

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), MCSP, Heather Gray, Prof D, MSc, MCSP, and Angus McFadyen, PhD

Focal hand dystonia (FHD) in musicians is a painless, task-specific motor disorder characterized by involuntary loss of control of individual finger movements. The aim of this study was to investigate the long-term effects of a combined behavioral therapy intervention aimed at normalizing finger movement patterns. **METHODS:** Eight musicians with FHD had taken part in the 1-year study involving intensive constraint-induced therapy and motor control retraining at slow speed as the interventions. Four of these subjects volunteered to take part in this 4-year follow-up. A quasi-experimental, repeated measures design was used, with 9 testing sessions over 4 years. Video recordings of the subjects playing two pieces were used for data analysis. The Frequency of Abnormal Movements scale (FAM) was the main outcome measure. It was hypothesized that there would be significant differences in FAM scores achieved over the 4-year period. **RESULTS:** The results from the ANOVA revealed a significant decrease, by approximately 80%, in the number of abnormal movements for both pieces over the 4-year period ($F=7.85$, $df=8$, $p<0.001$). Tukey's post-hoc test revealed that 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. **CONCLUSIONS:** A 1-year period of intensive task-specific retraining may be a successful strategy with long-term, lasting effects for the treatment of musician's FHD. Results suggest that retraining strategies may need to be carried out for at least 6 months before statistically significant changes are noted. *Med Probl Perform Art* 2013; 28(1):33-46.

Mr. Berque is Specialist Musculoskeletal Chartered Physiotherapist, Department of Physiotherapy, Glasgow Royal Infirmary; Dr. Gray is Senior Lecturer in Physiotherapy, School of Health and Life Sciences, Glasgow Caledonian University, and Research Associate, Institute of Health & Wellbeing, University of Glasgow; and Dr. McFadyen is Statistical Consultant, AKM-STATS, Formerly Reader in Health Statistics at Glasgow Caledonian University, Glasgow, Scotland.

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Address correspondence to: Mr. Patrice Berque, 4 Carlston Steadings, Campsie Road, Torrance, Glasgow G64 4ED, Scotland, UK. Tel +44 1360 621896. patrice.berque@btinternet.com.

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Focal task-specific dystonia (FTSD) affecting musicians' hands (also termed focal hand dystonia, FHD) is a task-specific, painless motor disorder characterized by an involuntary loss of control and coordination of individual finger movements, accompanied by abnormal co-contractions of agonist and antagonist muscles.¹⁻⁴ In most cases, it only occurs in the context of instrument playing, more often involves digits 3, 4, and 5 (D3 to D5) of the hand, and is thought to be related to the intense and prolonged practice of highly skilled movement patterns. The hand that performs the most complex movement patterns on the instrument is usually affected: the right hand of pianists, the right hand of guitarists, the left hand of flautists, and the left hand of string players.³⁻¹¹

FTSD has been reported to affect 0.2% to 0.5% of professional musicians, and males are much more affected than females.^{3,4,6,10-13} The condition can be disabling enough to curtail a professional career.^{7,9,12,13}

LITERATURE REVIEW

Neurophysiology of Focal Dystonia

Extensive research on the neurophysiology of FTSD has been carried out in recent years and has identified a wide range of abnormalities throughout the central nervous system. Findings revealed reduced inhibition in the sensory cortex;¹⁴⁻¹⁷ altered sensory perception;^{4,18,19} abnormal motor preparation and increased motor cortex excitability;^{20,21} impaired sensory-motor integration, whereby experimentally induced proprioceptive stimulation (muscle vibration) to individual hand muscles resulted in increased excitability of cortical projections to all neighboring hand muscles in musician's dystonia, whereas excitability was reduced in neighboring non-vibrated hand muscles of healthy non-musicians;^{3,4,22-25} and maladaptive cortical plasticity.^{2-4,17,26}

Indeed, Elbert et al.,² using magnetoencephalography (MEG) and magnetic resonance imaging (MRI) in a study involving a small sample of eight musicians affected by FHD, eight non-affected musicians, and a control group of non-musicians, revealed a significantly reduced distance or fusion of the cortical representations of the digits in the primary somatosensory cortex (S1) for the affected hand of FHD musicians, compared to non-dystonic musicians and controls. Sim-

TABLE 1. Ordinal Dystonia Scales Used to Monitor Effects of Treatment

TCS ³⁰	Stage Definition	ADDS ²⁹	Stage Definition
Stage 0	Unable to play	Stage 0	No dystonia
Stage 1	Plays several notes but stops because of blockage or lack of facility	Stage 1	Mild difficulty playing
Stage 2	Plays short sequences without rapidity and with unsteady fingering	Stage 2	Moderate difficulty playing
Stage 3	Plays easy pieces but is unable to perform more technically challenging pieces	Stage 3	Marked difficulty playing
Stage 4	Plays almost normally but difficult passages are avoided for fear of motor problems		
Stage 5	Returns to concert performances		

TCS, Tubiana and Chamagne dystonia scale; ADDS, Arm Dystonia Disability Scale.

ilar findings emerged from a study by Bara-Jimenez et al.,²⁶ who suggested that this maladaptive cortical reorganization may be linked to altered sensory perception with abnormalities in temporal and spatial discrimination, which they demonstrated in individuals affected by writer's cramp.^{18,19}

Furthermore, there is considerable evidence that FTSD is associated with functional disturbances of the basal ganglia, whereby a reduction in the influence of the indirect pathway (inhibitory) would lead to overactivity of the direct pathway (excitatory) and, therefore, abnormal motor output with overflow into inappropriate muscles.¹⁵⁻¹⁷

Instruments to Measure Musician's Dystonia

Ordinal dystonia evaluation scales (DES) have been developed and validated, although they were not originally designed to measure musician's dystonia.²⁷ The Burke-Fahn-Marsden scale (BFM) had been studied and considered as valid (Spearman's $r=0.89-0.98$, $p<0.01$), showing good intra-rater (Spearman's $r=0.98$) and inter-rater (Spearman's $r=0.85-0.96$) reliability ($p<0.01$).²⁸ Moreover, the same authors²⁹ developed additional scales to allow a more specific and sensitive assessment of focal dystonias. The arm dystonia disability scale (ADDS)²⁹ is one of those and includes a subsection dealing with playing a musical instrument (Table 1). The Tubiana and Chamagne scale (TCS)³⁰ was written specifically to assess musicians and has the highest range of values (0 to 5), but it has never been validated (Table 1).

Measurement is an important aspect of healthcare to establish the impact of treatment and monitor change. Validity is an indicator of whether the outcome measure is appropriate for its intended purpose, with reliability being an indicator that the measurement is reproducible.³¹ Although commonly used in several studies on musician's dystonia^{25,27,32-36} and writer's cramp,^{37,38} these ordinal scales have been evaluated for intra- and inter-reliability in one study only. In a systematic review of methods of evaluation of musician's dystonia, Spector and Brandfonbrener³⁹ confirmed that neither the TCS nor ADDS had been evaluated for reliability. In a concurrent study, Spector and Brandfonbrener²⁷ showed that the BFM had good intra-rater (ICC=0.82) and inter-rater (Spearman's $r=0.76$) reliability. The ADDS also showed good intra-rater (ICC=0.81) and acceptable inter-rater (Spearman's $r=0.68$) reliability. However, their inter-rater reliability results must be taken with care, given that

correlation (Spearman's ρ) is a measure of association and does not always indicate actual agreement.

