

Setting the stage for prevention and treatment: New therapeutic approaches in musician's dystonia

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Musician's dystonia (MD) is probably the most challenging disorder in musicians' medicine. It has a tremendous negative impact on the careers of affected musicians and is difficult to treat. We have investigated treatment effects of established therapies and new approaches on playing-related motor control in pianists with MD.

Keywords: focal dystonia; musician's dystonia; retraining; botulinum toxin; transcranial direct current stimulation

Focal dystonia in musicians (MD) is a task-specific movement disorder which presents itself as a loss of voluntary motor control in extensively trained movements while the musician is playing the instrument (Altenmüller 2003). For those who are affected, the disorder is highly disabling, and in many cases, it terminates musical careers. According to estimations, 1% of all musicians are affected. Defective inhibition on different levels of the central nervous system is thought to be involved in the pathophysiology (Lin and Hallett 2009). Treatment of patients with MD is still a challenge. Therapeutic options include anticholinergic medication with trihexyphenidyl (Trhx), botulinum toxin (BT) injections, and pedagogical retraining (PR) (Jabusch and Altenmüller 2006). Transcranial direct current stimulation (tDCS) has been shown to modulate excitability of the motor cortex: anodal tDCS facilitated motor learning, and cathodal tDCS improved performance in overlearned tasks

(Nitsche *et al.* 2003, Antal *et al.* 2004). It may be hypothesized that tDCS facilitates retraining effects in musicians with dystonia. In this paper, we present outcome data in 33 pianists with focal dystonia after an average follow-up period of 24 months. Pianists were treated using BT, Trhx, or PR, either as monotherapies or in combination. Treatment monitoring was made using an established protocol that allows assessment of motor control in a relevant musical task. Additionally, short-term development after tDCS-supported retraining in a single patient is reported.

METHOD

Participants

Study 1: 33 pianists with MD (mean age=37 years, range=21-71 yrs, 24 men, 9 women) were included in the follow-up study. *Study 2:* the short-term follow-up study was performed in a 43 year-old male pianist suffering from MD. All patients were diagnosed at the Institute of Music Physiology and Musicians' Medicine, Hanover University of Music and Drama. The diagnostic procedure included a complete neurological examination as well as visual inspection while patients were playing the piano.

Procedure

Assessment of motor control: motor control at the piano was assessed in scale playing because this motor task is early affected during onset of MD. MIDI-based scale analysis was done according to the following protocol (Jabusch *et al.* 2004). Scales were performed with the affected hand on a digital piano that was connected to a computer. Sequences of 10 to 15 C major scales were played over two octaves in both playing directions. Scales were played using the conventional C major fingering. The tempo was standardized and paced by a metronome (one keystroke every 125 ms). The temporary unevenness of inter-onset intervals (IOI) has previously been identified as a valid, reliable, and precise indicator of the impairment of motor control in pianists with dystonia (Jabusch *et al.* 2004). For each participant, temporary unevenness of IOI was analyzed for the affected hand and for both playing directions by calculating the median standard deviations of IOI (mSD-IOI) of all scales. The mSD-IOI score of the more severely affected playing direction was used for further analyses. Motor control was assessed before and after follow-up in Study 1 as well as before and after each treatment condition in Study 2.

Treatment, Study 1: therapeutic approaches, as monotherapies or in simultaneous or successive combination, included the following options: PR was applied in patients who preferred a non-medication treatment approach. PR took place under the supervision of a piano instructor (LB) specialized in dystonia retraining. PR included elements based on the following principles reported previously (e.g. Boulet 2003): (1) movements of affected body parts were limited to a level of tempo and force at which the dystonic movement would not occur, (2) compensatory movements (e.g. of adjacent fingers) were avoided, partially under the application of splints, (3) instant visual feedback with mirrors or monitors helped patients to recognize dystonic and non-dystonic movements. BT injections were applied in patients in whom primary dystonic movements could be clearly distinguished from secondary compensatory movements. Target muscles were identified by visual inspection of the dystonic movement patterns while patients were playing their instruments. A lyophilized botulinum toxin A powder (Dysport®, Ipsen Ltd., Berkshire, UK) was injected using an EMG-guided technique. Trhx was applied as monotherapy when treatment with PR or BT was not desired or possible and no contraindication was present. Adjustment of the dosage was made depending on beneficial effects and side effects. Patients with little response during PR were additionally treated with BT or Trhx when no contraindications were present.

Treatment, Study 2: in a double-blind single case study, tDCS was combined with retraining on the piano that took place according to the aforementioned principles. The patient was treated with three stimulation protocols consecutively, with a minimum of 5 weeks between treatment sessions: anodal tDCS, cathodal tDCS, and placebo stimulation (3x5 days). In the verum stimulation conditions, tDCS (2 mA) was applied for 20 mins on the primary motor cortex contralateral to the affected hand. During stimulation, the patient practiced slow, non-dystonic movement patterns on the piano. Motor control was assessed before and 1 min, 60 mins, 120 mins, and 180 mins after the respective treatment session.

Statistical analyses: Mann-Whitney-U tests were applied to analyze performance differences. The alpha level was set at 0.05. In Study 2, alpha adjustment for multiple testing was made according to Bonferroni-Holm.

