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Vibro-Tactile Stimulation as a Non-Invasive Neuromodulation Therapy for Cervical Dystonia: A Case Study

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Abstract— **Introduction: Cervical dystonia (CD) is a type of focal dystonia that is characterized by involuntary neck postures. The underlying neurophysiology mechanism of CD is unknown, but there is increasing empirical evidence that motor deficits of CD are associated with somatosensory and proprioceptive deficits in the upper limb area. Vibro-tactile stimulation (VTS) is a non-invasive somatosensory stimulation approach where afferent signals from the vibrated muscle and tactile mechanoreceptors modulate cortical activity. Previous studies have shown that VTS could be an effective neuromodulation therapy for treating laryngeal dystonia. This proof-of-concept study examined the effect of VTS on alleviating the involuntary cervical muscle contractions in a female participant with intermittent torticollis. Methods: VTS was applied sequentially on four neck positions: bilateral trapezius (TRP) and bilateral sternocleidomastoid (SCM). Each VTS site was vibrated continuously for six minutes. The kinematics and underlying neck muscle activities during dystonic neck movements were examined with acceleration and surface electromyography (sEMG). To quantify the efficacy of VTS, two acceleration features and one sEMG feature were derived: (1) number of acceleration peaks per minute; (2) peak amplitude of acceleration (PAA); (3) change in power of sEMG after VTS. Results: The frequency of intermittent dystonic neck movements decreased by 60% after VTS. In addition, PAA during dystonic episodes was drastically reduced after VTS when compared to baseline. Third, the effectiveness of VTS in alleviating dystonic muscle spasms depended on the site of vibration. For this participant, the left trapezius muscle was shown as the optimal vibration site reducing sEMG signal power by 15% across all recorded muscles. Conclusion: This case study offered preliminary insight into the assumed effectiveness of neck muscle VTS as a treatment for CD. Our participant experienced pain relief after VTS with measured improvements reflected by electrophysiological and kinematic data. A systematic study with a larger sample size is required in the future to validate the effectiveness of VTS for treating symptoms in CD.**

I. Introduction

Cervical dystonia (CD) is a type of focal dystonia characterized by involuntary neck muscle contractions, resulting in abnormal cervical movements or postures [1]. The pattern of dystonic neck movement and postures is variable in patients with CD. Symptoms are clinically categorized into torticollis, laterocollis, retrocollis, or anterocollis [1]. Torticollis is the most common form of CD, where the head remains upright and may turn to one side intermittently then releases. Laterocollis is the second most common form of CD, where the head is pulled laterally to one side of the shoulder. Retrocollis and anterocollis happen respectively when the head is pulled backward (head extension) and towards the chest (head flexion).

The underlying neurophysiological mechanism of CD is unknown. It has been long hypothesized to be associated with somatosensory and proprioceptive deficits [2], [3]. Khosravani et al. investigated the effect of botulinum neurotoxin (BoNT) injection on wrist proprioception and corresponding sensorimotor cortical activities in CD patients [4]. During an active movement wrist position matching task, an excessive rise of premotor/motor cortical β-oscillations (13-30Hz) with a significantly larger error in matching wrist position was observed in CD patients when compared to healthy controls. This abnormal active position sense of CD patients is in line with the hypothesis of the proprioceptive dysfunction in CD. This can open an avenue for a potential treatment using sensory stimulation and altering afferent signals.

Vibro-tactile stimulation (VTS), as a form of somatosensory stimulation, is known to alter afferent signals from the vibrated mechanoreceptors in muscles and tactile receptors in the skin [5]. VTS was shown to improve voice quality in spasmodic dysphonia, as another type of focal dystonia [6]. However, the effect of VTS on CD is not clear.

To address the clinical need of an alternative behavioral therapy for individuals with CD, this proof-of-concept study investigated, if VTS of neck muscles is a suitable non-invasive form of neuromodulation that induces measurable improvements in CD. To this end, this study pursues two aims: First, to obtain preliminary data on the assumed effectiveness of VTS in reducing the extent and frequency of dystonic head postures in CD. Second, to examine the dependency of muscle vibration site on reducing dystonic muscle activities.

II. Method

A. Participant

We here report the case of a 10-year-female with juvenile CD. The participant receives regular Botox injections as a symptomatic treatment. She was at the end of her Botox treatment cycle when participating in this study. The participant presented with intermittent right torticollis with concurrent left shoulder elevation. Additionally, the participant self-reported experiencing pain and discomfort as the dystonic movement/posture occurred. This study is exempt from the Institutional Review Board review of the University o Minnesota.

