

Slow-Down Exercise Reverses Sensorimotor Reorganization in Focal Hand Dystonia: A Case Study of a Pianist

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Abstract

Focal hand dystonia in musicians, which is characterized by involuntary flexion and/or extension of fingers while playing musical instruments, is a disabling neurological disorder that can even threaten their careers. The present study investigated whether or how a non-invasive intervention for focal hand dystonia called “slow-down exercise” affects motor performance, muscular activity, and somatosensation in a dystonic pianist. The patient was asked to perform a simple five-finger exercise at three different tempi on a digital piano, just prior to, 1, 2, 3, 6, and 12 months after the onset of the slow-down exercise training. As the rehabilitation proceeded, the patient improved the regularity of piano keystrokes, as objectively quantified using musical instrument digital interface signals. Measurements of surface electromyographic activity from the forearm muscles demonstrated that the patient gradually regained the inherent bilateral difference in the co-contraction level of the extensor digitorum communis and flexor digitorum superficialis muscles. Furthermore, the practice of slow-down exercise lowered the two-point discrimination thresholds of affected fingers, indicating the restoration of tactile spatial resolution. These findings not only confirm the effectiveness of slow-down exercise for the treatment of focal hand dystonia but also provide objective evidence that a simple behavioral intervention can reverse the reorganization of sensorimotor neural networks in dystonic patients.

Keywords: Focal hand dystonia; Musician; Pianist; Slow-down exercise; Electromyographic activity; Co-contraction; Tactile spatial discrimination

Introduction

Focal hand dystonia in musicians, also referred to as musician's cramp, is a highly disabling movement disorder that is characterized by involuntary flexion and/or extension of fingers while playing musical instruments [1,2]. Based on the fact that these symptoms often disappear when playing at a slow tempo, a non-invasive training method called “slow-down exercise (SDE)” has been developed [3]. The SDE training involves the selection of a short passage that triggers the symptoms, followed by the practice of the passage at a tempo that is slow enough for dystonic patients to play without involuntary movements, while gradually increasing the performing tempo. This simple training has been shown to be highly effective in treating focal hand dystonia in pianists: it significantly lowered the Arm Dystonia Disability Scale (ADDS) [4] scores in 20 affected pianists [3]. However, it remained unclear which components of motor skill are improved by the SDE training. To address this, the present study aimed to objectively quantify the changes in motor performance in the process of SDE training. To this end, we measured the regularity of piano keystrokes by using the musical instrument digital interface (MIDI) signals [5].

Clinical observations suggest that focal hand dystonia is accompanied by the co-contraction of antagonistic muscles in the arm [2]. However, very few attempts have been made to directly assess the changes in the level of co-contraction during rehabilitation of affected musicians. We thus examined whether the SDE training modifies the co-contraction level of forearm antagonistic muscles by recording surface electromyographic (EMG) activity while the patient was playing the piano.

Accumulating evidence suggests that tactile spatial resolution of affected fingers is impaired in dystonic patients [6,7], which seemingly reflects enlarged and overlapping cortical representations of affected fingers in the primary somatosensory cortex [8,9]. If the SDE training can also reverse this maladaptive somatosensory reorganization, the discrimination acuity of affected fingers should improve as the

rehabilitation proceeds. Based on this assumption, we also examined the changes in two-point discrimination thresholds of affected fingers during the SDE training period.

Materials and Methods

Patient

A female classical piano player with a four-month history of focal hand dystonia participated in this study. The patient was 25 years old at the onset of SDE training, and was right-handed with a laterality quotient of the Edinburgh Handedness Inventory [10] of 89. The patient started to learn the piano at the age of four, and had received 20 years of intensive piano training. As a highly skilled amateur pianist, she used to give an average of 4-5 public performances in concerts and competitions each year before the onset of focal hand dystonia, and had received a prize in a prestigious national competition. However, the development of focal hand dystonia deprived her of the ability to play the piano and forced her to completely withdraw from public performances. Her complaint was that the fifth digit of the right hand involuntarily extended, while the third and fourth digits flexed, during piano playing (Figure 1). The patient underwent complete neurological examinations (e.g., radiography of the cervical spine) with no abnormalities noted, and was diagnosed with focal hand dystonia by one of the authors (NS). The patient was prescribed no medication. The patient gave written

