connectivity of the CST extended to the primary motor cortex, we found fewer CST fibers and a smaller cross-sectional area of the CST at the level of the pons on the left than on the right side. These DTI findings revealed a decreased CST tract volume. Visually, it is apparent that the integrity of the CST of the left side was reduced compared to that of the right side. However, it is noteworthy that the CST maintained connectivity to the primary motor cortex, possibly resulting in our patient's paretic hand motion.

In our patient, corticospinal (re)organization occurred relatively late in development, given the prenatal nature of the injuries. Therefore, our patient's motor function recovery was poor, and fine mirror movement was not observed [17]. In a previous study, children with extensive unilateral early brain lesions but relatively preserved hand functions consistently showed evidence for ipsilateral, fast-conducting cortico-spinal pathways allowing the contralesional hemisphere to exert motor control over the paretic hand [6]. Corticospinal (re)organization occurs very early in development, and abnormal ipsilateral projections can establish appropriate linkages with the cortical and subcortical networks required for effective arm and hand control [6]. However, when the (re)organization occurs relatively late in development, the abnormal ipsilateral projections cannot access these networks [4], as was the case with our patient. Although our patient had a bilateral hemisphere injury, corticospinal reorganization occurred in the less damaged hemisphere. This type of case has been reported in a previous study [7].

This study had several limitations. First, we were unable to perform the TMS study at the age of 4 years when her brain injury occurred. Second, there are limitations associated with using DTI to show ipsilateral corticospinal projections.

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