

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

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Focal hand dystonia (FHD) in musicians is a painless 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 behavioural therapy intervention, aimed at normalising 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 recordings 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 hypothesised 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 post-hoc 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; 25:149–161.

LITERATURE REVIEW

Focal task-specific dystonia (FTSD) affecting musicians' hands (also termed focal hand dystonia, FHD) is a painless

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motor disorder characterized by an involuntary loss of control and coordination of individual finger movements.¹⁻⁴ 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.^{3,5,8-12} It more often involves digits 3, 4, and 5 (D3 to D5) of the hand^{2,4,13,14} and is thought to be related to the intense and prolonged practice of rapid, alternating, and highly precise finger movement patterns.^{2,4,5,14} The condition can be disabling enough to curtail a professional career.^{7,9,15,16}

Prevalence Amongst Musicians

FTSD has been estimated to affect between 5% and 14% of musicians consulting performing arts clinics in the US.^{8,15,17} This would give an estimated prevalence of 0.2% to 0.5% in the population of professional musicians.^{3,4,18} Focal hand dystonia (FHD) occurs much more frequently in males than females. One study^{11,19} revealed that 73% of instrumentalists affected were men, two others reported 80%¹⁴ and 83%.⁵ Symptoms usually begin in the third or fourth decade,^{4,11,14,15} when musicians are at the peak of their professional career. Every instrument group is affected, but guitarists, pianists, and woodwind players are most commonly affected,^{11,15} representing 70% of patients with dystonia in one study.⁵ 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.^{3-5,7,9,10,13-15}

Neurophysiology of Focal Dystonia

Extensive research has been carried out on the neurophysiology of FTSD in recent years, and findings converge towards abnormal or decreased reciprocal inhibition and increased excitation at spinal cord, brainstem, and cortical levels. This would explain the co-contractions of agonist and antagonist muscles observed in FTSD.^{1,6,8,10,11,20,21} Decreased inhibition in the sensory cortex could drive excessive motor output.^{20,22,23} Transcranial magnetic stimulation studies have shown increased motor cortex excitability^{20,24} and decreased cortical inhibition^{23,25} in individuals with writer's cramp, and

deficiencies in sensory integration and abnormalities in sensorimotor integration in musicians affected by FTSD.^{3,4,23,26} 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.^{3,20,21}

Byl et al.,²⁷ in a primate study in which two monkeys were trained on a repetitive behavioural task, showed that motor disturbances developed with degradation of hand movement reminiscent of FHD. This was accompanied by a disordered cortical representation of the hand in area 3b of the somatosensory cortex. These findings suggested that extensive repetitive and simultaneous stimulation of the digits may contribute to a maladaptive cortical reorganisation of the representational zones of the digits and form the basis of FHD.^{2-4,11,12,14,23,27}

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 nonaffected musicians, and a control group of nonmusicians, 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 nondystonic musicians and controls.² Similar findings emerged from a study by Bara-Jimenez et al.,²⁸ and the authors suggested that this maladaptive cortical reorganisation may be linked to altered sensory perception^{4,23} with abnormalities in temporal and spatial discrimination, which they demonstrated in FHD subjects affected by writer's cramp.^{29,30}

Management: Behavioural Therapy Interventions

The management of FHD remains imperfect and uncertain,^{11,15,16,19} and to date there is no "cure."³ In a recent retrospective study looking at the long-term outcome of a wide range of treatment strategies in 144 patients, Jabusch et al.³¹ revealed that 54% of dystonic musicians had improved. Oral anticholinergic medication and botulinum toxin injections are, however, palliative, and repeated injections are necessary.^{3,11,14,31} Some authors believe that the strong linkage of musicians' movements and emotions to the limbic system may enhance development of memory processes, which would explain the difficulties in retraining appropriate movement patterns.⁵

