

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

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Focal Hand Dystonia

(Elbert et al. 1998, Lederman 2002, Schuele & Lederman 2003, Brandfonbrener et al. 2004, Frucht 2004, Lim et al. 2004, Conti et al. 2008, Frucht 2009, Altenmüller & Jabusch 2010)

- Painless motor disorder.
- Involuntary loss of fine motor control and coordination of individual finger movements.
- Deterioration of sensorimotor skills, task-specific.
- Usually involving 3rd to 5th digits.
- Estimated prevalence of less than 1% of the population of professional musicians.

Focal Hand Dystonia – Neurological Changes

(Ikoma et al 1996, Elbert et al. 1998, Hallett 1998, Bara-Jimenez et al. 2000, Hallett 2004, Rosenkranz et al 2005, Lin & Hallett 2009, Altenmüller & Jabusch 2010)

- Reduced inhibition and increased excitation at spinal cord, brainstem, and cortical levels, leading to excessive motor output with overflow into inappropriate muscles.
- This would explain co-contraction of agonist and antagonist muscles observed in FHD.
- Altered sensory perception;
- Impaired sensorimotor integration;
- maladaptive cortical plasticity.

Fusion of Cortical Representations

(Elbert et al., 1998)

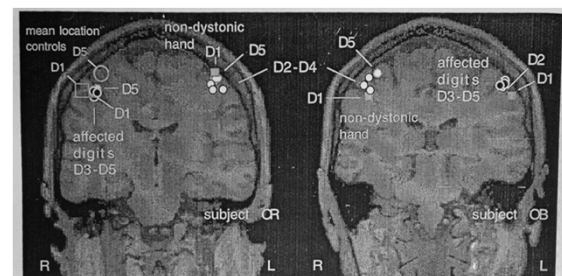


FIG. 2. A coronal MRI section through the somatosensory cortices of 2 musicians (OR and OB) with focal hand dystonia onto which are projected the dipole locations of digits 1-5 (D1-D5) resulting from contralateral stimulation. The large open symbols in the right hemisphere of subject OR indicate the mean location of dipoles for D1 and D5 in normal control subjects.

FHD – Management Strategies

- Limb immobilisation (Priori et al., 2001);
- Learning-based sensory training (Byl et al., 2009);
- Sensory retraining – Braille reading (Zeuner et al., 2002);
- Proprioceptive retraining (Rosenkranz et al., 2009);
- Constraint-induced therapy (Candia et al., 2002);
- Motor Control Retraining – “Slow-Down Exercise” (Sakai, 2006).

AIMS

- Investigate the long-term effects of a combined behavioural therapy in musicians affected by FHD, 3 years after completion of the initial 12-month study = 4-year follow-up:
 - Constraint-induced therapy.
 - Motor control retraining (Slow-Down Exercise).
- Subsidiary aim: reliability study of the outcome measures: ADDS, TCS, FAM scales.

A Combination of Constraint-induced Therapy and Motor Control Retraining in the Treatment of Focal Hand Dystonia in Musicians

Patrice Berque, BS (Hons), MCSP, Heather Gray, MS, MCSP, Cassandra Harkness, BS, MCSP, and Angus McFadyen, PhD

Focal hand dystonia (FHD) in musicians is a painful task-specific motor disorder characterized by an involuntary loss of control of individual finger movements. The aim of this study was to investigate the effects of an innovative behavioral therapy intervention, aimed at normalizing movement patterns, in musicians affected by FHD. Methods: Eight musicians volunteered to take part in this retraining protocol. Intensive constraint-induced therapy and motor control retraining at slow speed were the interventions. Video recording of the subjects playing two pieces were used for data analysis. The Frequency of Abnormal Movements scale (FAM), the change in metronome speed achieved during motor control retraining, and two ordinal dystonia evaluation scales were chosen as outcome measures. It was hypothesized that there would be significant differences in the FAM scores and metronome speeds over a 12-month period. Results: For the main outcome measure, the FAM scale scores, the two-factor repeated measures ANOVA revealed a very significant decrease in the number of abnormal movements per second of instrumental playing over the 12-month period ($F = 6.32$, $df = 7$, $p < 0.001$). Tukey's posthoc tests carried out for the FAM scores revealed that significant changes occurred after 8 months of therapy. Discussion: These results suggest that a combination of constraint-induced therapy and specific motor control retraining may be a successful strategy for the treatment of musicians' FHD. Furthermore, the results suggest that retraining strategies may need to be carried out for at least 8 months before statistically significant changes are noted. *Med Probl Perform Art* 2010; 25:139-153.

