Rare disease

Effect of vibration stimulation on dysbasia of spastic paraplegia in neuromyelitis optica: a possible example of neuronal plasticity

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Summary

We analysed the effect of vibration stimulation (VS) on dysbasia of neuromyelitis optica (NMO). The patient was a 36-year-old woman who was diagnosed with NMO and had difficulties in walking. VS was applied to the lower limb muscles on the left, more spastic, side with an ordinary vibrator. The performance of standing up and walking improved with VS. Even with improved performance after VS, the amount of surface EMG of the lower limbs did not increase as a whole, but the EMG patterns among the lower leg muscles changed remarkably. VS produced reciprocity within antagonistic muscles. Variability of EMG amplitudes decreased remarkably during the walking cycle, not only on the vibrated side, but also on the non-vibrated side. The effect lasted longer than several dozen minutes after the cessation of VS. We conjectured that central pattern generator (CPG) and neuronal plasticity were the result of VS.

BACKGROUND

Neuromyelitis optica (NMO) is characterised by mostly severe attacks on the optic nerves and the spinal cord. NMO was regarded as a variant of multiple sclerosis (MS); however, recent evidence points to a distinct pathogenesis from MS. Walking disturbance is seen as a cardinal symptom of spinal cord lesion, causing severe disability. In terms of rehabilitation, vibration stimulation (VS) is used for several purposes, such as reducing spasticity and enhancing the strength of weak muscles.^{1–2} It is known that muscle stretch receptors are highly responsive to VS. Increased sensory inputs by VS may modulate the spinal circuit. Hagbarth *et al* first discovered that if human skeletal muscles are mechanically vibrated for some time, tonic contraction of the stimulated muscles is observed, whereas the antagonistic muscles are relaxed. This is referred to as tonic vibration reflex (TVR), and many studies have been conducted on healthy subjects as well as subjects with various neurological disorders.^{3–5} We present a case report of an NMO patient with dysbasia, who improved with VS to the lower limb muscles. We discuss the plausible mechanisms of the beneficial effect of VS.

CASE PRESENTATION

A 36-year-old woman, once diagnosed as having optic neuritis, repeatedly suffered visual disturbance as well as muscle weakness in both lower limbs, urination disorder and girdle sensation in the chest. Flexion of her neck provoked a lancinating pain that ran down to the lower limbs. She was admitted to our university hospital. On neurological examination, there was no abnormality in the cranial nerves, except for signs related to the right optic nerve. She showed difficulties in both standing and walking. In the Manual Muscle Test (MMT) of the lower limbs, the proximal muscles were 1/1–0 (right/left),

whereas the distal muscles were 4–3/3, and the muscle tone of the lower proximal leg muscles, particularly on the left side, was hypotonic. Power and muscle tone of the upper limbs were normal. Deep tendon reflex was absent in bilateral lower extremities. The heel–knee test was not possible. The patient complained of pain in the lower limbs, and showed hyperaesthesia to light touch. There was also decreased vibratory sense in the lower limbs. Neck flexion induced signs of Lhermitte and she had grasping pain in both gastrocnemii. She had urinary retention because of flaccid bladder.

INVESTIGATIONS

In immunological tests, serum antiaquaporin 4 antibody (AQP-4) was positive. Cerebrospinal fluid (CSF) analysis showed an increase in both CSF protein (181 mg/dl) and CSF cell count (152 cells/ μ l), while the oligoclonal IgG band was negative, and myelin basic protein was 691 pg/ml. There were high signal lesions extending over three vertebrae (C1–C6 and Th8–Th10 levels) in T2-weighted spinal MRI. She was diagnosed as NMO, and so steroid pulse therapy and apheresis were performed.

OUTCOME AND FOLLOW-UP

After the treatment, her right visual acuity and lower limb weakness showed amelioration. The muscle tone of the lower limbs altered gradually from flaccid to spastic and deep tendon reflexes of the lower limbs became exaggerated at 10 days after admission. Physiotherapy was started from the early phase of admission and the patient gradually showed improvement in walking. During physical training, it was noticed that motor performance was clearly improved for some period by VS. We conducted EMG analysis as described hereafter to examine the effect and underlying mechanisms of VS. Since she could not

walk independently, after this EMG examination on the 50th day after admission, the patient was transferred to a chronic rehabilitation hospital. After rehabilitation, the patient can now walk without aid.

VS was applied from the gluteal muscles to distal muscles through the lower limbs, on the left side, which was the dominant side of spasticity, for 2 min by a commercial massaging device with a frequency of 100 Hz. We measured the time of standing up and down from a chair five consecutive times and walking as fast as possible for 12 m. We also performed surface EMG and video recording. Thereafter, full-wave rectification of the surface EMG and quantitative coordination to kinesiology were performed with Teraview, a motion analysis software. The phases of the walking cycle were decided visually based on video data. The Mann-Whitney U test (StatView) was used for statistical analysis. $p < 0.01$ was used as the statistically significant level.

1. Effect on performance: The average standing-up time was 4.7 ± 1.7 (mean $\pm$ SD) and 2.8 ± 0.3 s, before and after VS, respectively. The average sitting-down time was 2.4 ± 0.2 and 2.3 ± 0.2 s, respectively. VS significantly shortened only the time of standing up from a chair ($p < 0.01$; figure 1). For the 12 m walking task, the average walking time was 27.3 and 23.2 s, before and after VS. The time reduced significantly with VS.
2. The total amount of EMG of each muscle for a single walking cycle (columns) and EMG activities changed during the walking phase (line graphs) (figure 2).

To understand the mechanisms of improved performance, we analysed quantitatively the amount of EMG for each muscle during one step. The integrated EMGs were smoothed and averaged for nine steps before VS and eight steps after. On the left lower limb, the amount of EMG on the soles decreased significantly after VS, but increased slightly on the antagonistic anterior tibial muscle. Some changes existed in the quadriceps and hamstrings.