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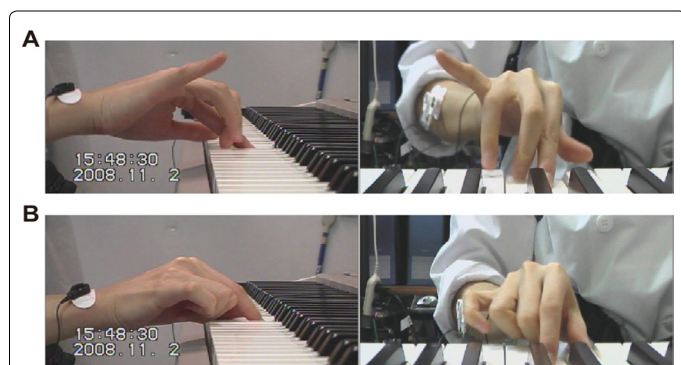


Figure 1: The patient's right hand during performance of the five-finger exercise at 160 bpm before the onset of SDE training. The fifth digit overextended just prior to its keystroke (A), and the third and fourth digits subsequently overflexed during downward movements (B).

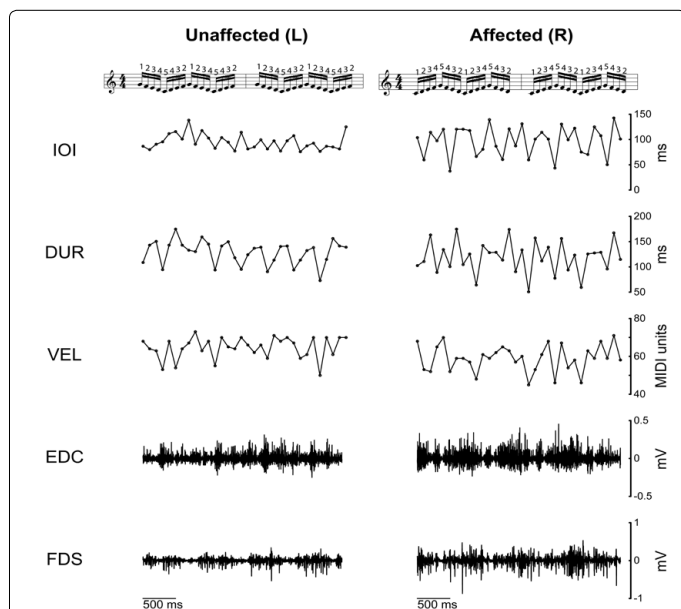


Figure 2: Typical examples of MIDI and raw EMG data. The left column shows the data recorded from the unaffected hand and the right column the affected hand. L, left; R, right; IOI, inter-onset intervals; DUR, tone durations; VEL, key velocities; EDC, extensor digitorum communis muscle; FDS, flexor digitorum superficialis muscle.

informed consent prior to the experiment, which was conformed to the Declaration of Helsinki, and fully agreed to publish this study.

Procedure

We asked the patient to practice the SDE [3] using a simple five-finger motor task (1-2-3-4-5 and back; Figure 2) every day. The patient chose to practice this specific passage because it evoked dystonic movements (i.e., involuntary extension of the fifth digit and flexion of the third and fourth digits) most substantially. The patient firstly identified a tempo at which she could play the passage without any dystonic symptoms by using a metronome, and repeatedly practiced the passage at this tempo. As soon as she became comfortable with the tempo, she slightly increased the performing tempo and practiced the passage at this new tempo. In this way, the patient gradually increased the performing tempo. If the dystonic symptoms emerged during this process, she decreased the tempo to a comfortable level. The patient underwent this basic SDE training for 30 minutes per day for 12 months.

According to the previous successful clinical trial [3], the patient was allowed to freely perform additional SDE and practice normal musical pieces during the training period. Interviews with the patient indicated that besides the basic 30-minute training she practiced the SDE with various piano etudes (e.g., “The Virtuoso Pianist in 60 Exercises” by Charles-Louis Hanon, “Le déliateur” by Ernest Van de Velde) for ~30 minutes per day with the aim of regaining finger strength and independence. The patient reported that she focused on relaxing arm and shoulder muscles between individual keystrokes in performing the SDE. To this end, she applied self-massage to these muscles before starting the training. After three months of intensive SDE training the patient resumed practicing normal musical pieces (0.5 - 1.5 hours per day), occasionally applying the SDE to difficult passages.