RESULTS

Study 1: monotherapies were applied in 23 patients: BT (n=8), Trhx (n=1), PR (n=14). Treatment combinations were applied in five patients: PR+BT (n=3), PR+Trhx (n=2). Five patients refused any treatment. Follow-up monitoring after an average period of 24 months (range=3-57) revealed the

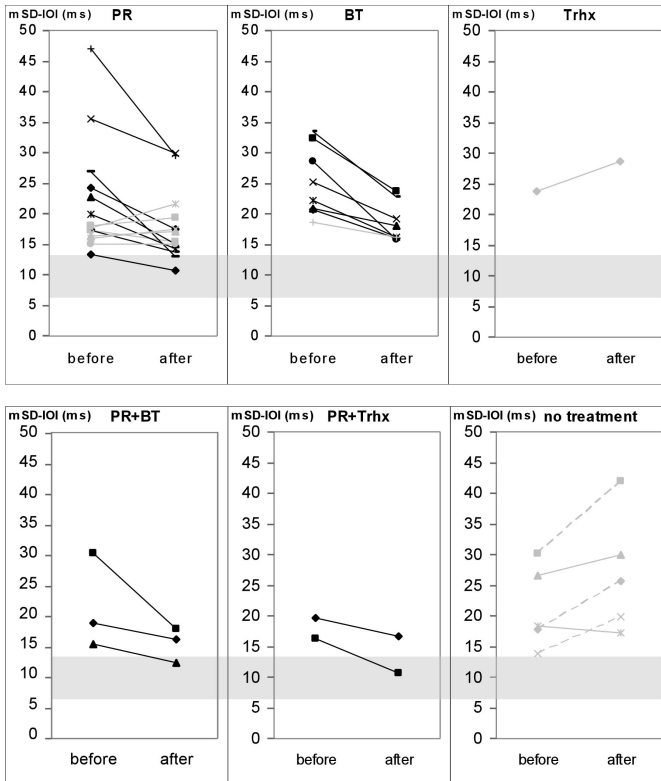


Figure 1. Study 1: each line indicates the mSD-IOI values of one patient before (left endpoint) and after follow-up (right endpoint). A high score for mSD-IOI denotes a high level of temporal unevenness in the scales (impaired motor control), while a lower score for mSD-IOI denotes a lower level of unevenness (less or non-impaired motor control). Solid black lines indicate significant improvement of motor control, solid grey lines indicate no significant change in motor control, dashed grey lines indicate significant deterioration in motor control. Gray window: range of motor control of healthy pianists.

following outcome. A significant improvement of motor performance was seen in 20 patients (71% of the 28 treated patients) after BT (n=7), PR (n=8), PR+BT (n=3), and PR+Trhx (n=2). Four of these patients (14% of all treated patients) returned to a level of normal motor control as seen in healthy pianists (PR: n=2, PR+BT: n=1, PR+Trhx: n=1). No change of motor performance was seen in 8 patients after treatment (BT: n=1, Trhx: n=1, PR: n=6). In the

five untreated patients, motor performance remained unchanged ($n=2$) or deteriorated ($n=3$). Detailed outcome of individual patients is displayed in Figure 1.

Study 2: motor control at baseline did not differ between the stimulation conditions. In the placebo condition, motor control was improved 1 min ($p<0.001$) and 120 mins ($p<0.05$) after treatment as compared with before treatment. In the anodal condition, motor control was improved 1 min ($p<0.001$), 60 mins ($p<0.001$), 120 mins ($p<0.01$), and 180 mins ($p<0.05$) after treatment. In the cathodal condition, motor control was improved 1 min, 60 mins, 120 mins, and 180 mins after treatment (each $p<0.05$). Inter-treatment comparisons revealed a better performance outcome in the cathodal condition compared with placebo 180 mins after treatment ($p<0.001$).

DISCUSSION

The results underline the potential benefit of a behavioral approach in the treatment of pianists with MD. PR as a monotherapy resulted in improved motor control in the majority of patients after follow-up. This result indicates a beneficial effect of an active involvement of patients. As a limiting factor, PR requires patience and compliance of affected pianists. BT injections were successful in all but one of the patients treated with this option. When the muscles involved in the dystonic movements can be clearly identified, BT treatment may be recommended to patients, especially to those who are not able to invest the time and patience required for PR. A treatment attempt with Trhx was unsuccessful in the only patient treated with this option as a monotherapy. Other publications report an improvement rate of 33% in patients with MD who tolerated the medication (Jabusch and Altenmüller 2006). The applicability is, however, limited due to the frequent occurrence of side effects. Treatment combinations such as PR+BT or PR+Trhx are promising, especially in patients with little response to PR alone. In Study 2, retraining resulted in prolonged effects when combined with cathodal tDCS as compared to placebo stimulation. Further studies with large numbers of patients and longer follow-up periods are, however, required to determine the effects of (1) combination therapies based on PR and (2) tDCS-supported retraining. Our results show that the currently available therapies help to improve the situation in the majority of patients. However, they also demonstrate that only exceptionally, affected pianists return to normal motor control.

This observation underlines the crucial importance of a successful prevention. Previous findings suggest that different risk factors such as exagger-

ated perfectionism and anxiety can trigger the manifestation of MD (Jabusch and Altenmüller 2006) in predisposed musicians. These risk factors should be addressed in the musical education from the first lesson onward. Music teachers should strive to create a friendly, supportive atmosphere focusing on creativity, curiosity, and playful experiences in the world of sounds. We assume that the risk of MD may be reduced by including retraining principles even in the teaching process for healthy pianists and by the application of “healthy” practice strategies. Reasonable practice schedules, an economic playing technique, prevention of overuse and pain, mental practice, a variety of movement patterns, maintenance of motivation, avoidance of mechanical repetitions and frustration, sufficient breaks, warm-up and cool-down exercises, regular physical exercise, and sleep are the cornerstones of healthy musical practice. Future projects will have to investigate the preventive potential of these strategies.

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