The difficulty in relying on ordinal scales to measure FHD has motivated research into the development of objective measures for the quantitative evaluation of musician's dystonia.

Using an experimental design, Jabusch et al.³³ developed a MIDI-based scale analysis software to measure objectively the extent of the disorder in eight pianists affected by FHD. The software was able to analyze irregularities in loudness and different timing parameters. Test-retest reliability had been ascertained in a pilot study and found to be excellent (intra-class correlation coefficients ICC=0.94-0.98). The eight subjects were matched for age, gender, and handedness with eight healthy pianists. Owing to the small sample size, non-parametric tests were carried out. The results revealed statistically significant differences between affected hands (FHD subjects) and reference hands (healthy subjects) between the two groups. This was also reflected by a high correlation between mean ADDS scores and the MIDI-based results. Between-group analysis of the non-affected hands did not reveal any significant differences. Finally, significant differences in each group between both hands were only observed in pianists with dystonia. The authors concluded that the tool was effective and reliable for the quantification of FHD in pianists. Unfortunately, this tool is only available for pianists and requires specific equipment, not easily available.³⁹

Spector and Brandfonbrener²⁷ developed an instrument that could be applied to all types of instrumentalists. The frequency of abnormal movements scale (FAM) was used as an objective measure in which the number of abnormal movements of the main dystonic finger and the other affected fingers could be identified by objective raters. A digital video camera was used to videotape subjects playing a musical piece for five minutes, and the number of abnormal movements was counted over the 5-minute window. The FAM score was therefore the number of abnormal movements per second of instrumental playing. Participants underwent constraint-induced therapy treatment, as described by Candia et al.⁴⁰ The authors used a quasi-experimental study, and subjects were monitored at day 1 (baseline measure), week 1, and month 6 of the retraining program. Detailed information on the protocol, tests of intra- and inter-reliability, and blinding of the assessors allowed the monitoring of some aspects of internal and external validity. The FAM scale was compared to the BFM and ADDS and showed high intra- and inter-

rater reliability (ICC=0.92–0.96, Spearman's $r=0.87$ – 0.90), high internal consistency (Cronbach's $\alpha=0.96$), high concordance (weighted kappa=0.94), it approached statistical significance regarding responsiveness to change ($p=0.06$), and it proved superior to the BFM and ADDS with regards to all these statistical parameters. Spector and Brandfonbrener²⁷ concluded that ordinal scales give a global clinical impression of the severity of musician's dystonia, whereas the FAM scale provides an objective quantification of the frequency of abnormal movements, being therefore more specific. However, the same reservations mentioned earlier with regards to using Spearman's ρ for inter-rater reliability would apply to the FAM results.

The FAM was further validated by the present authors,³⁶ in their study investigating a combined behavioral therapy intervention over a 12-month period in musicians affected by FHD. They obtained excellent intra-rater reliability, using an intra-class correlation model (ICC Model (2,1)), with ICC values ranging between 0.985 and 0.999 ($p<0.001$) and a set of narrow 95% confidence intervals (CI) (0.851–1.000). Berque et al.³⁶ also found good inter-occasion (or test-retest) reliability for the FAM scores (ICC Model (2,1)), and concluded that the FAM could, therefore, be considered as a useful objective clinical outcome measure for musicians with FHD.

Management: Behavioral Therapy Interventions

The management of FHD remains imperfect and uncertain, and to date there is no "cure."^{3,7,13,41} Oral anticholinergic medication and botulinum toxin injections are palliative; repeated injections are necessary and may not be effective for all patients, depending on dystonic patterns and precise localization of the dystonic muscles.^{3,42–44} Given these limitations, the search for new behavioral therapy interventions for the management of FHD has emerged.^{4,17,45} With the recent advances in the neurophysiology of FTSD and brain imaging previously described, new behavioral strategies for musician's dystonia and writer's cramp, based on principles of neuroplasticity, have aimed to reverse the disturbed somatotopic cortical map for the affected hand.¹¹

Limb immobilization in a static splint for 4 weeks led to decrease of the cortical representation of the hand and improvement of arm dystonia, but only for some individuals.^{32,45}

Byl and colleagues^{46–48} used a complex program of learning-based sensory training (LBST) in individuals with FHD, aimed at redefining spatial and temporal processing capabilities. The subjects improved significantly in obtaining fine motor control, accuracy, sensory discrimination, and functional independence. However, since many different features of retraining were included in the protocol, it was difficult to evaluate which element was most successful.

Sensory retraining, in the form of Braille reading, revealed improvements in spatial discrimination and decrease in symptoms of FHD in individuals with writer's cramp.^{49,50}

In a recent study using proprioceptive retraining, aiming at reversing the excessive sensorimotor reorganization seen in

musician's dystonia^{22,24} towards the pattern seen in healthy subjects, Rosenkranz et al.²⁵ showed that a 15-minute intervention with proprioceptive input (muscle vibration) restored the sensorimotor organization in pianists with FHD to the pattern seen in healthy pianists. Furthermore, they showed a statistically significant improvement in motor control, as measured with the MIDI-based scale analysis developed by Jabusch et al.,³³ and a significant improvement in the BFM and TCS ordinal scores. Unfortunately, improvement was short-lived, usually lasting no more than 24 hours,²⁵ and further research is required to assess the long-term benefits of this retraining strategy.

Sakai³⁵ developed a motor control retraining technique, named "slow-down exercise" (SDE), on 20 pianists affected by FHD. An exercise program was developed in which participants underwent basic movement training at decreased speed, making sure that the dystonic patterns would not occur at this reduced speed. The pianists would increase metronome speed every 2 weeks as long as they could maintain a normal movement pattern. Results showed a significant improvement from 2.2 ± 0.41 to 4.6 ± 0.50 ($p<0.05$) with the TCS scores over time,³⁵ although there was no mention of the frequency of follow-up measurements and of the type of statistical tests used. The duration of the treatment period was not standardized between subjects, ranging from 1 to 6 years. Furthermore, no descriptive or inferential statistical analysis was carried out on the metronome speed increase achieved over time.

Most of these studies have shown encouraging results, but most of them were not randomized controlled trials;⁵¹ samples were small, interventions were carried out over short periods (< 6 months), most of them did not use a control group, and subjective ordinal scales were often used to measure progress.