We assessed the patient's motor performance six times, namely just prior to, 1, 2, 3, 6, and 12 months after the onset of SDE training. The patient performed four cycles of the five-finger exercise (Figure 2) on a digital piano (P-60s; Yamaha, Hamamatsu, Japan), with each hand separately. The patient was asked to play mezzo-forte (medium loudness) and in legato-style (notes were played in a smooth, connected manner). The tempo was paced by a metronome, which was set to quarter notes. We asked the patient to perform the task five times at each of three different tempi, namely 80, 120, and 160 beats per minute (bpm). When the patient made a mistake (i.e., wrong, omitted, or extra notes), we asked her to perform the task again to obtain data of five correct trials for each hand at each tempo. The keyboard of the digital piano was equipped with a MIDI, the output of which was fed to a personal computer via a MIDI translator (MIDI sport uno; M-audio Japan, Tokyo, Japan). For recording and generating MIDI-files, we used a commercially available music editing software (Singer Song Writer Lite 4.0 for Windows; Internet, Tokyo, Japan). The software was used to measure inter-onset intervals (time intervals between the onset of a note and that of the subsequent note, IOI in ms), tone durations (time intervals between the onset of a note and the end of the same note, DUR in ms), and key velocities (an indirect measure of loudness ranging from 0 to 127, VEL in MIDI units). The patient's performance was quantified by calculating the coefficients of variance (CV) of these three parameters [5,11], which were averaged across five trials for each hand and for each of the three tempi. Since producing regular keystrokes is a fundamental skill component in piano playing, we used these measures of regularities of keystroke timing and loudness to quantify the improvement in the patient's ability to play the instrument. The very last notes of the task (G4 for the left hand and C4 for the right hand) were excluded from the analyses because they tended to be elongated.

While the patient was performing the motor task, surface EMG activity was recorded from the extensor digitorum communis (EDC) and flexor digitorum superficialis (FDS) muscles of the unaffected non-dominant (i.e., left) arm and the affected dominant (i.e., right) arm. Bipolar Ag/AgCl disposable electrodes (10 mm diameter) were placed at the estimated motor point of each target muscle with a 20-mm center-to-center distance. At each electrode position, the skin was cleaned using alcohol to reduce skin resistance. During EMG recording, special care was taken to prevent the connecting cables from disturbing the patient. The EMG signals were amplified 500 times and sampled at 1,000 Hz with a MP100 data acquisition system (Biopac Systems Inc., CA, USA) interfaced with a personal computer. Figure 2 shows typical examples of raw EMG data along with MIDI data. EMG signals were high-pass filtered at 20 Hz, full-wave rectified, and low-pass filtered at 50 Hz. Subsequently, baseline noise (i.e., mean resting EMG amplitude of pre- and post-performance periods) was subtracted from the signals. To examine the co-contraction level of antagonistic muscles in the forearm,

we computed the co-contraction index [12,13] for the EDC and FDS muscles. The rectified and smoothed EMG signals were first scaled to a mean of 1, and the normalized EMG signals of the two muscles were overlaid. Then for each point in time, the minimum value of the normalized signals was calculated, discarding the portion of EMG signals in one muscle that is not matched by those in the other muscle. The co-contraction index was computed by averaging the resulting time-varying signals during performance of the five-finger exercise (i.e., period between the onset of the first note and that of the last note).

In addition to motor performance, we investigated changes in tactile spatial resolution of the affected fingers (third, fourth, and fifth digits) over the period of rehabilitation. To this end, we assessed the two-point discrimination threshold [14] for each digit five times, namely just prior to, 1, 3, 6, and 12 months after the onset of SDE. During the assessment the patient sat with the forearm in supination, resting comfortably on a desk, and with eyes closed. The ends of the arms of the esthesiometer were placed simultaneously on the volar aspect of the middle phalanx of a digit without application of any pressure except for the weight of the instrument. We conducted eight assessment trials for each digit, with four trials in each of two different conditions: in the decrease condition, the experimenter started at a distance longer than 20 mm, gradually decreasing the distance until the patient could not differentiate between the two points. In the increase condition, the experimenter started at a distance of 0 mm, gradually increasing the distance until the patient could differentiate between the two points.