untary loss of control and coordination of individual finger movements.^{1,2} It is a disorder associated with a sudden or insidious deterioration of sensorimotor skills which, in most cases, only occurs in the context of instrument playing.^{3,7} Involuntary spasms, cramping sensations, abnormal hand posture, finger curling, loss of coordination during specific fingerings, fingers sticking on the keys of the instrument, irregularities in rhythm and tempi are common findings.^{3,5,8} It more often involves digits 3, 4, and 5 (D3 to D5) of the hand^{3,9,10} and is thought to be related to the intense and prolonged practice of rapid, alternating, and highly precise finger movement patterns.^{2,4,10} The condition can be disabling enough to curtail a professional career.^{3,10,11}

Prevalence Amongst Musicians

FTSD has been estimated to affect between 5% and 14% of musicians consulting performing arts clinics in the US.^{11,12} This would give an estimated prevalence of 0.2% to 0.5% in the population of professional musicians.^{1,10} Focal hand dystonia (FHD) occurs much more frequently in males than females. One study¹³ revealed that 73% of instrumentalists affected were men, two others reported 80%¹⁴ and 83%¹⁵. Symptoms usually begin in the third or fourth decade.^{4,10,11}

LITERATURE REVIEW

Subjects

Instrument	Dystonia	Side	Onset	Compliance
Guitar 1	D3, D4, D5	R	2006	95%
Guitar 2	D3, D4, D5	R	1982	76%
Flute 1	D4, D5	L	2002 (D5) 2006 (D4)	95%
Flute 2	D4, D5	R	2004	95%
Piper 1	D5	R	2005	77%
Piper 2	D3, D4	R	1995	40%
Oboe	D4, D5	R	2006	88%
Accordeon	D3, Wrist, D2, D4	R	2005	N/A

Subjects



Outcome Measures

- 2 test pieces: easy and medium difficulty;
- Frequency of Abnormal Movements (FAM) scale (Spector & Brandfonbrener, 2005);
- 2 ordinal Dystonia Evaluation Scales:
 - Tubiana & Chamagne Scale (TCS),
 - Arm Dystonia Disability Scale (ADDS);
- Change in metronome speed achieved during Slow-Down Exercise (Sakai, 2006).

Hypothesis

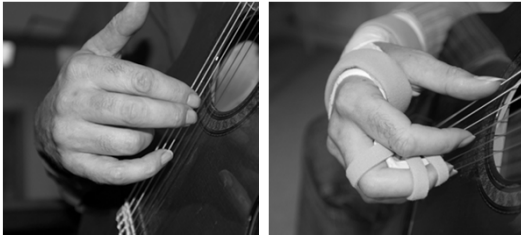
Significant differences in FAM scores, ordinal scale scores, and metronome speeds at the 4-year follow-up.

Study Design

- Repeated Measures Design: subjects tested at Day 1, Day 8, then every 2 months up to Month 12; and Year 4.
- Standardised protocol;
- Standardised metronome speed for each piece.

Constraint-Induced Therapy

(Berque et al., 2010)



Guitar Player – Day 1

(Berque et al., 2010)



Home Protocol

- Week 1: constraint-induced therapy only. 2 hours per day;
- Constraint-induced: ½ hour to 1 hour per day;
- Slow-Down Exercise: ½ hour per day;
- No monitoring of subjects between Month 12 and Year 4.

Outcome Measures – Reliability

(Spector & Brandfonbrener 2005, Spector & Brandfonbrener, 2007)

- Lack of evaluation of the clinical utility of common outcome measures used in studies on FHD:
 - TCS never evaluated for reliability.
 - ADDS evaluated in one study only (Spector & Brandfonbrener, 2005).
 - FAM developed by Spector & Brandfonbrener and evaluated in their study (Spector & Brandfonbrener, 2005).

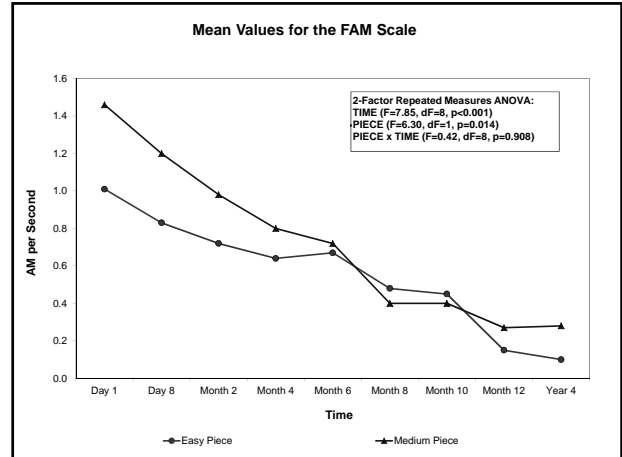
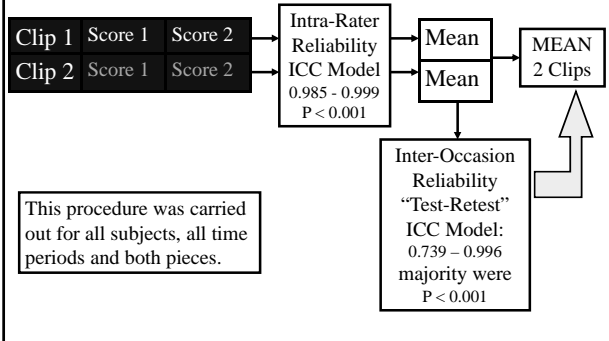
TCS & ADDS – Reliability Tests