Task-Specific Constraint-Induced Therapy for Musician's Dystonia

More recently, attention has focused on another approach, involving motor retraining using constraint-induced therapy. Based on the assumption that behavioral mechanisms underlie both the cortical disorder and the involuntary loss of coordination,^{2,4} Candia et al.⁴⁰ developed a novel treatment intervention, sensory motor retuning (SMR), using intensive constraint-induced therapy in an attempt to regain normal finger movement patterns and reverse the cortical fusion observed in individuals affected by FHD.² Splints were used to immobilize specific fingers, depending on the pattern of the dystonia. The splints could be tailored to the individual's hand while holding a normal playing posture on their instrument. The main dystonic finger that was being trained was not splinted, while one or several adjacent fingers were splinted to reduce co-contraction. The main dystonic finger was required to carry out task-specific repetitive exercises for one hour per day on the instrument in coordination with the remaining free digits.^{40,52} Determining which fingers would need to be splinted was achieved by immobilizing, in turn, each finger participating in the abnormal movement pattern



FIGURE 1. Dystonic patterns for Subjects 1 and 3.

while the musician was playing with the remaining digits. In all cases, immobilizing one or two digits enabled a freer and more independent movement pattern of the main dystonic finger with reduced co-contractions.⁴⁰

Unfortunately, the sample was small ($n=11$), and there was no control group, thereby questioning internal validity of the study.⁵³ In a follow-up study, however, Candia et al.⁵⁴ explained that having a control group may have been unethical and would have caused attrition due to lack of motivation of the participants in the control group. Data collection was performed using an ordinal dystonia evaluation scale (0 to 4 DES), developed in a previous study,⁵² and a dexterity and displacement device measuring smoothness of movement. Pianists and guitarists showed a statistically significant improvement in DES scores between pre-treatment, post-treatment, and follow-up scores ($F_{2,20}=16.5, p<0.0001$), and in smoothness of movement ($t=2.9, p<0.05$).⁴⁰ The authors concluded that constraint-induced therapy was of value in the treatment of focal hand dystonia in pianists and guitarists, but not in wind players, and suggested that normalization of movement patterns occurred through normalization of the cortical representational maps.⁵⁴ However, there was no indication that the outcome measures used had been validated and there was no attempt to ascertain intra- and inter-rater reliability. There was also no evidence of blinding of those measuring outcomes. Moreover, Candia et al.⁴⁰ analyzed their ordinal DES scores using a one-way and then two-way ANOVA models without specifically saying these models were repeated measures models or that assumptions of these parametric procedures were appropriate given the ordinal data.⁵⁵ Given this lack of detail and the small sample, their statistical significant results should be treated with some caution. Finally, the treatment period was not standardized and varied greatly between individuals, with a final follow-up ranking from 2 to 25 months.

The woodwind players (two flautists and one oboist) did not improve, perhaps due to the fact that wind players were asked to blow into their instruments only occasionally and that finger-mouth coordination was, therefore, not addressed in the study protocol. Indeed, a recent study by Hirata et al.⁵⁶ showed altered hand and mouth relationship (reduced dis-

tance between lip and hand representations) in the somatosensory cortex of musicians affected by embouchure dystonia. Furthermore, the three wind players in the study by Candia et al.⁴⁰ had only been treated for 1 week, 2 months, and 4 months, respectively, at their final follow-up. Owing to the neurological nature of FHD and when considering comments made by Zeuner et al.,³⁷ who concluded that a 4-week period of retraining was not long enough to produce excitability changes or reorganization of the motor cortex in 10 individuals with writer's cramp, the retraining periods used by Candia et al.⁴⁰ for their wind players may be too short to make assumptions of treatment failure.

Combined Behavioral Therapy Intervention: Constraint-Induced Therapy and Motor Control Retraining

In an attempt to take account of these shortcomings, the present authors³⁶ carried out a study whereby the aim was to investigate the effects of a combined behavioral therapy intervention over a 12-month period in eight musicians affected by FHD. Constraint-induced therapy (SMR) and motor control retraining (with SDE) were the behavioral interventions. A standardized protocol was used, and compliance was monitored closely. A quasi-experimental repeated measures (within-subject) design was used. All participants were tested at regular intervals during the study period (day 1, day 8, then every 2 months up to month 12) whilst playing an easy piece and a medium-difficulty piece.

Intensive, constraint-induced therapy involved playing specific finger combinations, aimed at normalizing movement patterns for the main dystonic finger (Fig. 1) (Table 2), with a splint in situ immobilizing the selected adjacent fingers to reduce co-contractions (Fig. 2).^{36-38,40} A metronome was used at all times, and speed was modified during the performance of each sequence in order to challenge the subjects and make the motor control retraining more specific.^{36,40} Following an intensive 1-week period of constraint-induced therapy (2hrs/day), participants were asked to practise these finger sequences daily with their splints for 30 to 60 minutes.³⁶ In addition, they were asked to practise motor control retraining at slow speed (SDE) while maintaining good movement pat-

TABLE 2. Subjects Characteristics

Subjects	Instrument	Gender	Age*	Affected Fingers	Dystonic Pattern	Dystonia Onset	Compliance [†]
1	Guitar	Male	53	Right D3, D4, D5	D4 Flexion D3 Extension D5 Extension	2006	95%
2	Guitar	Male	47	Right D3, D4, D5	Flexion	1982	76%
3 [#]	Flute	Male	47	Left D4, D5	Flexion	D5: 2002 D4: 2006	95%
4	Flute	Female	48	Right D4, D5	Flexion	2004	95%
5	Bagpipes	Male	55	Right D5	Flexion and Adduction	2005	77%
6	Bagpipes	Male	51	Right D3, D4	Flexion	1995	40%
7 [#]	Oboe	Male	48	Right D4, D5	D4 Abduction D5 Flexion	2006	88%
8 [#]	Accordeon	Male	30	Right wrist, D2, D3, D4	Extension	2005	N/A

*Age at the start of the initial study—Day 1 of the protocol, i.e., during 2007 for all subjects except subject 4, who started in 2008.

[†]Compliance: % of days when the protocol was practised during the initial 12 month-period.

[#]These subjects did not complete the initial 1-year study: Subject 3, despite making good progress, was excluded after Month 10 after receiving a botulinum toxin injection, but was included for the follow-up study at year 4, since he had not received any injections for over 2 years. Subject 7 withdrew after month 6 and Subject 8 after month 2, due to lack of progress.

terns, without splints, for 30 minutes/day.^{35,36} Contrary to Candia et al.,⁴⁰ the wind players were asked to play continuously during a specific sequence, alternated by a sequence without blowing in the instrument, in order to take account of finger-mouth coordination as a possible source of dystonic patterns.⁵⁶ For the same reasons, SDE was practised while blowing in the instrument. The outcome measures used were the FAM scale;²⁷ the change in metronome speed achieved during SDE;³⁵ and two ordinal dystonia evaluation scales, the TCS³⁰ and the ADDS²⁹ (Table 1).

Although some data were missing for three individuals who did not complete the study period, the results from the two-factor repeated measures ANOVA revealed that a retraining protocol combining constraint-induced therapy (SMR) and motor control retraining (SDE) led to statistically significant improvements in FAM scores with a decrease, by approximately 80%, in the number of abnormal movements per second of instrumental playing over time, for both pieces of music combined ($F=6.32$, $df=7$, $p<0.001$),³⁶ indicating a trend towards normalization of movement patterns, in keeping with

the findings of Candia et al.⁴⁰ Similar statistically significant trends were obtained for the other outcome measures.

Furthermore, the results of the Tukey's post-hoc test for the FAM scores suggested that that retraining had to be carried out for at least eight months before statistically significant changes in FAM scores were noted. These findings were made possible by the study design (repeated measures design), since all participants were followed over a 12-month period, and were tested at similar time intervals. This notion of a minimum period of retraining had not been reported before in musicians with FHD. Indeed, in the studies by Candia et al.⁴⁰ and Sakai,³⁵ the follow-up period for the final measurement was not standardized and varied greatly between subjects.