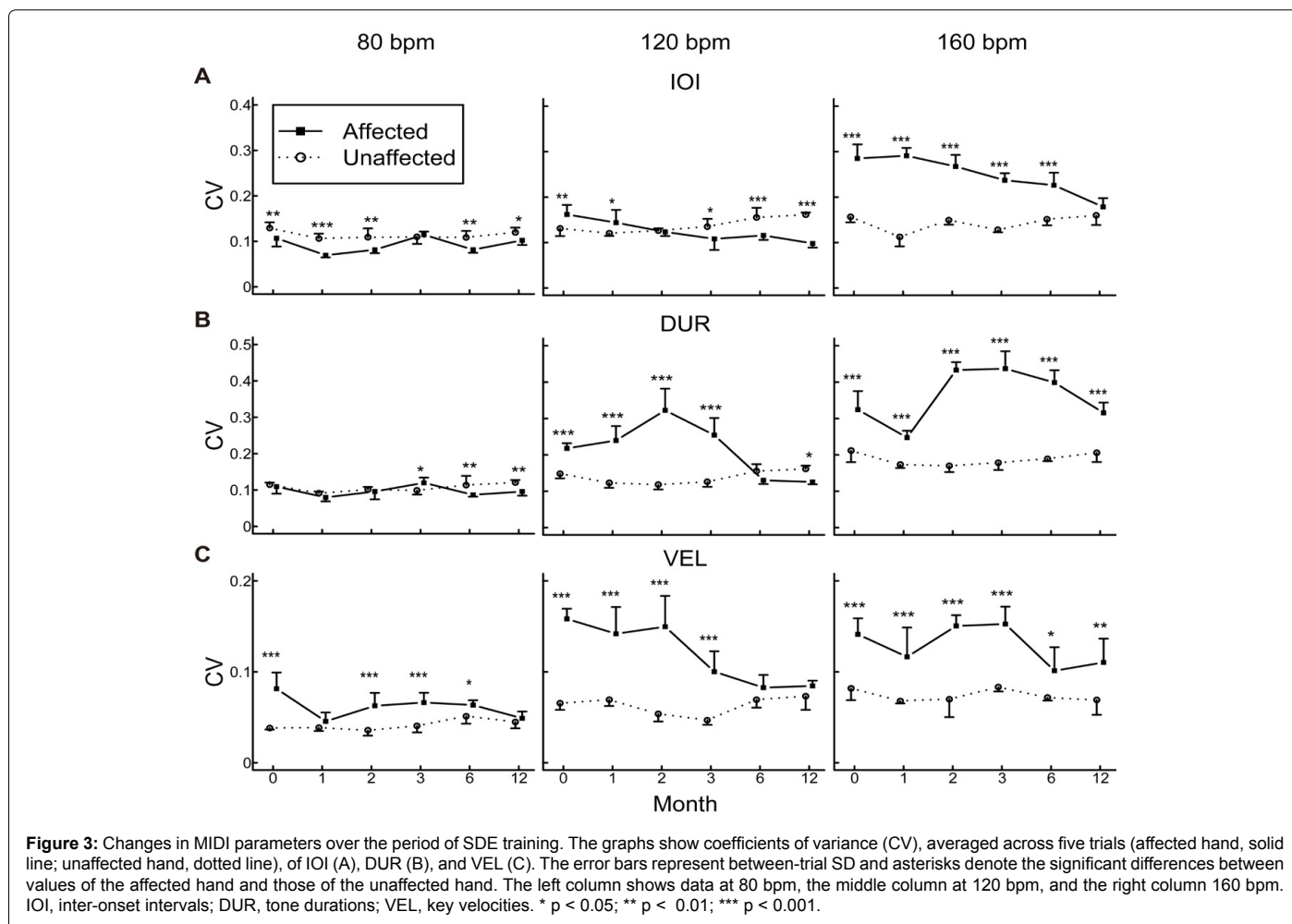
Assessments were performed in a randomized order, and catch trials were randomly applied to enhance measurements. The two-point threshold was determined by averaging the thresholds obtained from the eight trials, for each digit.

