

Improved motor control after alteration of somatosensory input: Prognostic value of “glove effect” in pianists with dystonia

Hans-Christian Jabusch¹, Jakobine Paulig², Michael Großbach², Laurent Bouillet³, and Eckart Altenmüller²

¹ Institute of Musicians' Medicine, Dresden University of Music
Carl Maria von Weber, Germany

² Institute of Music Physiology and Musicians' Medicine, Hanover University
of Music, Drama, and Media, Germany

³ Department of Music Physiology, International Piano Academy, Lake Como, Italy

Musician's dystonia (MD) impairs motor control and musical performance. Alteration of somatosensory input (e.g. by wearing a latex glove) may improve motor control in some MD patients. The potential association between this so-called sensory trick phenomenon and the outcome after consequent treatment with botulinum toxin and/or pedagogical retraining was assessed in 24 pianists with MD using objective performance measures. Outcomes were significantly better in those patients with a significant pre-treatment sensory trick response (positive glove effect) than in others. The glove effect seems to have a prognostic value in the treatment of patients with MD.

Keywords: musician's dystonia; sensory trick; glove effect; retraining; botulinum toxin

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. MD is still difficult to treat. Therapeutic options mainly include botulinum toxin injections, pedagogical retraining, and anti-cholinergic medication with trihexyphenidyl (Jabusch *et al.* 2005). Until today, the pathophysiology is not fully understood. Defective inhibition on different levels of the central nervous system is discussed to be involved in the

pathophysiology (Lin and Hallett 2009). Maladaptive neuronal plasticity leads to a fusion of the digital representations in the somatosensory cortex of patients (Elbert *et al.* 1998). Evidence for the crucial role of the afferent somatosensory input in the pathophysiology of MD comes from the so-called sensory-trick phenomenon. This phenomenon has been reported in patients with various forms of focal dystonia (e.g. with cervical dystonia): alteration of the somatosensory input by touching the face may reduce or even abolish involuntary muscle activity (Wissel *et al.* 1999, Schramm *et al.* 2004). Analogously, in patients with MD, alteration of somatosensory input (e.g. by wearing a latex glove) may result in short-term improvement of motor control of the affected hand (Altenmüller 2003). According to clinical observations, we hypothesized that the extent of this improvement (“glove effect”) may predict the long-term outcome after treatment in patients with MD.

Pianists represent a large fraction of musicians with dystonia (Altenmüller 2003). For reliable assessment of motor control in pianists with MD and for monitoring of treatment effects in these patients, MIDI- (music instrument digital interface) based scale analysis has been evaluated and established (Jabusch *et al.* 2004). Using this protocol, the present study was designed to investigate the potential association between the “glove-effect” and the long-term outcome after treatment in pianists with dystonia.

METHOD

Participants

Thirty professional pianists (22 men and 8 women; mean age=40.5±13.1 years, range=21.4-68.5 years) suffering from MD were enrolled in the study. Patients were diagnosed at the outpatient clinic of the Institute of Music Physiology and Musicians’ Medicine (IMMM) of the Hanover University of Music, Drama, and Media and underwent complete neurological examination by a neurologist specialized in movement disorders (EA). Dystonic symptoms occurred in the typical manner as painless cramping of one or more fingers while patients were playing the piano. One pianist suffered from MD in both hands. Data from both hands of this patient were included in the statistical analysis as two separate cases.

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, which 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) in 26 patients. Due to severely impaired motor control in four patients, the desired IOI was 187.5 ms in three patients and 250 ms in one patient. 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. Performance tests were carried out separately under three conditions: (1) baseline test before treatment (termed “baseline”), (2) playing with latex glove (sensory trick) before treatment (termed “glove”), and (3) follow-up test after treatment (termed “follow-up”).

Treatment protocol: Therapeutic approaches, as monotherapies or in combination, included the following options: Botulinum toxin injections (BT) were applied in those patients in which 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. Pedagogical retraining (PR) was taking place under the supervision of the piano instructor Laurent Boulet 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 were avoided, partially under the application of splints, and (3) instant visual feedback with mirrors or monitors helped patients to recognize dystonic and non-dystonic movements. Trihexyphenidyl (Trhx) was frequently applied in addition to BT or PR if no contraindication was present. Adjustment of the dosage was made depending on beneficial effects and side effects.

Statistical analyses: Glove-effect and outcome of individual patients were analysed by Mann-Whitney U tests using the respective mSD-IOI values. Group differences were analysed by Mann-Whitney U tests. The alpha level was set at 0.05.

RESULTS

The mean age at onset of dystonic symptoms was 33.5 years; median duration of symptoms at the time of the baseline and glove performance tests was 3 years (range=0.2-44.6). Follow-up tests were conducted on average 4.8 ± 2.5 years after baseline tests. Scale analysis tests revealed the following results: median mSD-IOI_{baseline} was 20.0 ms (range=10.4-58.0); median mSD-IOI_{glove} was 20.3 ms (10.3-56.7); median mSD- IOI_{follow-up} was 21.8 ms (10.4-37.4).