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B. Apparatus

To record muscle activity, the sEMG system (Biometrics ltd.) was used for this study. Four sEMG electrodes were respectively attached to bilateral sternocleidomastoid (SCM) and trapezius (TRP) muscles to record electrical activity. One ground electrode was attached to C7 spinal segment (Fig. 1).

To record the acceleration of head movements, one wired accelerometer (Biometrics ltd., Model S3-1000G-HA) was attached to the forehead. The 3-axis accelerometer provided simultaneous measurements in three orthogonal directions. The range of acceleration for detection is within ±1000G.

To apply vibration on neck muscle, small, low-voltage (~1V) encapsulated vibrators (Precision MicrodrivesTM, Model 307 – 100) were attached noninvasively to different locations of the cervical area (Fig. 2). Although it has long been established that VTS in the range of 40-100 Hz stimulates the mechanoreceptors and muscle spindles that affect motor behavior [5], [7], the knowledge on the optimal frequency of VTS for modulating CD is still inconclusive. In this study, the vibration frequency was experimented with the frequency range 90-110 Hz.

C. Experimental Procedure

The study involved two conditions: (1) Unrestricted dystonia, and (2) VTS application.

Unrestricted dystonia: This condition recorded sEMG and acceleration signals of cervical movements during rest and symptomatic state. Vibrators were not attached to the neck, thus there was no VTS applied. During the unrestricted dystonia condition, the participant was asked to sit and keep her head in a neutral, comfortable position. The participant was instructed to not resist any involuntary muscle contraction and let her neck assume the dystonic posture once it became symptomatic. The duration of each unrestricted dystonia condition was 3 minutes.

VTS application: This condition aims to examine the efficacy of VTS on alleviating involuntary muscle contractions in CD. Four vibration sites were tested on the following regions: the skin above the right and left SCM and right and left TRP. For each vibration site, two vibrators are attached and activated simultaneously. The maximum duration of each VTS trial was initially set as 6 minutes and adjusted by the participant's response. For instance, vibration was stopped whenever the participant reported increased pain or discomfort. Since there is no gold standard for choosing the ideal VTS duration for CD, the current 6-min duration was determined by using a similar value that was previously applied to modulating SD [6].

Data collection began with an unrestricted dystonia condition as the baseline to record the initial muscle behavior during rest and symptomatic state. The VTS was then applied sequentially on each of the four vibration sites. An unrestricted dystonia condition followed right after the end of VTS condition. The study ended with another unrestricted dystonia condition as the retention condition to record changes in cervical muscle behaviors 10 minutes after VTS. There was a 3-min rest after each VTS condition to avoid muscle fatigue. The complete protocol is summarized in Table 1.

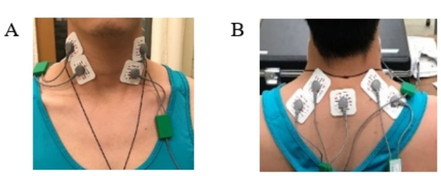

Figure 1. Attachment of sEMG electrodes on A) sternocleidomastoid and B) trapezius. A ground electrode is attached to the C7 spinal segment.

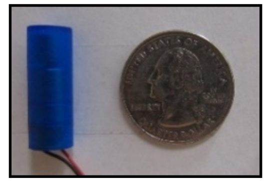

Figure 2. The vibrator used for this study. Length: 25 mm. Diameter: 9 mm

D. Data analysis

Data analysis and signal processing were conducted with MATLAB2019a (The MathWorks Inc., Natick, MA, U.S.A.). Raw sEMG data were filtered with a 16th order 100 Hz low-pass Butterworth filter and subsequently with an 8th order 50-70 Hz band-stop Butterworth filter to eliminate the power line noise. Electrophysiological signatures of dystonic neck postures were identified by time-frequency analysis of filtered sEMG signal. First, in order to quantify the magnitude of involuntary neck muscle contractions, power of rectified sEMG was calculated which measures the time-averaged energy contained in the sEMG data. A higher power reflects a stronger muscle contraction. The time normalized power of sEMG is calculated in (1):

$$p = \left| \frac{\sum_{t_1}^{t_2} x_N^2}{t_2 - t_1} \right| \quad (1)$$

where x_N is the amplitude of rectified sEMG signal at sample index N, t_1 and t_2 are two time intervals.

