A Combination of Constraint-induced Therapy and Motor Control Retraining in the Treatment of Focal Hand Dystonia in Musicians
A Long-term Follow-up Study

Patrice Berque* BSc (Hons) MMACP MCSP, Heather Gray§ ProfD MSc BSc MCSP, Angus McFadyenδ PhD

* Glasgow Royal Infirmary, Department of Physiotherapy, Alexandra Parade, Glasgow G31 2ER

§ School of Health & Life Sciences, Glasgow Caledonian University, Cowcaddens Road, Glasgow G4 0BA

δ Statistical Consultant, AKM-STATS. Formerly Reader in Health Statistics, Glasgow Caledonian University, Glasgow G4 0BA

Introduction

Focal hand dystonia (FHD) in musicians is a painless task-specific motor disorder characterized by involuntary loss of control of individual finger movements (Fig. 1). It is associated with decreased cortical inhibition, and maladaptive cortical reorganization showing fusion of the representational zones of the digits in the primary somatosensory cortex.[1] Research investigating the long-term effects of rehabilitation strategies for FHD is lacking.

Aims

The aim of this study was to investigate the long-term effects of a combined behavioural therapy intervention, aimed at normalising finger movement patterns.

Methods

- Eight musicians with FHD volunteered to take part. Six musicians completed the initial 1-year period, and four agreed to participate in the 4-year follow-up.
- Inclusion and exclusion criteria were checked again, and all four participants met them.
- One year of intensive constraint-induced therapy (Fig. 1) and motor control retraining at slow speed were the interventions.[2]
- Participants would practice their specific exercises between one and two hours each day during the initial intensive 1-year period.
- Participants were no longer monitored between the end of the intensive 1-year period and follow-up at year 4.
- A quasi-experimental repeated measures (within-subject) design was used, with 9 testing sessions over 4 years.
- Video recordings of the subjects playing two pieces of music were used for data analysis.
- The **Frequency of Abnormal Movements scale (FAM)**,[3] and two ordinal **Dystonia Evaluation Scales (DES)**[4,5] were chosen as outcome measures.
- It was hypothesized that there would be significant differences in FAM scores and DES scores achieved over the 4-year period.

FIGURE 1. Dystonic patterns and constraint-induced therapy

Results

1. Reliability Testing of the Outcome Measures

Intra-rater, inter-rater, and test-retest reliability were tested using an intraclass correlation model: ICC Model (2,1).[6]

FAM Scale:

- **Inter-rater reliability:** Excellent with ICC = 0.985-0.999, $p < 0.001$, narrow 95% confidence intervals (CI = 0.985 – 1.000).
- **Test-retest reliability:** Good to very good, with ICC = 0.739 – 0.996, majority with $p < 0.001$, but wider 95% CI.

Dystonia Evaluation Scales:

- **Intra-rater reliability:** ICC = 0.700 – 1.000 with fairly narrow 95% CI (0.600 – 0.999).
- **Inter-rater reliability:** ICC = 0.760 – 0.900 with reasonable 95% CI (0.390 – 0.990).

2. FAM Scale Scores: Comparison over Time

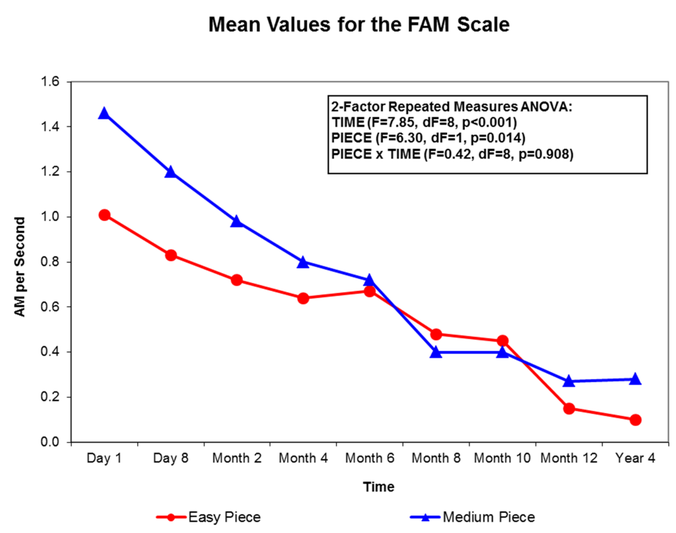

FIGURE 2. Frequency of abnormal movements (FAM) scale: mean values for each piece and all subjects.