Statistical analyses

To examine the effects of SDE training on MIDI parameters and the co-contraction index, we first conducted a three-way ANOVA of side (affected or unaffected) \times month (0, 1, 2, 3, 6, or 12 months) \times tempo (80, 120, or 160 bpm). When the three-way interaction was found to be significant, we subsequently examined the simple interaction effects of side \times month by performing a two-way ANOVA for each tempo. For the tempo at which side \times month interaction effect was significant, simple-main effects of side were examined with a one-way ANOVA for each month. As for the two-point discrimination threshold data, a three-way ANOVA of side (affected or unaffected) \times month (0, 1, 3, 6, or 12 months) \times digit (third, fourth, or fifth digit) was performed. The p value of < 0.05 was regarded as statistically significant.

Results

The SDE training significantly improved the regularity of keystrokes in the patient. Figure 3 displays the changes in the CVs of three MIDI parameters during the training period. Three-way ANOVAs revealed significant effects of side \times month \times tempo interaction for IOI ($F(10, 143) = 9.91, p < 0.001$), DUR ($F(10, 143) = 15.38, p < 0.001$), and VEL



($F(10, 143) = 3.08, p < 0.01$). Tests of simple effects performed on IOI data showed significant effects of side \times month interaction at all of the three different tempi (80 bpm: $F(5, 48) = 3.38, p < 0.05$; 120 bpm: $F(5, 48) = 12.70, p < 0.001$; 160 bpm: $F(5, 47) = 19.13, p < 0.001$). Although the CV of IOI tended to be lower for the affected dominant hand than the unaffected hand ($p < 0.05$) at 80 bpm, at faster tempi the IOI regularity of the affected hand markedly improved during the training period. At 120 bpm, the CV of IOI was higher for the affected hand just prior to, and one month after the onset of SDE training. However, the difference between the two hands disappeared one month later, followed by significantly lower CVs for the affected hand ($p < 0.05$). At 160 bpm, the difference in IOI regularity between the affected and unaffected hands gradually reduced, and it disappeared 12 months after the onset of SDE. The improvement in the regularity of DUR and VEL became visible slightly later than that in IOI regularity. Two-way ANOVAs revealed significant effects of side \times month interaction at all tempi for DUR data (80 bpm: $F(5, 48) = 3.98, p < 0.01$; 120 bpm: $F(5, 48) = 29.60, p < 0.001$; 160 bpm: $F(5, 47) = 19.43, p < 0.001$), and for VEL data (80 bpm: $F(5, 48) = 6.45, p < 0.001$; 120 bpm: $F(5, 48) = 12.56, p < 0.001$; 160 bpm: $F(5, 47) = 2.44, p < 0.05$). Although at 80 and 120 bpm the CV of DUR became significantly lower for the affected hand 6 months and 12 months after the onset of SDE training respectively, at 160 bpm it remained higher for the affected hand compared to the unaffected hand throughout the training period ($p < 0.001$). Similarly, while the affected hand consistently showed more irregular VEL than the unaffected hand ($p < 0.05$) at 160 bpm, the difference in VEL regularity between the two hands disappeared 12 months and 6 months after the onset of SDE at 80 and 120 bpm respectively. Because of the dramatic improvement in

the ability to play the piano, the patient successfully returned to public performance 15 months after the onset of SDE.

The practice of SDE also influenced the co-contraction level of antagonistic muscles in the forearm (Figure 4). A three-way ANOVA on the co-contraction index revealed a significant effect of side \times month \times tempo interaction ($F(10, 143) = 4.35, p < 0.001$). Follow-up ANOVAs showed significant effects of side \times month interaction at 80 bpm ($F(5, 48) = 8.42, p < 0.001$) and 160 bpm ($F(5, 47) = 12.20, p < 0.001$). Additional analyses revealed that at 80 and 120 bpm the co-contraction level was consistently lower for the affected forearm than for the unaffected forearm ($p < 0.01$). At 160 bpm, on the other hand, we found no difference in the co-contraction level between the two arms just prior to, and one month after the onset of SDE. The co-contraction level became significantly lower for the affected forearm two months after the onset of SDE ($p < 0.05$).