TABLE I. EXPERIMENTAL PROCEDURE

Session	Condition	Duration (min)
Baseline	Unrestricted dystonia	3
VTS	Right SCM	6
	Rest	3
	Right TRP	6
	Rest	3
	Left TRP	6
	Rest	3
	Left SCM	6
Post VTS	Unrestricted dystonia	3
Rest		7
Retention	Unrestricted dystonia	3

Second, to investigate the effect of VTS on alleviating dystonic muscle activities, a percentage decrease in the power of rectified sEMG was obtained for each VTS trial by comparing it to the baseline. Change in the power of sEMG is calculated in (2):

$$\text{Change in power of sEMG} = \frac{p_{ref} - p_{test}}{p_{ref}} \times 100\% \qquad (2)$$

where p_{ref} represents power of sEMG in baseline, p_{test} represents power of sEMG in VTS condition.

The kinematic data of neck movements were recorded by the triaxial accelerometer in lateral, longitudinal, and vertical axis. Acceleration data were denoised down to level 6 using wavelet denoising and subsequently filtered with a 6th order 10Hz low-pass Butterworth filter. Movements can be identified by peaks in the plot of filtered acceleration data. Thus, to locate the dystonic neck movements, the upper envelope of the original acceleration data was obtained using spline interpolation over local maxima separated by at least 300 samples. Then the peaks of the upper envelope were filtered with a threshold of 0.3g and separated by at least 200ms. The proper value of threshold was determined by sequentially comparing different values of threshold, where the ideal threshold was expected to keep major peaks that represented the dystonic neck movements and to eliminate minor ones that were caused by body sway. The filtered peaks were then cross-validated with sEMG data to ensure that they represented the dystonic neck movements. Second, the average number of dystonic movements per minute (DM/min) was determined by the number of peaks in acceleration data during baseline, post VTS, and retention condition, which reflected changes in the frequency of dystonic neck movements. Third, the peak amplitude of acceleration (PAA) during dystonic episodes was calculated to investigate the severity of each dystonic neck movement.

III. Results

Compared to the baseline condition, the frequency and amplitude of involuntary muscle contractions decrease (Fig. 3A). Meanwhile, the average peak amplitude of acceleration after VTS is observed to be lower than baseline condition (Fig. 3B). An average number of 15 dystonic movements per minute (15 DM/min) was recorded in the baseline condition. Compared to baseline, the frequency of dystonic neck movements decreases by 60% to 6 DM/min in post VTS condition. However, the frequency approached baseline after 7 minutes of retention (Fig. 4).

Fig. 5 shows the distribution of PAA in baseline, post VTS, and retention condition. The PAA in post VTS (median: 0.59) is lower than the PAA in baseline (median: 0.90). However, after 7 minutes of retention the median PAA again approached the baseline level (median:0.89). Meanwhile, the inter-quartile range of PAA drops by 53% in post VTS condition compared to the baseline. Additionally, while the PAA of baseline and retention condition is shown to be normally distributed, the PAA of post VTS condition is right-skewed.

The effect of vibration sites is illustrated in Fig. 6. Overall, all four vibration sites show positive rate of reduction (mean = 8.5%). Among all four vibration sites, left TRP site demonstrates the most prominent effect (mean = 14.7%) in reducing sEMG power, followed by left SCM site (mean = 10.7%). Furthermore, power of sEMG in right SCM and right TRP increases when VTS is applied on them (change in right SCM = -1.5%, change in right TRP = -5.9%). Results show that vibration on ipsilateral side of the dystonic movement (mean = 4.2%) has less reduction than vibration on contralateral side (mean = 12.7%). Overall, left TRP is shown

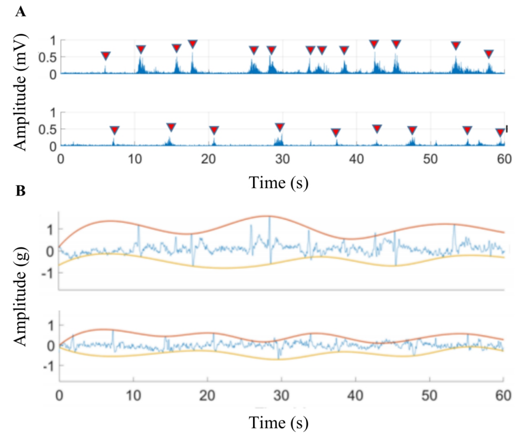

Figure 3. Changes in (A) rectified sEMG of right trapezius and (B) acceleration in lateral direction after VTS. (A) Frequency and magnitude of involuntary muscle contractions decreased. (B) Envelope amplitude of acceleration during dystonic neck movements decreased after VTS (blue: original acceleration data; red: upper envelope; yellow: lower envelope).VTS: vibro-tactile stimulation.