A statistically significant difference was seen only for the soleus ($p < 0.01$).

Since EMG activities changed in phases through the walking cycle, EMG activities of each muscle were related to the walking cycle. A single walking cycle was divided into 10 segments. After VS, the magnitude of variability in EMG of all divided segments decreased remarkably in all examined muscles, not only on the vibrated side, but also on the non-vibrated side (data not shown). In particular, the activities in the soleus decreased in the swing phase after VS.

The improvement in performance by VS lasted for a single session of physical exercise for approximately 40 min, but did not last until the next day.

DISCUSSION

In this study, we reported a case with dysbasia due to NMO with spastic paraplegia improved by VS. Corticosteroid is the basic therapy for neurological inflammation for treating NMO. However, it is stressed that VS and physiotherapy may change the disordered activities among lower limb muscles to a functionally well-organised level. As a method of intervention for spasticity, stimulation with a vibrator may be a good option. We analysed the mechanism of VS.

Physiologically, when a mechanical vibrator (100–200 Hz) is applied over a relaxed muscle, it causes tonic EMG activities and contraction of the vibrated muscle. This is called TVR and is a kind of autogenic reflex.^{6 7} It is noted that the increased Ia excitatory input by the vibrator is important for TVR. When a vibrator is applied to a spastic muscle, it enhances the tone of the spastic muscle, but when it is applied to an antagonistic muscle, it causes an apparent reciprocal inhibition of the spastic muscles. Regarding the change in performance after VS as seen in this case, if TVR works to increase the EMG activities in weak muscles, one may expect the EMG to increase in vibrated muscular activity in general, but this

Figure 1 Effect on performance. The time taken to stand up became shorter and the patient's walking speed became faster. Average and SD are shown only for times of standing up and sitting down.

Figure 2 Total amount of electromyography (EMG) activities during a single walking cycle and change in EMG activities in the walking phase. Left column: The total amount of EMG during a single walking cycle changed in several muscles. Tibialis anterior (TA) muscle increased slightly, but not significantly, but soleus decreased significantly; these are antagonistic muscles. Right line graphs: The EMG activities of a single walking cycle were divided into 10 parts to examine the different contributions of each muscle to stance and swing phases in the walking cycle. The size of variability in EMG decreased remarkably. This was the case not only on the vibrated side, but also on the not vibrated side. After the vibration, in TA, there were biphasic increases both in stance and swing phase. On the contrary, the enhanced activity of the soleus in the late swing phase reduced significantly. These findings suggest that the reciprocity among antagonistic muscles was improved. Symbols and vertical lines show mean and SD.

was not the case in our patient. Instead, muscular activity of the soleus decreased, and that of the tibialis anterior increased. The soleus and tibialis anterior muscles are antagonistic for ankle movement. In patients with spasticity, it was shown that the soleus muscle is facilitated and tibialis anterior muscle is inhibited.⁸ Regarding VS which decreased the activity in the soleus muscle and increased the activity in the anterior tibial muscle, it is possible that VS acted not only through general excitation, but also through reciprocal innervation pathways.

The spinal cord is the most fundamental nerve module for walking. The modules are known as central pattern generators (CPGs), and they are neural circuits that autonomously generate rhythmic alternating contraction commands for the limbs' flexors and extensors, without relying on any other input.⁹ Regarding VS, a marked decrease of variabilities of EMG in total leg muscles could explain why VS acted not only on the segmental levels between L3 and S1, but also on CPG as a whole.

Neuroplasticity refers to the ability of the brain and spinal cord to change as a result of one's experience. Learning may

change the strength of the connections, by adding or removing connections. The effect of VS continued for as long as 40 min after the cessation of VS, which is longer than the synaptic mechanisms such as transmitter depletion and postsynaptic depletion. This finding may support the hypothesis of neuronal plasticity on CPG by VS.

Learning points

- ▶ Spastic paraparesis.
- ▶ Neuromyelitis optica.
- ▶ Disturbance in reciprocity.
- ▶ Vibration stimulation may improve reciprocity and performance.

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Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Hagbarth KE**, Eklund G. The effects of muscle vibration in spasticity, rigidity, and cerebellar disorders. *J Neurol Neurosurg Psychiatr* 1968;**31**:207–13.
2. **Ageranioti SA**, Hayes KC. Effects of vibration on hypertonia and hyperreflexia in the wrist joint of patients with spastic hemiparesis. *Physiother Canada* 1990;**42**:24–33.
3. **De Gail P**, Lance JW, Neilson PD. Differential effects on tonic and phasic reflex mechanisms produced by vibration of muscles in man. *J Neurol Neurosurg Psychiatry* 1966;**29**:1–11.
4. **Hagbarth KE**, Eklund G. Tonic vibration reflexes (TVR) in spasticity. *Brain Res* 1966;**2**:201–3.
5. **Eklund G**, Hagbarth KE. Normal variability of tonic vibration reflexes in man. *Exp Neurol* 1966;**16**:80–92.
6. **Gillies JD**, Burke DJ, Lance JW. Supraspinal control of tonic vibration reflex. *J Neurophysiol* 1971;**34**:302–9.
7. **Burke D**, Hagbarth KE, Lofstedt L, *et al*. The responses of human muscle spindle endings to vibration during isometric contraction. *J Physiol* 1976;**261**:695–711.
8. **Yanagisawa N**, Tanaka R, Ito Z. Reciprocal Ia inhibition in spastic hemiplegia of man. *Brain* 1976;**99**:555–74.
9. **Grillner S**. Biological pattern generation: the cellular and computational logic of networks in motion. *Neuron* 2006;**52**:751–66.

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