In addition, as the rehabilitation proceeded, tactile spatial resolution of the affected fingers has significantly improved (Figure 5). A three-way ANOVA on two-point discrimination threshold data with the factors side, month, and digit revealed significant effects of side \times month interaction ($F(4, 322) = 22.39, p < 0.001$) and side \times digit interaction ($F(4, 322) = 2.81, p < 0.05$). Additional analyses showed that for all the investigated fingers (i.e., third, fourth, and fifth digits) the discrimination thresholds were significantly higher for the affected hand than for the unaffected hand just prior to, and one month after the onset of SDE ($p < 0.01$). Interestingly, these differences disappeared three months after the SDE onset.

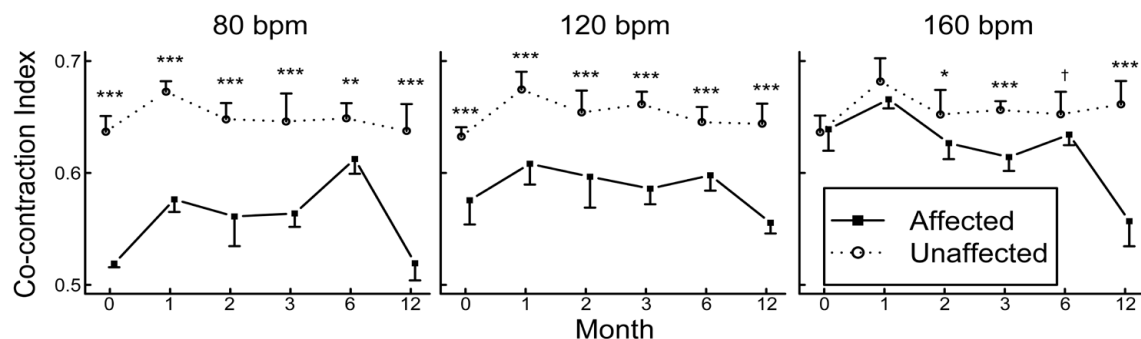


Figure 4: Changes in co-contraction index over the period of SDE training. The graphs show values of co-contraction index averaged across five trials (affected hand, solid line; unaffected hand, dotted line). The error bars represent between-trial SD and asterisks denote the significant differences between data from the affected arm and those from the unaffected arm. † $p < 0.10$; * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$.

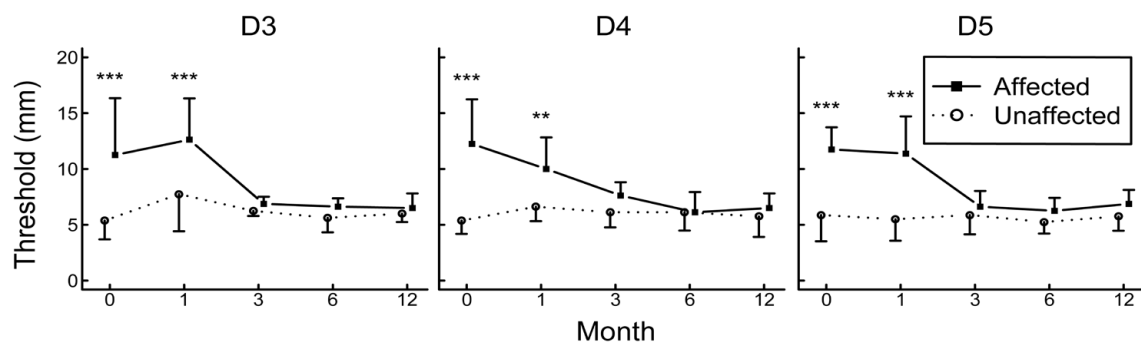


Figure 5: Changes in two-point discrimination threshold over the period of SDE training. The graphs show values of discrimination threshold averaged across eight trials (affected hand, solid line; unaffected hand, dotted line). The error bars represent between-trial SD and asterisks denote the significant differences between data from the affected hand and those from the unaffected hand. D3, third digit; D4, fourth digit; D5, fifth digit. ** $p < 0.01$; *** $p < 0.001$.