Moreover, although this was not the main aim of their study, Spector and Brandfonbrener²⁷ analyzed the treatment effects of their SMR protocol at day 1, day 8, and month 6. They noted significant improvement at day 8, but no further improvement between 1 week and 6 months of therapy. Their results are in contrast with the findings from Berque et al.³⁶ However, the lack of improvement observed by Spector



FIGURE 2. Constraint-induced therapy for Subjects 1 and 3.

and Brandfonbrener²⁷ may be due to a lack of subject compliance, and a short retraining period (<6 months). Similarly, a short retraining period, combined with the fact that their wind players did not blow constantly in their instrument, may explain the lack of improvement for the wind players in the study by Candia et al.,⁴⁰ in contrast with Berque et al.³⁶

Although confirming similar trends, when comparing the results of the FAM scores and the DES scores in the study by Berque et al.,³⁶ Tukey's post-hoc test revealed that the FAM scale was more sensitive to change, as suggested by Spector and Brandfonbrener,²⁷ showing significant changes taking place from 8 months of therapy for the FAM data, compared to month 10 for the TCS and month 12 for the ADDS. Furthermore, this confirmed what was hypothesized by Spector and Brandfonbrener²⁷ in that ordinal scales give a global clinical impression of the severity of musician's dystonia, whereas the FAM scale provides an objective quantification of the frequency of abnormal movements, being therefore more specific. These observations on dystonia evaluation scales are also in keeping with Zeuner et al.³⁷ who commented on the difficulty of relying on subjective self-assessment scales and on the ADDS to rate FHD.

The study by Berque et al.³⁶ has, however, some limitations: no control group was used; the sample was small ($n=8$), and only six completed the 1-year study; no inter-rater reliability tests were carried out for the FAM scores; no intra or inter-rater reliability tests were carried out on the DES scores and metronome speed data; since the protocol combined constraint-induced therapy and motor control retraining, the improvements shown cannot be attributed to one intervention alone, but to a combination of both.

Long-Term Treatment Effects of Constraint-Induced Therapy for Musician's Dystonia

Owing to the neurological nature of FHD, long-term studies are required to determine whether constraint-induced therapy can yield long-term benefits for musicians with FHD. Apart from the study by Berque et al.,³⁶ which followed eight musicians affected by FHD at regular intervals over a 12-month period using a standardized protocol and monitoring compliance closely, there is only one case report of a pianist relating to long-term treatment follow-up⁵⁷ using constraint-induced therapy (SMR). The authors reported the case of a pianist, affected by severe FHD who was treated successfully using constraint-induced therapy over a 12-month period. Using a 10-point ordinal dystonia evaluation scale, the functional level progressed from 29% to 100% over the period. When assessed at follow-up 8 years later, the pianist had maintained the progress achieved, and had returned to professional musical performance. No reliability tests were carried out on the chosen 10-point ordinal scale.

STUDY AIMS

The aim of the current study was to investigate the long-term effects of a combined behavioral therapy intervention aimed

TABLE 3. Mean Years to Follow-up

Subjects	Instrument	Years to Follow-up
1	Guitar	3.42
2	Guitar	2.83
3	Flute	3.00
4	Flute	2.54
Mean (SD)		2.95 (0.37)

SD, standard deviation.

at normalizing finger movement patterns, in the musicians affected by FHD who completed the original 12-month study.³⁶ The present study was a 4-year follow-up, and the objective was to determine whether or not participants have continued to progress or at least have maintained the level achieved since the end of the initial study. It was hypothesized that there would be significant differences in FAM scores, DES scores, and metronome speeds at the 4-year follow-up. A subsidiary aim was to assess intra- and inter-reliability of the two dystonia evaluation scales used, the ADDS and the TCS, since this had not been possible during the initial study.

METHODS

The study was approved by the Research Ethics Committee of the School of Health and Life Sciences at Glasgow Caledonian University. All participants provided written informed consent.

Subjects

Four instrumental musicians affected by focal hand dystonia (Fig. 1) volunteered to participate in the 4-year follow-up study: two guitar players and two flute players (Subjects 1 to 4 in Table 2). Eight musicians had participated in the initial 1-year study;³⁶ six were professional musicians, two were amateur musicians. There was only one female in the group.

Table 2 summarizes the characteristics of the individuals who took part in the two studies. Some data were missing for three participants who did not complete the 1-year period. Despite making good progress, Subject 3 received a botulinum toxin injection after month 10 and had to be excluded after that point. Subjects 7 and 8 decided to withdraw after month 6 and month 2, respectively, due to lack of progress. For this long-term follow-up study, the mean years to follow-up was 3 years since completion of the initial 1-year study (Table 3).

In an attempt to limit threats to internal validity, especially the concepts of "history" and "maturation,"⁵³ inclusion criteria were checked again. Each of the four participants met the inclusion criteria for the study:^{40,58} they had not been diagnosed with any other neurological condition, nor had they received a botulinum toxin injection within the past year; they did not suffer from any other movement disorders, nor did they have any other neurological signs or suffer from nerve entrapment syn-

TABLE 4. Practice Profile*

Question	Subject			
	1	2	3	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:				
< 15 min?				
Between 15 min and 0.5 hr?	✓		✓	✓
Between 0.5 and 1 hr?		✓		

* Adapted from Ackermann and Driscoll.⁶⁰

dromes; they were not undertaking any other medical treatment or therapy for their dystonia. Since Subject 3 had not received any botulinum toxin injections for more than two years, he was included in this 4-year follow-up study (Table 2).

Additionally, each participant met all the following clinical inclusion criteria:⁵⁹ motor skill impairments were specific to playing their instrument and could be described by errors in timing, force, or trajectory with cramping sensations or involuntary movements of the fingers; function was impaired as a result of degraded movement; skill loss could not be explained by a decrease in practice of their instrument.

Neurological Screening

In the initial 1-year study, extensive neurological tests were carried out on both hands to ascertain normal sensory function.³⁶ Since the participants met all the inclusion criteria described above, and no change in their health status had occurred within the 3-year period since the end of the initial study, these neurological tests were not repeated.

Retraining Protocol: Constraint-Induced Therapy and Motor Control Retraining

The aim of the initial 1-year study was to investigate the effects of an intensive retraining protocol combining constraint-induced therapy (SMR)⁴⁰ and motor control retraining (SDE)³⁵ over a 12-month period in musicians affected by FHD.³⁶ A very detailed account of the retraining protocol can be found in a recent publication of the present authors.³⁶

Practice Profile

Following the initial 1-year study, participants were no longer monitored. At the time of this 4-year follow-up, participants were therefore asked to fill in a small questionnaire, adapted from Ackermann and Driscoll,⁶⁰ in order to determine the level of compliance with their specific retraining program since the end of the initial study (Table 4).

Data Collection

For this 4-year follow-up, one testing session was carried out for each of the four individuals approximately 3 years (mean, 2.95 ± 0.37 yrs) after completion of the initial 1-year study

(Table 3). The same standardized protocol was used for this follow-up study and is summarized below.³⁶

The outcome measures used were those chosen for the initial study by Berque et al.,³⁶ i.e. the FAM scale, the ADDS, the TCS, and the metronome speed score achieved during SDE. The two test pieces chosen for the initial study were used for data collection at follow-up, and the same tempi were respected.³⁶ A metronome (Zen-on metriona, quartz metronome, Seiko Instruments & Electronics Ltd, Maidenhead, Berkshire, UK), previously tested for accuracy and precision, was used at all times.