Sensory trick, glove effect: Mann-Whitney U tests showed significant improvement of fine motor control through wearing a glove in six cases (19%) in comparison with the baseline (positive glove-effect [PGE]; all p-values<0.05), significant deterioration in nine cases (29%; all p-values<0.05), and no significant effect in 16 cases (52%; both groups considered as displaying no PGE). The glove-effect (GE) was described as $mSD-IOI_{glove} - mSD-IOI_{baseline}$. Median GE was 1.3 ms (range=-34.1-21.1), negative values indicating improved motor control with the glove compared to baseline.

Details of treatment: Eight patients (27%) were consequently treated with BT (consequent treatment was defined as minimum 3 BT injections); 21 patients (70%) consequently took part in PR (defined as minimum 4 months PR); 7 patients (23%) were consequently treated with Trhx (defined as minimum 4 months Trhx application). None of the patients treated with Trhx showed a PGE; therefore, further statistical analyses were made on patients who were consequently treated either with BT or PR or both (n=24). This group of 24 patients will be referred to as “treated group.” The treated group included five patients with a PGE and 19 patients with no PGE (deterioration with the glove n=6; no differences n=13).

Long-term outcome and its association with glove effect: Mann-Whitney U tests revealed significant improvement of MD symptoms in 12 cases (39%; all p-values<0.05), significant deterioration in seven cases (22%; all p-values<0.05), and no significant change in 12 cases (39%) in the follow-up test. Outcome was defined as $mSD-IOI_{follow-up} - mSD-IOI_{baseline}$ (outcome value). Median outcome value was -2.0 ms (range=-20.6-12.4). In the treated group, patients with a PGE showed significant better outcome values compared with the patients with no PGE (Mann-Whitney U test; p=0.03). Mann-Whitney U tests for therapy details revealed that patients with a PGE and those with no PGE did not differ in the duration of follow-up as well as in treatment details regarding the following variables: duration of treatment (BT), number of injections and dosages (BT), duration of treatment (PR), number of sessions, and intervals between sessions (PR); all p-values were>0.05.

DISCUSSION

The present study investigated the potential association between the sensory trick phenomenon and the outcome after treatment in patients with MD. To provoke the sensory trick, patients played wearing a latex glove on the affected hand. The study focused on affected professional pianists only. The selection of affected professional pianists as study participants warranted a high homogeneity of the sample. Playing with glove, only six patients (19%) showed a statistically significant improvement of motor control compared with baseline. This finding is in agreement with other publications reporting that only a minority of patients with focal dystonia exhibited a reduction of dystonic muscle contractions by using a sensory trick maneuver (e.g. Gómez-Wong *et al.* 1998, Schramm *et al.* 2004).

Patients were treated with currently available treatment options, using BT, PR, and Trhx. Outcome tests after an average follow-up period of 4.8 years indicated that 39% of patients showed a significant improvement while 39% displayed no change of symptoms and 22% had a significant deterioration. The percentage rate of patients with an improvement in the follow-up test is lower in the present study than that in a previous follow-up study showing improvement in 71% of patients (Jabusch *et al.* 2009) using the same performance measures. The following reasons may be responsible for this observation: (1) different samples with only partial overlap were investigated in both studies, (2) sample sizes were relatively small in both studies, (3) performance measures may be distorted in the abovementioned four patients with severely impaired motor control because no adjustment procedure was available to correct for increased target-IOI values, (4) average follow-up duration in the present study was more than twice as long as that of the previous study. A review of the literature on outcome studies reveals that longer follow-up periods seem to be associated with worse outcomes.

The subgroup of patients who were treated consequently (either by a minimum of 3 BT injections or by a minimum of 4 months of PR) displayed an association between the glove effect and the outcome: patients with a PGE showed significant better outcome values compared with the patients with no PGE. This finding strengthens the clinical impression that a positive glove effect has a prognostic value in the treatment of patients with MD. Patients who displayed a significant pre-treatment response to the sensory trick had a better long-term outcome after treatment than those with a little initial sensory trick response. We speculate that a strong response to the sensory trick may indicate that the dystonic movement patterns might be easier to modulate and that therefore treatment might be more effective in these patients.

Further research has to be conducted to clarify if pathophysiological alterations in patients with MD such as blurring of receptive fields in the somatosensory cortex are less pronounced in patients with a stronger sensory trick response.

Address for correspondence

Hans-Christian Jabusch, Institute of Musicians' Medicine, Dresden University of Music Carl Maria von Weber, Wettiner Platz 13, Dresden 01067, Germany; *Email:* jabusch@hfmdd.de

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