Test	ICC	p-value	95% CI
Intra-rater (PB)			
Day 1 TCS	0.70	0.023	0.04-0.93
Day 1 ADDS	0.92	<0.001	0.67-0.98
Month 2 TCS	0.90	<0.001	0.60-0.98
Month 2 ADDS	1.00	<0.001	-
Intra-rater (RH)			
Day 1 TCS	0.76	0.010	0.20-0.95
Day 1 ADDS	0.93	<0.001	0.72-0.99
Month 2 TCS	0.90	<0.001	0.60-0.98
Month 2 ADDS	0.94	<0.001	0.73-0.99
Inter-rater			
Day 1 TCS	0.90	<0.001	0.39-0.98
Day 1 ADDS	0.88	0.001	0.55-0.98
Month 2 TCS	0.76	0.003	0.16-0.95
Month 2 ADDS	0.83	0.003	0.40-0.96

TCS – Reliability Tests

TCS	Stage Definition
Stage 0	Unable to play
Stage 1	Plays several notes but stops because of blockage or lack of facility
Stage 2	Plays short sequences without rapidity and with unsteady fingering
Stage 3	Plays easy pieces but is unable to perform more technically challenging pieces
Stage 4	Plays almost normally but difficult passages are avoided for fear of motor problems
Stage 5	Returns to concert performances

FAM – Reliability Tests



Practice Profile

(adapted from Ackermann & Driscoll, 2010)

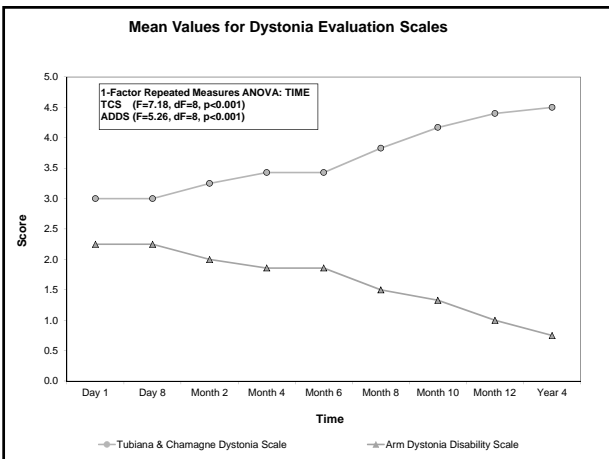
Question	S. 1	S. 2	S. 3	S. 4
On average, how many days per week did you practise your specific exercises?	5	4	4	6
On average, how many practice sessions would you normally do per day for your specific exercises?	1	1	1	2
How long have your average practice sessions been for your specific exercises: - Less than 15 minutes? - Between 15 minutes and half an hour? - Between half an hour and one hour?	√	√	√	√

Guitar Player – Month 12

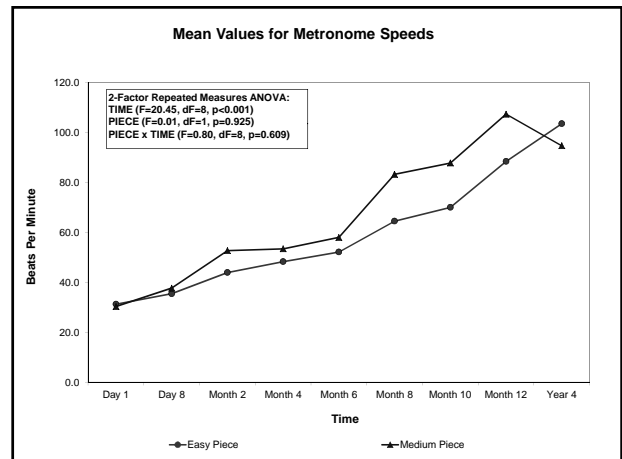
(Berque et al., 2010)



Mean Values for Dystonia Evaluation Scales



Mean Values for Metronome Speeds



Limitations

- No control group;
- Small sample;
- Skewed follow-up results for the medium difficulty piece;
- Two strategies were used.

Clinical Recommendations

- A 1-year retraining protocol may lead to long-term benefits for musicians with FHD;
- Progress maintained with only 15 to 30 minutes of daily specific practice;
- Intensive retraining for more than 6 months;
- The FAM scale is a useful and valid clinical tool;
- The TCS and ADDS showed good to very good intra- and inter- reliability.

Co-authors

- Heather Gray, Senior Lecturer, Glasgow Caledonian University.
- Angus McFadyen, Statistical Consultant, formerly Reader in Health Statistics at Glasgow Caledonian University.