At the 4-year follow-up session, participants were rated by the researcher once using two ordinal dystonia scales, the TCS³⁰ and the ADDS²⁹ (Table 1). The FAM, developed by Spector and Brandfonbrener²⁷ and slightly modified by Berque et al.,³⁶ was used as an objective measure. The FAM score is the number of abnormal movements per second of instrumental playing.

A digital video camera (Lumix DMC-FX3, Panasonic, Osaka, Japan) was used to videotape participants playing the two musical pieces. Individuals were asked to play continuously for at least 3 minutes. Each piece was recorded twice, as per initial protocol which assessed subject "inter-occasion" (test-retest) reliability.³⁶ A 3-minute window of playing was used for data analysis.

SDE scores were determined as previously described.³⁶

Data Analysis

Video segments were transferred to a computer for analysis. The main author carried out the analysis. The number of abnormal movements was counted over the 3-minute window. Abnormal movements from the main dystonic finger and from the other affected fingers were counted separately for each video. They were then added up and the total was divided by the duration of the excerpt (180 seconds) to obtain a value per second of playing (ratio data).^{27,36,55}

Each video clip was scored twice, with a 1-week interval, to avoid possible memory bias. Since the results from the ICC Model (2,1) in the initial study showed excellent intra-rater reliability,³⁶ the mean of the two scores for each clip was used in the present study. Similarly, since the ICC Model (2,1) revealed generally good inter-occasion reliability (test-retest) between the two recordings of each piece, the mean of the

two video clips (occasions) was therefore used for the statistical analysis of variance ANOVA.

Dystonia Evaluation Scales: Reliability Tests

In an attempt to determine the usefulness of the dystonia evaluation scales,³⁹ intra-rater and inter-rater reliability were tested for the TCS and ADDS. These tests were carried out by the main author (PB) and by a second rater (RH) of a high musical standard.

Two time periods were chosen and tested independently: time period 1 (day 1) and time period 3 (month 2) of the initial study. These periods were chosen since video data were available for all eight participants. After month 2, Subject 8 had indeed withdrawn from the initial study.³⁶

Since each piece had been recorded twice, only the first recording of each piece (easy and medium-difficulty) was selected for the tests. There were therefore two videos per individual except for Subject 2, who had not been able to play the medium-difficulty piece due to the severity of his dystonia. Each rater was asked to watch both pieces before giving one score only for each DES. It was assumed that watching both the easy and the medium-difficulty pieces would give a broader clinical impression of the severity of the participants' dystonia. Both researchers independently rated the videos. The procedure was repeated 2 weeks later in order to minimize memory bias.

In order to minimize expectancy effects from the researcher (Rosenthal effect),⁵³ the second rater (RH) was blinded to the experiment and did not have any knowledge of the time periods being tested or previous knowledge of the participants. It was unfortunately not possible to blind the main researcher, who had extensive knowledge of each participant's dystonia and had followed them over the 4-year period.

The second rater (RH) had received a 1-hour training session to be familiarized with scoring videos, using the TCS and ADDS. This training session included an explanation of the dystonic patterns for each subject (Table 2), using videos from other time periods, which were not included in the actual reliability tests.

Statistical Analysis

SPSS software version 19 was used for descriptive and inferential analysis (SPSS Inc., Chicago, IL). A quasi-experimental repeated measures (within-subject) design was used, incorporating nine testing sessions from the initial study and this follow-up: day 1, day 8, month 2, month 4, month 6, month 8, month 10, month 12, and year 4. The researcher was manipulating two independent variables: the factors "time" (nine testing sessions) and "piece" (easy and medium-difficulty).

For the FAM scores, the potential sources of variation identified were: intra-rater reliability; inter-occasion reliability (within-subject and within-day variation); between-subject and within-day variations; between-conditions comparison (easy vs medium difficulty piece); and between-sessions comparison (nine testing sessions over 4 years). Intra-rater and

inter-occasion reliability had already been tested previously, using an intra-class correlation model: ICC Model (2,1).³⁶ A two-factor parametric repeated measures analysis of variance (ANOVA) was carried out, with the number of abnormal movements per second (AM/sec) as the dependent variable. Diagnostics were performed after the model was fitted to confirm normality and homoscedasticity.⁵⁵

For the DES scores, one-factor repeated measures parametric ANOVA models were used, with the respective scores obtained for each DES as the dependent variable, and normality and homoscedasticity were confirmed.⁵⁵ Intra-rater and inter-rater reliability were tested using an intra-class correlation model: ICC Model (2,1).⁶¹

For the metronome speed scores, a two-factor parametric repeated measures ANOVA was carried out, with the metronome speed achieved without occurrence of abnormal movements as the dependent variable, and once again appropriate diagnostics were performed.⁵⁵

Where applicable, Tukey's post-hoc tests were carried out to determine which time periods were significantly different. All tests were performed using a 5% level of significance ($\alpha=0.05$).

RESULTS

Intra- and Inter-Rater Reliability Tests for the Ordinal Dystonia Evaluation Scales

Results from the ICC Model (2,1) showed good to very good intra-rater reliability for both raters, with ICC values ranging between 0.700 and 1.000 and a set of fairly narrow 95% confidence intervals (CI) (0.600–0.999), except for the values relating to day 1 for the TCS, where the CI was wider (Table 5). Since the intra-rater scores were good, the mean of the two scores for each rater was used when assessing inter-rater reliability.

For the inter-rater reliability test, the ICC Model (2,1) revealed good to very good results, with ICC values ranging between 0.760 and 0.900 and a set of reasonable 95% CI (Table 5). Once again, the values relating to the TCS revealed a wider CI, but for month 2.

FAM Scores: Comparison Over Time

For the 4-year follow-up (day 1 to year 4), the results from the two-factor repeated measures ANOVA showed a statistically significant trend: the number of AM per second of instrumental playing decreased by approximately 80% over time for both pieces combined ($F=7.85$, $df=8$, $p<0.001$) (Fig. 3), indicating a significant improvement with normalization of movement patterns over the 4-year period. Similar results had been obtained during the initial 1-year study.³⁶ Since these results were significant, Tukey's post-hoc test was carried out to determine which time periods were statistically significant over the 4-year period. Results showed significant improvements only between day 1 and any period from month 6; between day 8 and any period from month 8; between month

TABLE 5. Ordinal Dystonia Evaluation Scales: Intra- and Inter-Rater 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, Tubiana and Chamagne dystonia scale; ADDS, Arm Dystonia Disability Scale.

2 and year 4; between month 4 and year 4; and between month 6 and year 4 (with p -values ranging between $p < 0.001$ and $p = 0.044$). This would suggest a late effect of therapy and remains consistent with results obtained previously.³⁶

In contrast with the initial 1-year study, which showed significant differences only between day 1 and any period from month 8, and day 8 and any period from month 8, these long-term follow-up results showed that further progress could be obtained over a longer time period since month 2, month 4, and month 6 were significantly different from year 4.

