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Abnormal somatosensory evoked potentials in a child with motor conversion disorder: A case report

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A 12-year-old female child complained of bilaterally limited lower limb activity for 4 days before admission to the pediatric neurology department. She could not walk alone, but she could maintain a squatting posture for 10 min when using the toilet and was also observed to raise her legs inadvertently. She had suffered trauma to her right hand at 4 years old, and the activity of the hand remained slightly limited as of the current admission. The girl's parents both had mental retardation; therefore, she was in the care of her grandfather and had little contact with her parents. Before the onset of the disease, she had been required to start school, at which time her stress level increased.

The patient had restricted movement of the joints in both lower extremities, with normal touch, blunted pain, and significant lower limb weakness (muscle strength I–II). Upper limb muscle tone and strength were normal. There were no sensory deficits. The abdominal wall reflex was normal. The Achilles tendon reflex and the knee reflex were absent. Pathological reflexes were absent. No other pathological results were found on examination.

Blood work (routine blood test, liver and kidney function, cardiac biomarkers, muscle enzymes, coagulation test, ammonia test, pyruvate-blood test, lactic acid) showed normally, and viral markers (IgM antibodies to enterovirus, Epstein–Barr virus, coxsackievirus, adenovirus, cytomegalovirus, rubella virus, simple Herpes virus) were negative. Plain and enhanced spinal magnetic resonance imaging (MRI) showed no abnormalities. Nerve conduction in the lower limbs was normal. Somatosensory evoked potential (SSEP) detection was first performed on the third day after disease onset in our hospital outpatient clinic. The cathode was located between the medial malleolus and the Achilles tendon, where stimulation caused big toe abduction. The anode was placed 3 cm distal to the cathode and delivered constant current pulses of 100 msec, applied at 3 Hz, with a stimulus intensity of 8 mA; the data were sampled every 100 msec for analysis, and each run represented the average of two hundred stimuli. Cortical waves (P40) were recorded over the scalp at Cz and referenced to Fz. Spinal waves [cauda equina (CE)] were recorded over the L5 spinous process and popliteal fossa (PF) waves were recorded