Discussion

The present study investigated how the non-invasive SDE training modifies motor performance, muscle co-contraction, and tactile spatial resolution in a pianist with focal hand dystonia by combining measurements of MIDI parameters, forearm EMG activity, and two-point discrimination thresholds of fingers.

The MIDI data confirmed the effectiveness of SDE training in treating focal hand dystonia. The improvement in motor performance was especially remarkable for IOI at faster tempi. Although the affected right hand showed more irregular IOI than the unaffected left hand before the onset of SDE training at 120 and 160 bpm, only two months of training successfully extinguished the difference in IOI regularity between the two hands at 120 bpm, and 12 months of training did so even at 160 bpm. The IOI data at 80 bpm, on the other hand, indicated that the affected dominant hand was superior to the unaffected non-dominant hand in terms of timing control of keystrokes already before the onset of SDE training. This suggests that there was inherent functional asymmetry between the dominant and non-dominant hands in this patient. Compared to the IOI, the changes in the regularity of DUR and VEL were observed later in the training period. At 120 bpm the differences in the regularity of these two parameters between the two hands disappeared only after six months of training. At 160 bpm the regularity of DUR and VEL remained higher for the affected hand than for the unaffected hand throughout the training period. These results indicate that the regularity of IOI, which enormously influences the quality of musical performance, is more sensitive to performance improvement in dystonic pianists compared to the other MIDI parameters. The present findings confirmed the validity and reliability of MIDI parameters in assessing changes in motor performance of dystonic pianists. Nowadays, many commercially-available digital pianos are equipped with MIDI terminals, and the handy measurements of MIDI signals would be useful for precisely and objectively quantifying motor performance of dystonic pianists for the purpose of diagnosis and the evaluation of treatment efficacy.

Previous literature suggested that the co-contraction level of antagonistic muscles is generally increased in patients with focal hand dystonia [2]. Interestingly, however, the present EMG data demonstrated that this effect is tempo-dependent. Contrary to our assumption, at 80 and 120 bpm the co-contraction level of antagonistic muscles was lower for the affected forearm than for the unaffected forearm already before the onset of SDE training. This might be explained by the fact that the co-contraction level is generally lower in the dominant arm than in the non-dominant arm during finger movements, possibly reflecting superior dynamic control of the dominant hand [15]. The lower co-contraction level in the affected dominant arm at 80 bpm may thus be related to its superior timing control as compared to the unaffected non-dominant arm. At 160 bpm, on the other hand, the co-contraction level in the affected forearm was comparable to that in the unaffected forearm just prior to the onset of SDE training. This indicates that the disorder had deprived the dominant hand of its functional superiority over the non-dominant hand. As the rehabilitation proceeded, however, the co-contraction level in the affected forearm gradually decreased and the patient regained the inherent difference in the co-contraction level between the dominant and non-dominant arms. These results provide the objective evidence of decrease in co-contraction associated with functional recovery in dystonic patients. Based on the finding, we suggest that the co-contraction index has potential value in clinical assessment or biofeedback training for focal hand dystonia.

The SDE training also influenced tactile spatial resolution of the

affected fingers. Although the values of spatial resolution can vary depending on the measuring tools employed [14,16,17], we confirmed that the two-point discrimination thresholds of the third, fourth, and fifth digits of the unaffected hand (~5 mm) fell within the normal range of healthy adults with the current method [14,18]. We found that the thresholds of the same digits of the affected hand were approximately twice as high as those of the unaffected hand just prior to the SDE onset. These differences, however, disappeared after three months of training. This improvement in discrimination thresholds seemingly coincided with the improvement in keystroke regularity, suggesting the importance of tactile sense for fine control of finger movements. The results indicate that the SDE training successfully reversed the excessive reorganization in the primary somatosensory cortex [8,9] by allowing the patient to receive distinct sensory inputs from individual fingers.

Overall, the present results clearly demonstrate the effectiveness of SDE training in helping dystonic musicians recover normal motor and somatosensory functions, and importantly, evidence the brain's capacity to reverse sensorimotor reorganization induced by focal hand dystonia through its plasticity. Future research should attempt to associate the behavioral changes observed here with brain activity by using functional neuroimaging in order to fully elucidate the underlying neurological mechanisms.

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