Furthermore, Tukey's post-hoc test did not reveal any statistically significant difference between month 12 and the follow-up at year 4. However, Figure 3 shows a trend whereby the progress achieved during the first year of intensive

retraining was improved further for the easy piece and maintained for the medium-difficulty piece.

FAM Scores: Comparison Between Easy and Medium-Difficulty Pieces

For the 4-year follow-up (day 1 to year 4), the results showed a statistically significant difference between the two pieces ($F = 6.30$, $df = 1$, $p = 0.014$) (Fig. 3). The number of AM per second was significantly greater for the medium-difficulty piece than for the easy piece, although the interaction between the factors "time" and "piece" shown in Figure 3 was not significant ($F = 0.42$, $df = 8$, $p = 0.908$). However, Figure 3 shows a trend whereby the differences between the two pieces occurred in the first 6 months of the retraining period. During this period, individuals generally found the medium-difficulty piece harder to play, and this is reflected by the larger number of AM per second. After 6 months, this difference virtually disappeared until month 12 (Fig. 3). Between month 12 and the follow-up at year 4, the trend showed continued improvement for the easy piece, and no further improvement for the medium-difficulty piece.

It must be noted that, although Subject 2 (one of the three participants who was originally unable to play the more difficult piece) had made enough progress between month 12 and year 4 to be able to play the medium difficulty piece without stopping, he was not included in the ANOVA model with regards to the medium-difficulty piece due to non-existence of previous data.

Dystonia Evaluation Scale Scores

For the 4-year follow-up (day 1 to year 4), the results of the one-factor repeated measures ANOVA carried out for each DES revealed a significant improvement in scores for both the TCS ($F = 7.18$, $df = 8$, $p < 0.001$) and the ADDS ($F = 5.26$, $df = 8$, $p < 0.001$) over the treatment period (Fig. 4).

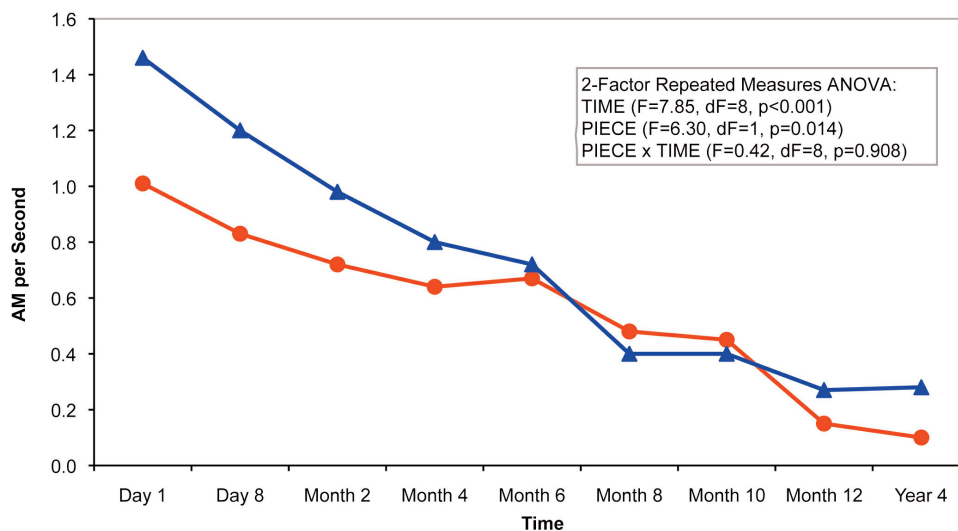


FIGURE 3. Frequency of abnormal movements (FAM) scale: mean values for each piece and all subjects. Circles indicate easy piece, and triangles indicate medium-difficulty piece.

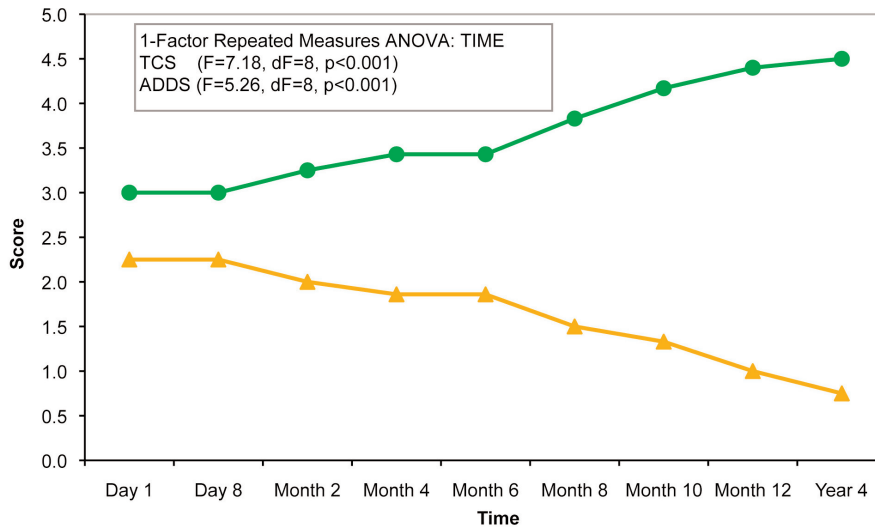


FIGURE 4. Ordinal dystonia evaluation scale scores: mean values (circles, TCS; triangles, ADDS).

Tukey's post-hoc tests for the TCS showed statistically significant improvements only between day 1 and any period from month 10; between day 8 and any period from month 10; between month 2 and any period from month 12; between month 4 and year 4; between month 6 and year 4; and between month 8 and year 4 (with p -values ranging between $p < 0.001$ and $p = 0.043$). The results showed no statistically significant difference between month 12 and the follow-up at year 4.

Tukey's post-hoc tests for the ADDS showed statistically significant improvements only between day 1 and any period from month 12; between day 8 and any period from month 12; between month 2 and year 4; between month 4 and year 4; and between month 6 and year 4 (p -values ranging between $p < 0.001$ and $p = 0.045$). The results showed no statistically significant difference between month 12 and the follow-up at year 4.

These post-hoc tests would suggest a late effect of therapy, in keeping with results obtained previously.³⁶

Metronome Speed Scores

For the 4-year follow-up (day 1 to year 4), the results from the two-factor repeated measures ANOVA showed a very significant trend: the metronome speed achieved by subjects without occurrence of AM increased significantly over time (from approximately 30 to 100 bpm) for both pieces combined ($F = 20.45$, $df = 8$, $p < 0.001$) (Fig. 5). Tukey's post-hoc test showed significant improvements only between day 1 and any period from month 4; between day 8 and any period from month 8; between month 2 and any period from month 8; between month 4 and any period from month 10; between month 6 and any period from month 12; between month 8 and year 4; between month 10 and year 4 (with p -values ranging between $p < 0.001$ and $p = 0.045$).

For the follow-up period, Tukey's post-hoc test did not reveal any statistically significant difference between month

12 and year 4. This could be explained by the decrease in mean speed noted for the medium-difficulty piece (Fig. 5).

The results showed no statistically significant difference between the two pieces ($F = 0.01$, $df = 1$, $p = 0.925$) and no significant interaction between the factors "time" and "piece," as shown in Figure 5 ($F = 0.80$, $df = 8$, $p = 0.609$). Indeed, the steady improvement over time was very similar for both pieces except at year 4 for the medium-difficulty piece, as stated above (Fig. 5). These factors would explain the lack of a significant interaction effect.

