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Musician’s dystonia in pianists: Long-term evaluation of retraining and other therapies

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Objective: Musician’s dystonia is characterized by loss of voluntary motor control in extensively trained movements on an instrument. The condition is difficult to treat. This retrospective study reports on the interventions received by a homogeneous cohort of pianists with musician’s dystonia and the subjective and objective changes reported in task performance.

Methods: This is a retrospective descriptive study. Fifty four pianists with musician’s dystonia who had received care in a Movement Disorders Clinic completed a self report questionnaire regarding type and effectiveness of treatment received over the last 4 years. Pianists’ fine motor control was assessed objectively by measuring the temporal regularity of their scale playing.

Results: Nearly all patients (98%) reported deficits in motor tasks other than musical playing. Half of the patients were taking medications (Botulinum toxin (53%), Trihexyphenidyl (51%)). Subjects reported participating in multiple therapies: retraining (87%), hand therapy (42%), relaxation techniques (38%), physiotherapy (30%), psychotherapy (23%), acupuncture (21%) and body techniques (21%). Self-reported improvements in motor performance were reported by 81.5% of the subjects with 5.6% reporting a complete recovery. Objective gains in task-specific motor performance were documented in 42.9% of the subjects (with deterioration in 4.8%). Retraining therapy, relaxation techniques and change in teacher explained 52% of the variance in subjective outcomes.

Conclusions: Musician’s dystonia not only interferes with musical performance but other fine motor tasks. Objectively, approximately 50% of patients improved task performance following participation in a variety of intervention strategies, but subjectively, 80% of subjects reported improvement.

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of ascending and descending scales separately. Then the maximum of these two inter-onset-intervals (IOIs) of the notes and computed the median of these for extracted and the analysis was performed similar to the procedure published pre-

One pianist was eliminated from the objective (scale playing) analysis because the were applied. One pianist was eliminated from the objective (scale playing) analysis due to organizational problems, not all patients could be recorded playing scales following BTX-A injections. Twenty-two pianists (33% of our total population) filled this criterion.

Due to the retrospective nature of this study, patients were informed about the study at the time of the follow-up and anonymity of the data handling was guaranteed. The study was conducted in accordance with the ethical guidelines Standard Protocol Approvals, Registrations, and Patient Consents of the Hannover Medical University.

2. Piano scale playing

For each subject, scale playing regularity was measured at baseline (first visit) and again within an interval of three months before or after the questionnaires were filled out. Patients were invited to play two-octave C-major scales that were recorded on a MIDI piano for analysis using a validated protocol published earlier [16]. In brief, the patients played a total of 10–15 ascending and descending scales with their affected hand, interleaved, accompanied by a metronome at 120 BPM and playing four notes per beat. Three patients could not play at this tempo. These patients played at 80 or 40 BPM. As a consequence of the non-linear dependence of unevenness on tempo, adjustment factors (between 0.52 and 0.89) obtained from healthy pianists’ scale playing performance at different tempi (unpublished data) were applied. One pianist was eliminated from the objective (scale playing) analysis because the first and follow-up measurements were made with different tempi. In offline analysis, the errorfree sequences of ascending and descending scales were extracted and the analysis was performed similar to the procedure published previously in Ref. [16]: For each scale run, we calculated the standard deviation of the inter-onset-intervals (IODs) of the notes and computed the median of these for ascending and descending scales separately. Then the maximum of these two numbers was taken, yielding a single value (except for one patient that was affected in two hands; we took the maximum score of the two hands and calculated our analysis with that). A high value of this quantity means that playing is rather uneven (as would be typical for dystonia patients). A low value means higher precision, that is, better motor control [16].

2.3. Questionnaire

The questionnaire was an extended version of the questionnaire used in a previous study [5]. It covered the following categories:

- Severity of dystonia symptoms — Patients were asked to summarize the development of their dystonia on the following scale: no more impairment, considerable improvement, moderate improvement, minimal improvement, no change, or deterioration. This is referred to as a categorical improvement rating. Additionally, patients were also asked to rate their playing capacity before therapy (retrospectively) as well as their playing capacity today (at fill-out) as a percentage of their playing capacity before they experienced any problems. We will call this the percentual improvement rating.