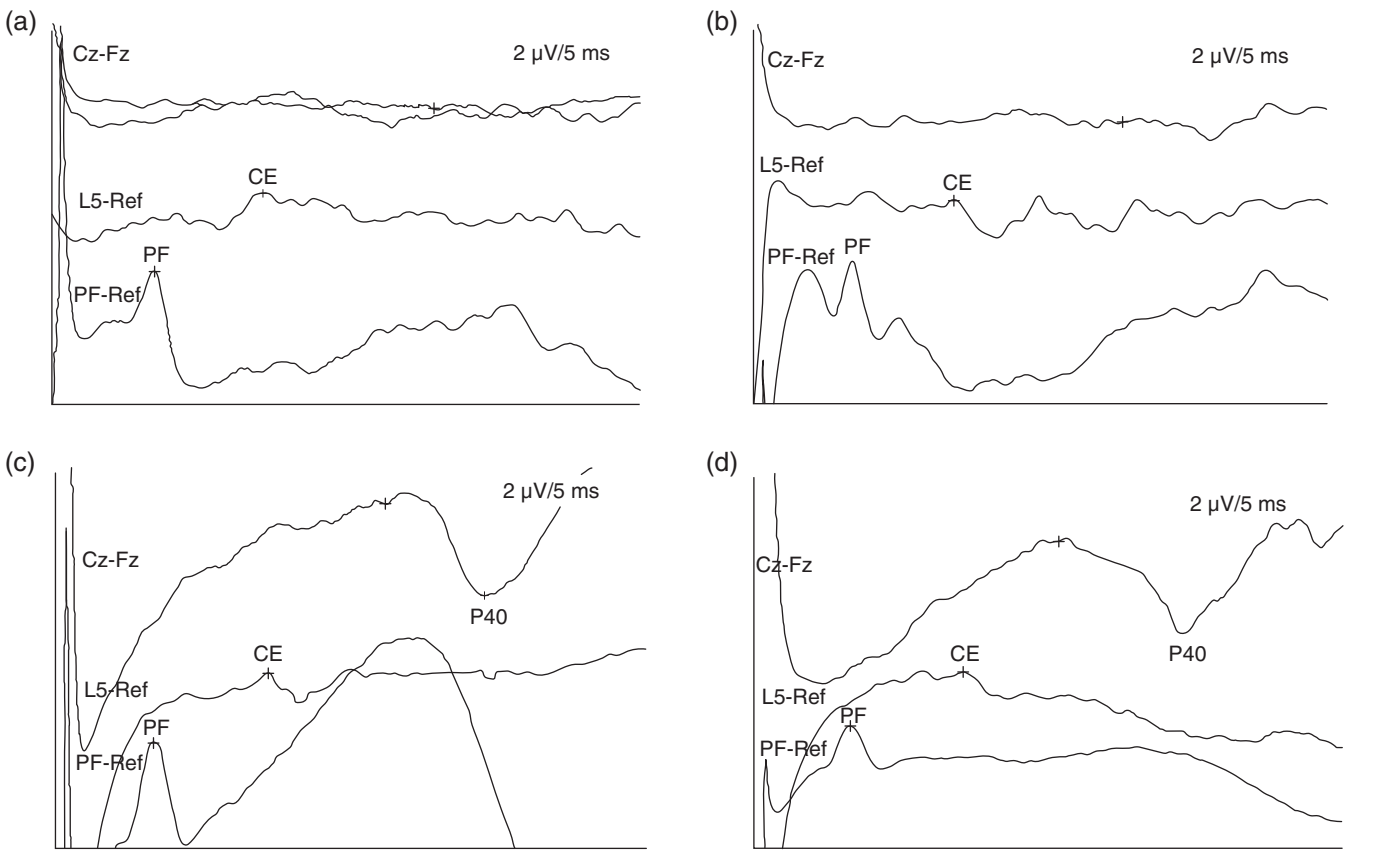

Fig1 Bilateral posterior tibial nerve somatosensory evoked potential (SSEP). (a) SSEP lower limb stimulate right tibial (3rd day of the disease course). (b) SSEP lower limb stimulate left tibial (3rd day of the disease course). (c) SSEP lower limb stimulate right tibial (11th day of the disease course). (d) SSEP lower limb stimulate left tibial (11th day of the disease course). CE, cauda equina; PF, popliteal fossa; L5, L5 spinous process; Ref, reference.

over posterior tibial nerve at PF, referenced to another surface electrode placed 3–4 cm away. In order to ensure that the results were correct, each recording was repeated more than two times. We used a Nihon Kohden Neuropack MEB-9200K for recording. When the bilateral tibial nerves were stimulated, cortical P40 waves were bilaterally absent, and bilateral CE waves and PF waves with normal latencies (17.9 ms for the right tibial nerve CE, 17.5 ms for the left tibial nerve CE, 8.75 ms for the right tibial nerve PF, 8.75 ms for the left tibial nerve PF) were elicited (Fig. 1a,b).

A neurological and psychiatric joint consultation arrived at a diagnosis of motor conversion disorder which might be triggered by anxiety regarding starting school. After 4 days of hospitalization with psychological counseling and B vitamin supplementation, she gradually walked. On the 11th day of the disease course, she could walk on her own. Then, we re-examined SSEPs. Bilateral cortical P40 waves, CE waves, and PF waves were normal latencies (36.4 ms for the right tibial nerve P40, 36.4 ms for the left tibial nerve P40, 18 ms for the right tibial nerve CE, 17.9 ms for the left tibial nerve CE, 8.35 ms for the right tibial nerve PF, 8.25 ms for the left tibial nerve PF) (Fig. 1c,d). At a 7-month telephone follow-up, the patient was attending school and she had no physical symptoms such as limb weakness.

We reviewed the literature on conversion disorder patients with SSEP abnormalities[1–4] and our case. The present case and the reviewed cases indicate functional neurological disorder may have abnormal SSEP and provide some clues regarding the mechanism of conversion disorder. We speculated that the possible mechanism of the loss of cortical SSEP was the same as Yazici *et al.*[4] and Vuilleumier's,[5] indicating that top-down inhibition works at the thalamus level. But the case had two limitations; first, she did not undergo brain MRI, and second, vitamin B level testing was not available in our hospital and vitamin B deficiency could be considered a differential because she was treated with supplementation. SSEP testing is less expensive and easier to perform than neuroimaging. Further studies on SSEP may clarify the neurobiological markers of conversion disorder and provide an improved reference for diagnosis and treatment.

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Disclosure statement

The authors have no competing interests to declare.

Ethics approval and consent to participate

The ethics committee of West China Second University Hospital judged that there was no need to review this case.

Consent for publication

Written informed consent was obtained via her grandfather's oral consent for the publication of this case report.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

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Aripiprazole as a new treatment for the prolonged nocturnal sleep of patient with idiopathic hypersomnia

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In the latest journal, Honda *et al.*[1] proposed the new method for diagnostic examination of 24 h polysomnography (PSG) for idiopathic hypersomnia, identified by Bedric Roth in 1976.[2] These patients had marked sleepiness, often with prolonged nocturnal sleep but without clinical or electrophysiological features of REM sleep disturbance,[1,2] reported as chronic with a poor response to treatment.[3,4]

EDS of idiopathic hypersomnia was occasionally treated by stimulants such as modafinil and amphetamine analogs with some effectiveness.[3] However, it is more difficult to reduce nocturnal sleep time than to decrease daytime sleepiness with medications in the patients with idiopathic hypersomnia with long sleep time. The efficacy of aripiprazole was reported in the patients with delayed sleep phase syndrome and non-24 sleep wake disorders,[5–8] which did not phase advance but reduced nocturnal sleep duration possibly *via* dopaminergic activation.[8]

Here, we described a case with idiopathic hypersomnia with long sleep time who had been treated with low dose aripiprazole with reduction of prolonged nocturnal sleep time. The patient gave written informed consent. This study was approved by Ethics Committee of Akita University Graduate School of Medicine, conducted with clinical trial registration: UMIN-CTR Clinical Trial, R000033455.

The patient was a 15-year-old female high school student. She had been treated for orthostatic disturbance when she was a primary school student. At the beginning of junior high school, her nocturnal sleep time was 9 h, from 22:00 to 07:00. Thereafter, her sleep time gradually extended to 14 h, from 22:00 to 12 PM (Fig. 1a). After the family woke her up in the morning, she showed prolonged difficulty waking up with sleep drunkenness. However, Epworth sleepiness scale (ESS) was only seven points in this condition (11 points or more is the cutline for hypersomnolence).

The PSG showed long sleep time, short sleep latency, high sleep efficiency and normal sleep structures with no indication of sleep-disordered breathing and periodic leg movements (Fig. 1b). Results from the MSLT were shortened sleep onset latencies without SOREMPs. Brain