DISCUSSION

Long-Term Effects of the Treatment Protocol on Focal Hand Dystonia: FAM Scores

The present study revealed that a retraining protocol combining constraint-induced therapy (SMR) and motor control retraining (SDE) led to statistically significant improvements in FAM scores over time for both musical pieces (Fig. 3), in keeping with results from the initial study.³⁶ These improvements in FAM scores were also clinically significant, showing a large 80% reduction in the number of AM per second of playing. These results demonstrate a trend towards normalisation of movement patterns when tailored retraining takes place in a task-specific environment,^{36,40} i.e., playing the instrument. This is reinforced by the trend observed for the interaction between the factors "time" and "piece" shown in Figure 3, whereby the difference in number of AM per second of playing between the two pieces virtually disappeared after 6 months of therapy, except between month 12 and year 4. These findings are in keeping with the theory that in the presence of maladaptive and use-dependent cortical reorganisation,^{2,4,17,26,62,63} task-specific retraining aims to break apart cortical fusion and allow normal cortical segregation to be re-established, thereby restoring normal sensory and motor representations of the hand and fine motor control.^{2,4,40,62}

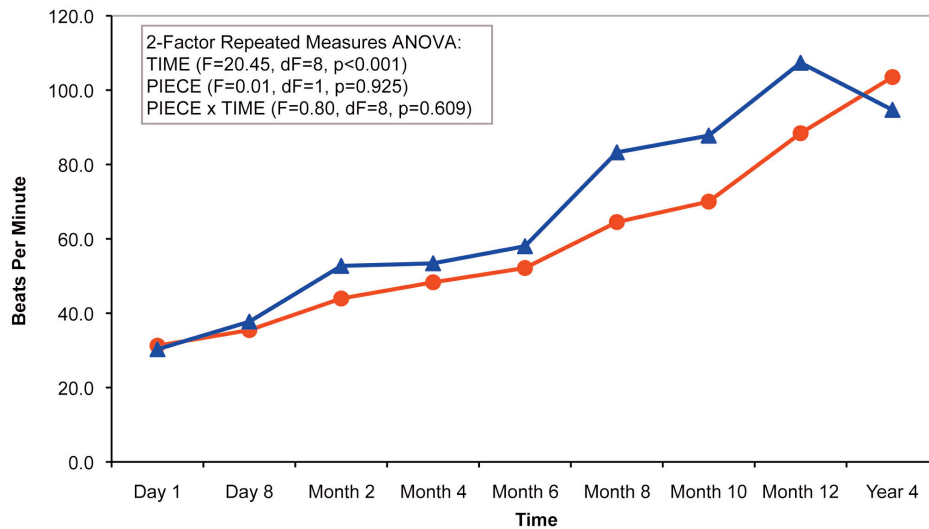


FIGURE 5. Metronome speed scores: mean values for each piece and all subjects. Circles indicate easy piece, and triangles indicate medium-difficulty piece.

Of important note, the results of the Tukey's post-hoc test suggested that retraining had to be carried out for at least 6 months before significant changes in FAM scores were noted. These findings were made possible by the study design (repeated measures design), since all individuals were followed over a 4-year period and were tested at similar time intervals. These findings are in keeping with the initial study by Berque et al.,³⁶ although the results from the initial study had shown that a minimum of 8 months of therapy was necessary to obtain statistically significant changes in FAM scores. This difference may be due to the small sample used in the follow-up study ($n=4$). This notion of a minimum period of retraining has been reported by the present authors in their initial study on musicians with FHD,³⁶ and they concluded that the lack of improvement in FAM scores observed by Spector and Brandfonbrener²⁷ and by the wind players in the study by Candia et al.⁴⁰ may have been due to a short retraining period. These comments would still seem valid when considering the results of the present study. Furthermore, this concept of a "minimum" duration of retraining has already been mentioned by Zeuner et al.³⁷ in their study on 10 individuals with writer's cramp. In the absence of changes in their neurophysiological measures, they concluded that 4 weeks of retraining was not long enough to produce excitability changes or reorganisation of the motor cortex.

Moreover, Tukey's post-hoc test revealed that further progress could be obtained over a longer time period than 12 months. Indeed, the results suggest statistically significant differences taking place between month 2 and year 4, month 4 and year 4, and month 6 and year 4. This has not been reported before and suggests that an intensive 12-month retraining protocol combining constraint-induced therapy and motor control retraining may lead to long-term benefits for musicians with FHD, although it must be borne in mind that there were only four participants in the follow-up study, and that they represented only two instrument groups (flute and guitar).

However, Tukey's post-hoc test did not reveal any statistically significant difference between the end of the initial study (month 12) and the follow-up period (year 4), thereby rejecting the hypothesis. The clinical trend between month 12 and year 4 showed continued improvement for the easy piece, and no further improvement for the medium-difficulty piece (Fig. 3). These results could be explained by the increase in mean FAM score for the medium-difficulty piece (Fig. 3), which was perhaps skewed due to a slight deterioration in Subject 1's dystonia compared to month 12. Indeed, Subject 1's score for the medium-difficulty piece would have an impact on the mean FAM in the small sample available for this follow-up ($n=4$). However, when considering that most individuals only spent an average of 15 to 30 minutes per day on their retraining program since the end of the initial study (Table 4), these results seem encouraging and suggest that progress achieved during the initial intensive 12-month retraining period was maintained at follow-up (Fig. 3) despite a small amount of daily specific retraining. This has not been reported before.

Comparison of the FAM and Dystonia Evaluation Scale Scores

The present study indicated that the retraining protocol led to statistically significant improvements in subjective ordinal dystonia scale scores for both the TCS and the ADDS over time (Fig. 4), thereby confirming the results obtained from the FAM scale and the trend towards normalisation of movement patterns already observed during the initial study.³⁶ These findings are in keeping with the improvement in DES scores obtained by Candia et al.⁴⁰ and Sakai³⁵ in their studies on musicians. However, as already noted, their protocols were not standardised with regards to the follow-up period, and reliability testing was not carried out for the subjective ordinal scales they used, contrary to the present study.

Interestingly, the results from the Tukey's post-hoc test showed that statistically significant changes took place near the end of the initial study period (month 10 for the TCS, and month 12 for the ADDS), indicating a late effect of therapy, in keeping with the results from the initial study.³⁶ For the ADDS however, in contrast with the initial 1-year study, which showed significant differences only between day 1 and month 12 and between day 8 and month 12, these long-term follow-up results would also suggest that further progress could be obtained over a longer time period since month 2, month 4, and month 6 were significantly different from year 4. This pattern is similar to what was noted for the TCS, although not quite to the same extent, confirming that the TCS may be more sensitive to change than the ADDS, as already suggested by Berque et al.³⁶ These trends are similar to what was described for the FAM scores over time, including the fact that there was no statistically significant difference between month 12 and the follow-up at year 4.

Finally, when comparing the FAM scores with the DES scores, Tukey's post-hoc test revealed that the FAM scale may be more sensitive to change, showing significant changes taking place from 6 months of therapy (Fig. 3), thereby confirming what was hypothesised by Spector and Brandfonbrener.²⁷

Reliability Tests for the Ordinal Dystonia Evaluation Scales

In view of the good to very good results obtained from intra- and inter-reliability testing for both ordinal scales, the dystonia evaluation scale scores obtained in both the initial study³⁶ and the present study can be treated with more confidence since the video data used for testing originated from the initial study.