- Motor deficits in other tasks — Each individual was asked to rate task specific problems on target tasks other than musical performance (e.g. hand writing, computer keyboard typing, using cutlery). The following ordinal scale as used to rate the severity of the impairment: no impairment, light impairment, moderate impairment, strong impairment.

- Therapies received — Each individual was asked to report which therapies they had participated in (see supplementary materials).

- Dosage and effectiveness of therapy — Each individual was asked to indicate the number of months they received each therapy and then to rate the effectiveness on a scale from 1 (very good) to 6 (bad).

Most of the patients returned the form by mail to our institute, whereas 6 out of 54 (11%) brought them back during the next appointment.

2.4. Medical file

For each subject who returned the questionnaire, the medical record was reviewed to obtain information about medication management of the dystonia and to fill in missing data from the questionnaire in terms of the history or the intervention. The following details were extracted from the medical record: (a) which hand was affected by dystonia and (b) how long the patient received BTX-A.

2.5. Statistical analyses — piano scale measurement

We calculated two tests to assess fine motor control objectively. To establish which patients improved significantly, we took the data from each pianist in turn and submitted the standard-deviations of the most impaired playing direction (ascending or descending) for the scales played at first-visit and at fill-out to a Bonferroni-corrected Mann–Whitney Test (alpha = 0.05/22). Further, we calculated improvement in the group as a whole by comparing the medians of the standard deviations for each pianist before and after treatment with the Wilcoxon signed-rank test.

Due to organizational problems, not all patients could be recorded playing scales reasonably close in time to fill-out. We therefore chose to include in our objective analysis only those patients of whom we had a scale measurement within plus/minus 3 months of questionnaire fill-out and (see above) who had not undergone treatment with BTX in between. Twenty-two pianists (33% of our total population) fulfilled this criterion.

2.6. Statistical analyses — questionnaire data

Group comparisons were performed using non-parametric statistics (Wilcoxon rank sum test or Mann–Whitney U). We report the corresponding effect sizes (r). Population statistics are reported as averages (with standard deviation in parentheses), unless specified otherwise. Because nine group comparisons were planned, we used alpha = 0.05/9 – 0.0056. An ordinal logistic regression was computed with categorical improvement rating as dependent variable. Predictors were age, duration of dystonia symptoms at fill-out, follow-up interval, therapies in which the patient participated (0 or 1). For retraining therapy we used the following scale: 0) did not undergo therapy, 1) started retraining, but interrupted, 2) is undergoing treatment until fill-out, 3) completed the retraining therapy program (we will refer to this latter variable as retraining status). We used the Akaika Information Criterion (AIC) as downward variable selection mechanism to avoid collinearity, as implemented in the stepAIC function in the MASS package. First, the optimal set of predictors was determined (i.e. therapies) and then the regressions were calculated with these predictors.

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3. Results

3.1. Description of the population

Our patients (73.6% male) were on average 44.8 (±12.9) years of age. The onset of dystonia symptoms was at 34.6 (±11.4) years. The participants were self-classified as left-handed (11.3%), right-handed (83.0%) or ambidextrous (5.7%). The hand affected by dystonia was left (17.0%), right (79.3%) or both (3.8%). The main instrument was piano (88.7%), organ (9.4%) or accordion (1.9%). The time between first-visit and fill-out was on average 51.6 (±25.4) months. At fill-out the patients had suffered 10.2 (±7.8) years from MD.

3.2. Participation in therapies

The patients had received the following treatments: retraining (87%), Botulinum toxin A (BTX-A) (53%), Trihexyphenidyl (Armatine®) or Parkopan® (51%), hand therapy (42%), relaxation techniques (38%), physiotherapy (30%), psychotherapy (23%), acupuncture (21%), body techniques (e.g. Feldenkrais, Alexander technique, Dis Pokinesis) (21%). Some pianists changed teachers (9%). For those who were treated with Trihexyphenidyl, we consulted the medical records to find they received 2.7 (±1.6) g cumulatively, over the duration of 21.5 (±17.8) months, corresponding to an average of 5.02 (±3.7) mg per day. For those treated with BTX-A, we calculated a standardized cumulative dose as follows: Botox® or Xeomin® units counted as one unit and 4 units of Dysport® counted as one unit, so as to obtain mean equivalent doses published in several review papers [17–20]. Patients in our cohort received an average cumulative dose of 100.6 (±100) such units in 4.1 (±3.0) injection sessions over the duration of 25.4 (±21.8) months, corresponding to an average of 24.1 (±17.6) units per session. Pianists participating in retraining therapy received an average of 41.0 (±29.5) sessions over a period of 38.2 (±38.9) sessions per a period of 38.2 (±38.9) months, which were 1.4 (±1.1) sessions per month.