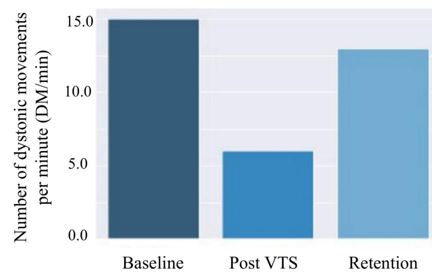

Figure 4. Number of dystonic movements per minute before and after VTS. The frequency of dystonic movements decreased by 60% post VTS compared to baseline but then increases after seven minutes of retention. DM/min: Number of dystonic movements per minute.

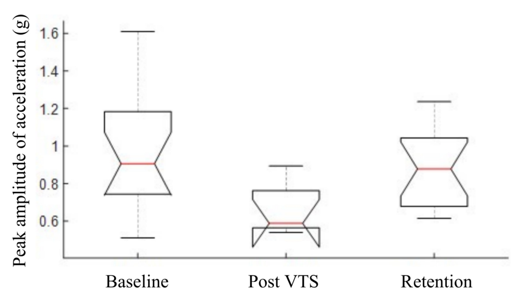

Figure 5. Peak amplitude of acceleration (PAA) during dystonic episodes in three conditions. The red line is the median, the top of the box is the 75th percentile, the bottom of the box is the 25th percentile. The whiskers represent the maximum and minimum values of the peak amplitude. VTS: vibro-tactile stimulation.

as the optimal vibration site with the highest average reduction in the power of sEMG.

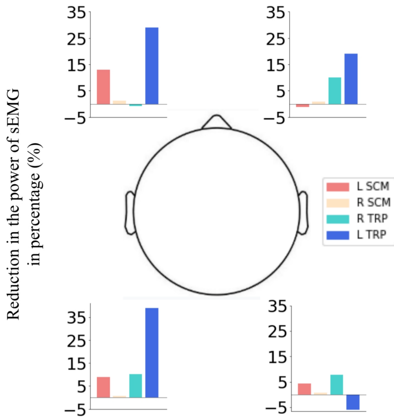

Figure 6. Change in the power of sEMG during four vibration sites. Bar plots are arranged by the anatomical location of vibration sites. Left TRP site is shown as the optimal vibration site with the highest average reduction in sEMG power. L: left. R: right. SCM: sternocleidomastoid. TRP: trapezius.

IV. Discussion

A. Effect of VTS on dystonic symptoms

The efficacy of VTS on CD was illustrated by examining electrophysiological and kinematic features of dystonic neck movements before and after VTS. The clearest evidence is the decreased frequency of intermittent dystonic neck movements after VTS. Additionally, a drastic reduction in PAA and power of sEMG was found after VTS, which demonstrates an alleviation in the extent of dystonic neck movements. Furthermore, the participant self-reported experiencing pain relief during and after applying VTS to her cervical muscles. However, it was also observed that the frequency and amplitude of dystonic movements approached baseline after 7 minutes of retention. It remains unclear how long the VTS efficacy would last as well as the influencing factors.

B. The role of vibration sites on the effectiveness of VTS

Four vibration sites were experimented with different effects shown on alleviating the intermittent dystonic movements. Among four vibration sites, left TRP was shown as the optimal vibration site. In general, vibration sites on the contralateral side of right torticollis (left SCM site and left TRP site) were more effective in reducing involuntary muscle contractions than vibration on ipsilateral sites. One thing to notice is that during vibration on the ipsilateral side of right torticollis (right SCM site and right TRP site), an increase was found in the sEMG power of right SCM and right TRP. This deterioration in muscle spasms demonstrates that VTS might induce negative effects in the vibrated muscles, which underlines the importance of the selection of vibration sites when applying VTS.

C. Future work

This case study requires a systematic investigation with a larger sample size to validate the effectiveness of VTS for treating symptoms in CD. Additionally, it is not clear if the current amplitude and placement of vibration were capable of targeting deeper neck muscles such as *splenius capitis*, which might further improve the effect of VTS. Thus, future investigation needs to focus on the impact of vibration amplitude and electrode placement during VTS application.

V. Conclusion

This proof-of-concept study examined the effect of VTS on alleviating involuntary cervical muscle contractions in one participant with intermittent torticollis. The frequency and extent of the dystonic postures were markedly reduced during and immediately after VTS application. Furthermore, the effectiveness of VTS in alleviating dystonic muscle spasms was shown to be dependent on vibration sites. Future studies are required to validate the effectiveness of VTS on CD with a larger sample size.

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