The results obtained from the ICC Model (2,1) for the ADDS showed very high scores for all time periods studied (day 1 and month 2), in keeping with the study by Spector and Brandfonbrener,²⁷ and further validate the use of the ADDS as a reliable subjective ordinal scale for musician's dystonia.

With regards to the TCS, the results from the ICC Model (2,1) represent the first attempt to validate this scale for reliability. The results are encouraging, since the intra-rater scores were consistently good to very good for both raters, and the inter-rater reliability scores were also high (Table 5). However, it must be noted that the scores for the time period day 1 were lower than for month 2, and this was the case for both raters. When analysing the scores given by each rater in detail, the results showed that half of the discrepancies occurred mainly between "stage 2" and "stage 3" of the scale (Table 1). Possible reasons for the discrepancies are: the TCS has a larger range of values (0 to 5) than the ADDS (0 to 3), and this makes scoring more difficult for raters; despite blinding of the second rater, there may have been a training effect since month 2 was scored directly after day 1; the wording of stages 2 and 3 may have caused some ambiguity (Table 1).

Although the TCS was specifically written to assess musician's FHD, it does not necessarily encompass, in each stage, all the common symptoms/characteristics observed in musicians with FHD, i.e., the ability to play without stopping, speed/tempo

specificity, musical piece specificity, irregularities in rhythm due to lack of coordination, frequency of abnormal dystonic movements, presence of compensatory movements.^{3,4,6,27}

Metronome Speed Scores

The present study revealed a statistically significant improvement in metronome speed achieved over time without occurrence of abnormal movements for both pieces (Fig. 5), in keeping with the results obtained by the present authors in their initial study.³⁶ These results were also clinically significant, showing a large increase in speed achieved from approximately 30 to 100 bpm for both pieces. These results are also in agreement with the trend observed in the study by Sakai,³⁵ who noted that the speed of performance increased until reaching 88.6% of normal performance speed in a group of 20 professional pianists. However, Sakai did not carry out any statistical analysis on their metronome speed data.

The Tukey's post-hoc test for the metronome speed scores showed that significant changes occurred more gradually than for the FAM scores over the 4-year period (Fig. 5), in keeping with the results obtained in the initial study.³⁶ The reason given was that the metronome speed scores did not have a finite end point, contrary to the FAM scores, which were converging towards "0." Indeed, individuals were encouraged to keep increasing the metronome speed above the "normal" standardised tempi of the test pieces as long as they could play without occurrence of abnormal movements. These findings would suggest that "change in metronome speed" could prove a sensitive tool to measure progress in musician's FHD. This tool still requires to be evaluated with regards to reliability and validity, as this was not carried out in the present study. Thus, the results of the metronome speed data need to be considered with some caution.

Finally, Tukey's post-hoc test did not reveal any statistically significant difference between month 12 and year 4, in keeping with the results from the other outcome measures used. This could be explained by the decrease in mean speed noted for the medium-difficulty piece (Fig. 5), which was perhaps skewed due to a slight deterioration in Subject 1's dystonia compared to month 12, and a very small sample ($n=4$).

Participants' Functional Improvement

Both flute players were professionals who belonged to one of the major symphonic orchestras in Scotland. Subject 3 was forced to take more than 8 months off work due to the severity of his dystonia at the start of the initial study. He made significant progress during the first 10 months of retraining, returned to orchestra playing, and was coping well. He then decided to try botulinum toxin injections and was therefore excluded from the initial study at this stage (Table 2). He had five botulinum toxin injections over a 15-month period. He stopped injections after this period, as he was still making good progress with his retraining programme, and has not had any injections since, i.e., more than 3 years now. He can play any piece of the orchestra repertoire. The other flute player, Subject 4, took only over 2 months off work and

returned to orchestra playing while following the retraining programme. She made steady progress over the 4-year period and still plays as principal flute in the orchestra.

One of the guitar players, Subject 2, was an amateur who had been affected by severe dystonia for over 25 years (Table 2). He made enough progress to be able to perform simple pieces in front of friends and family. The other guitar player, Subject 1, was a professional guitar teacher. He made excellent progress during the first year of retraining and was able to play more challenging pieces with minimal occurrence of abnormal movements. Despite a slight deterioration at the 4-year follow-up, he is still able to fulfil his professional duties.

Strengths and Limitations of the Study

The results from the present study were very encouraging, and this study represents the first attempt to monitor musicians affected by FHD over the long-term, using a quasi-experimental, repeated measures design with nine standardised follow-up sessions over 4 years for all participants. The trends observed may be explained by the combination of two retraining strategies, task-specificity of the protocol, and the use of outcome measures which proved reliable and responsive to change. There were, however, several limitations.

First, the study design meant that there was no control group for this study since recruitment of participants was limited to the individuals with dystonia available during the study recruitment period. In view of the results obtained by Zeuner et al.,³⁸ who concluded that nonspecific retraining was as effective as task-specific motor retraining in subjects affected by writer's cramp, future studies on musician's FHD should consider control groups in order to take account of possible threats to internal validity.

Second, the sample was small ($n=4$ at follow-up), and therefore the deterioration of Subject 1's dystonia had a significant impact on the FAM mean score and the metronome speed mean score for the medium-difficulty piece. Future studies should endeavour to use larger samples.

Third, no inter-rater reliability tests were carried out for the FAM scores for the initial study and this follow-up, and the FAM scores obtained should therefore be treated with some caution.

Fourth, due to the wider confidence intervals obtained for the results from the intra-rater reliability (day 1) and inter-rater reliability (month 2) tests regarding the TCS, the scores obtained over the 4-year period with this scale should be treated with some caution.

Finally, since the initial 12-month retraining protocol combined constraint-induced therapy and motor control retraining, the improvements shown cannot be attributed to one intervention alone, but to a combination of both.

CONCLUSION

The present study demonstrated that a 12-month intensive retraining protocol combining constraint-induced therapy and motor control retraining was associated with long-term bene-

fits for three musicians with FHD who received no other form of treatment and a fourth who received botulinum toxin injections before completing the initial intensive retraining protocol. Indeed, the progress achieved during the initial 12-month period was maintained at follow-up at year 4, and this has not been reported before. Furthermore, the progress achieved was maintained with a minimum amount of daily specific practice, i.e., 15 to 30 minutes. 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 plasticity changes are maintained in the long-term.

The study findings also suggest that this combined retraining protocol needs to be carried out for at least 6 months before significant changes in FAM scores are noted. This trend was confirmed by the significant improvements in dystonia evaluation scale scores obtained towards the end of the study period, thereby confirming what the present authors had suggested in their initial study, i.e., a minimum of 12 months of specific retraining is recommended.

Future studies could endeavor to ascertain whether the observed trends can truly be explained by this combined behavioral intervention, using a randomized controlled study design and larger samples.

When considering both the present study and the initial study,³⁶ the results from the reliability tests carried out for the FAM scale, the TCS, and ADDS are encouraging and show that these rating scales are reliable tools. However, more work is necessary to evaluate their validity.

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