3.3. Subjective report of improvement

Patients reported minor to considerable improvement (81.5%). Some (5.6%) even indicated they were no longer impaired (see Fig. 1). Participants reported an improvement (percentage of playing capacity at fill-out minus percentage at first visit) of -31% (median 25%, SD 28%). This gain was significant (Wilcoxon signed-rank test V = 44, z = -5.46, p < 0.0001, r = 0.74) (Fig. 2). In a few cases this improvement was to well above their playing level before dystonia onset.

3.4. Objective measure: scale playing evenness

Patients’ scale playing tended to become more regular at the fill-out visit (M = 18.2 ms, median = 17.5, SD = 5.6) relative to the baseline measurement (M = 20.3 ms, median = 17.7, SD = 7.1). However, this gain was only a statistical trend (Wilcoxon signed-rank V = 178, z = 1.67, p = 0.098, r = 0.36). The objective improvement correlated with the categorical improvement rating (Spearman ρ = 0.59, p = 0.003). Single-participant Mann–Whitney tests revealed that 43% of the pianists had improved significantly at fill-out, whereas 4.8% significantly deteriorated and 52% showed no change (Fig. 1).

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It revealed that over the course of four years of simultaneous 
therapy and acupuncture were negative but non-significant: 
the heterogeneous group of pianists suffering from musician's 
dystonia. However, patients tended to positively overestimate the 
development of their symptoms. We propose the following explanation. 
The objective measure was based on scale playing, which is the part 
of music performance most challenging to MD patients. However,
participation in various commonly applied therapies, patients 
reported considerable improvement in their symptoms (79.2%). 
However, among those for whom scale playing measurements 
were available, only 49% improved objectively. The present results indicate that retraining therapy ranks among the established therapies in terms of clinical importance. Retraining therapy is attractive because of its non-invasive nature. It is surprising that BTX-A did not appear as a predictor of subjective outcome. This can be only partially explained by the possibility that patients who responded less well to retraining turned to BTX-A afterwards. In fact, of the patients receiving retraining and BTX-A, only 42% started BTX-A after retraining, whereas 35% received BTX-A first and then participated in retraining. Finally, 24% participated in both.

The findings from this study are consistent with previous reports where 54% of a heterogeneous group of musicians with dystonia reported an improvement [5]. On the other hand, in another study, following sensory retuning up to 70% of the participants reported significant improvement [21]. Similar improvements were found applying BTX-A [4] or sensory training [22]. In another study, 12 out of 20 pianists using a “slow down technique” [23] reported a marked improvement (60%) and 8 a mild improvement [23] (40%). Unfortunately, the studies mentioned above applied different outcome measures and are therefore not directly comparable [24].

Objectively measured scale playing evenness revealed a more modest improvement than objective measures in other studies, such as the “Frequency of Abnormal Movement scale” (FAM [10]) or the “Finger Dexterity Scale” (FDS [24]). While perhaps more objective than the self-rated improvement scales, the latter objective outcome measures were not comparable to ours. It is not clear which of the objective measures have the most immediate relevance to the quality of piano playing. We found a significant correlation between the objectively measured improvement and the categorical subjective rating. However, patients tended to positively overestimate the development of their symptoms. We propose the following explanation. The objective measure was based on scale playing, which is the part of music performance most challenging to MD patients.

3.6. Impairment in other motor tasks

Four subjects did not fill out this section and could not be included. Ninety-eight percent of patients reported at least a minor deficit in at least one motor task other than playing their musical instrument. This was true both before and after participating in intervention, with 24.0% of the patients reporting at least one severe secondary impairment before therapy compared to 12.0% afterwards. This difference was not significant (Fisher Exact Test, \( p = 0.19 \), n.s.).