- The results from the two-factor repeated measures ANOVA for the main outcome measure, the FAM scale scores, revealed that the mean number of abnormal movements (AM) per second of instrumental playing decreased significantly by approximately 80% for both pieces over the 4-year period ($F = 7.85$, $df = 8$, $p < 0.001$)(Fig. 2).
- Tukey's post-hoc test revealed that statistically significant improvements occurred after 6 months of therapy (p-values between $p < 0.001$ and $p = 0.044$).
- Although the results were not significant between month 12 and follow-up at year 4, the trend revealed that the progress achieved during the first year of intensive retraining was maintained at year 4.

3. Dystonia Evaluation Scale Scores

- The results of the one-factor repeated measures ANOVA carried out for each DES revealed a statistically significant improvement in scores for both the Tubiana and Chamagne Scale[4] ($F = 7.18$, $df = 8$, $p < 0.001$) and the Arm Dystonia Disability Scale[5] ($F = 5.26$, $df = 8$, $p < 0.001$) over the 4-year treatment period, in keeping with the trends observed for the FAM scale.

Conclusions & Recommendations

- **A 1-year intensive retraining protocol** combining constraint-induced therapy and motor control retraining may lead to **long-term benefits** for musicians with FHD, with a significant trend towards **normalisation of movement patterns** over time.
- These results are very encouraging, and suggest that normalization of movement patterns and recovery of fine motor control occur through normalization of the cortical representational maps, and that these positive neuroplasticity changes are maintained in the long-term.
- Indeed, the progress achieved during the first year of intensive retraining was maintained at the 4-year follow-up with a minimum amount of daily specific practice, i.e. 15 to 30 minutes on average.
- Results also suggest that **retraining strategies may need to be carried out for at least 6 months** before statistically significant changes are noted.
- The **FAM scale** proves to be a **useful and reliable clinical tool** for measuring musician's dystonia.
- The results from the reliability tests carried out for the **Dystonia Evaluation Scales** are encouraging, and show that **these rating scales are reliable tools**. However, further research is necessary to evaluate their validity.

References

1- Elbert, T., Candia, V., Altenmüller, E., Rau, H., Sterr, A., Rockstroh, B., Pantev, C., Taub, E. 1998, "Alteration of digital representations in somatosensory cortex in focal hand dystonia.", *Neuroreport*, Vol. 9, pp. 3571-3575.

2- Berque, P., Gray, H., Harkness, C., McFadyen, A. 2010, "A combination of constraint-induced therapy and motor control retraining in the treatment of focal hand dystonia in musicians.", *Medical Problems of Performing Artists*, Vol. 25(4), pp. 149-161.

3- Spector, J.T., Brandfonbrener ,A.G. 2005, " A new method for quantification of musician's dystonia - the frequency of abnormal movements scale", *Medical Problems of Performing Artists*, Vol. 20, pp. 157-162.

4- Tubiana, R. 2000, "Musician's focal dystonia". in: *Medical Problems of the Instrumentalist Musician*, ed. R. Tubiana & P.C. Amadio, Martin Dunitz, London, p.340.

5- Fahn, S. 1989, "Assessment of the primary dystonias." in: *Quantification of Neurologic Deficit*, ed. T.L. Munsat, Butterworths, Boston, pp. 241-270.

6- Fleiss, J.L., Cohen, J. 1973, "The equivalence of weighted kappa and the intraclass correlation coefficient as measures of reliability", *Educational and Psychological Measurement*, Vol. 33, pp. 613-619.

Acknowledgements

- The study was approved by the Research Ethics Committee of the School of Health and Life Sciences at Glasgow Caledonian University.
- The authors want to thank Physiofirst for their contribution towards the costs associated with the completion of this study.

Further Information

Contacts: patrice.berque@btinternet.com; h.gray@gcu.ac.uk

Presented at Physiotherapy UK 2012, Liverpool, October 2012