The main motor impairments were in using a computer keyboard (44.5%) and hand writing (38.9%). There was only a trend for these numbers to decrease at follow-up (31.5% for keyboard, Fisher Exact Test, \( p = 0.01 \), not significant; and 27.8% for hand writing, \( p = 0.87 \), not significant).

3.7. Therapy participation as predictor of subjective improvement

Ordinal logistic regression using the predictors follow-up interval, retraining status, relaxation techniques, psychotherapy, acupuncture and teacher change yielded a model with a moderately good fit (\( R^2 = 0.52 \), model likelihood ratio test \( \chi^2(6) = 45.62, p < 0.0001 \) (Fig. 4). Retraining status (\( p = 0.0002 \)), relaxation techniques (\( p = 0.008 \)) and changing teacher (\( p = 0.008 \)) were all positive predictors of the subjectively rated outcome. Psychotherapy and acupuncture were negative but non-significant predictors.

4. Discussion

This retrospective study was designed to evaluate the subjective and objective effects of established therapies for the treatment of a homogenous group of pianists suffering from musician's dystonia. It revealed that over the course of four years of simultaneous participation in various commonly applied therapies, patients reported considerable improvement in their symptoms (79.2%). However, among those for whom scale playing measurements were available, only 49% improved objectively.

The present results indicate that retraining therapy ranks among the established therapies in terms of clinical importance. Retraining therapy is attractive because of its non-invasive nature. It is surprising that BTX-A did not appear as a predictor of subjective outcome. This can be only partially explained by the possibility that patients who responded less well to retraining turned to BTX-A afterwards. In fact, of the patients receiving retraining and BTX-A, only 42% started BTX-A after retraining, whereas 35% received BTX-A first and then participated in retraining. Finally, 24% participated in both.
in practice, patients tend to shift their selection of music materials away from those that provoke dystonic movements (e.g., music containing scales). As a result, they may feel less impaired than they really are. Additionally, patients’ positive overestimation may have reflected a particular attitude among musicians and their perceived self-control through active involvement in the treatment process. This discrepancy between subjective and objective measures underlies the need to establish objective measures of impairment in MD in order to better monitor therapy outcome.

In keeping with other reports, this study revealed the presence of motor control problems at tasks other than musical performance in the majority of patients. Although MD has traditionally been termed ”task-specific”, several studies reported deficits in additional, non-musical motor tasks [25]. In one study, symptoms not linked to the playing were found in 53% of the musician patients [7]. In our study, non-music related symptoms were reduced at fill-out relative to the first visit. This suggests that the therapies addressing MD also improved performance in other fine motor tasks. This could reflect some general organizational structure of the human motor system [26].

This study had some limitations. It included a small number of subjects. This may limit the interpretation of findings particularly based on multiple regression analysis. By design, this is a descriptive retrospective study and required subjects to reflect back on performance issues as long as four years earlier. There was no control for the amount of time a subject participated in each of the therapies. Objective measurements of task specific motor performance were only gathered on a subsample of subjects (less than 50% of the total number of subjects included in this study). The subjective measurement scales were not previously validated, which might explain some of the discordances between objective and subjective measures. Retraining in this study meant pedagogy and appropriate teaching by a music coach. However, retraining can also refer to retraining of the brain following the principles of neuroplasticity. This type of training could also be the focus in physiotherapy, occupational or hand therapy. In the present study, all physiotherapy and all hand therapy were assumed to include the same type of treatment. It is possible, however, that there may be significant differences in these interventions, some focusing on coaching the patient through sensory, motor or sensorimotor retraining rather than simple posture, flexibility and strengthening. Therefore the present findings must be interpreted cautiously.

In conclusion, this retrospective, descriptive study provides a perspective on the rehabilitation of pianists with musician’s dystonia. Pianists with a history of dystonic cramps are likely to have difficulty with comparable fine motor tasks in other contexts. Pianists with MD are likely to improve in performance after participating in a variety of rehabilitative interventions. In this four-year retrospective analysis, affected musicians had a more positive impression of their recovery of task specific motor control compared to objectively documented gains of performance values.

Conflict of interest

The authors declare no conflict of interest.

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Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.parkreldis.2013.08.009.

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