With the recent advances in the neurophysiology of FTSD and brain imaging, new behavioural therapy interventions have emerged,^{3,5,11} although they have only been performed on small sample groups ($n = 11$).⁷ Byl and McKenzie³² used various methods of sensory retraining in a group of 12 patients (5 musicians) with FHD. The patients improved significantly in obtaining motor control, accuracy, sensory discrimination, and physical performance. However, there was no control group and the intervention was carried out over a short period (3 to 6 months).³²

Based on the assumption that behavioural mechanisms underlie both the cortical disorder and the involuntary loss

of coordination,^{2,33,34} Candia et al.³⁴ developed a novel treatment intervention, sensory motor retuning (SMR), using constraint-induced therapy in an attempt to reverse the cortical fusion observed in FHD patients.² By immobilising one or several digits "compensating" for the dystonic movements, the "dystonic" finger was required to carry out repetitive exercises in coordination with the remaining free digits.^{33,34} Eleven musicians with dystonia were studied, but there was no control group. Measurements of outcome were made using an ordinal dystonia evaluation scale (DES) and a dexterity and displacement device, measuring smoothness of movement. Pianists and guitarists showed a significant improvement in DES scores between pretreatment, post-treatment, and follow-up scores ($F_{2,20} = 16.5$, $p < 0.0001$) and in smoothness of movement ($t = 2.9$, $p < 0.05$).³⁴

However, Candia et al.³⁴ analysed their ordinal DES scores using a 1-way and then 2-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.³⁵ Given this lack of detail and the small sample, their statistical significant results should be treated with some caution. The woodwind players did not improve, perhaps due to the fact that they were instructed to blow into their instrument only occasionally. Indeed, a recent study by Hirata et al.³⁶ showed altered hand and mouth relationship (reduced distance between lip and hand representations) in the somatosensory cortex of musicians affected by embouchure dystonia.

Sakai³⁷ developed a motor control retraining technique, named "slow-down exercise" (SDE), on 20 pianists affected by FHD. An exercise programme was developed in which patients 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 DES scores over time (0 to 5 DES), 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 standardised 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.

Finally, Zeuner et al.,³⁸ in a study involving 10 patients with writer's cramp, used a motor training model similar to Candia et al.,³⁴ using constraint-induced therapy. Subjects trained for 4 weeks. In keeping with Candia et al.'s results, they showed a significant improvement ($p = 0.042$) using the ordinal arm dystonia disability scale³⁹ (ADDS). However, they reported minor objective improvements in handwriting and no significant changes in their neurophysiological indicators, measured by transcranial magnetic stimulation and electroencephalography (EEG). They concluded that 4 weeks of motor retraining was not sufficient to reverse motor cortex abnormalities.

In a more recent study, Zeuner et al.⁴⁰ addressed the question of whether motor retraining needed to be task-specific



FIGURE 1. Dystonic patterns for subjects 1 and 3.

or not to retrain subjects affected by writer's cramp. One group of subjects ($n = 10$) carried out a task-specific motor retraining programme for 8 weeks, using splints.³⁸ The other group ($n = 11$) followed a non-task-specific retraining programme practising finger movements with a therapeutic putty. There were no writing exercises for this group. The affected forearm and hand had been immobilised for 4 weeks before starting the training programmes. Interestingly, the results of the repeated measures ANOVA for the ADDS were statistically significant for the factor "time" ($F_{2,4;48.1} = 3.80$, $p = 0.023$).⁴⁰ However, the interaction "time" by "type of training" was not statistically significant ($p > 0.2$).⁴⁰ Indeed, a comparable progress was achieved by both groups with regards to ADDS scores and objective improvements in handwriting. The authors concluded that non-task-specific retraining was as effective as task-specific motor retraining with splints.

Study Aim

The aim of the current study was to investigate the effects of a combined behavioural 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 behavioural interventions. A standardised protocol was used. Subjects were tested at regular intervals during the study period, whilst playing an easy piece and a medium-difficulty piece. The frequency of abnormal movements scale (FAM) developed by Spector and Brandfonbender,¹⁶ the change in metronome speed achieved during SDE,³⁷ and two ordinal dystonia evaluation scales (DES) were chosen as outcome measures. It was hypothesised that there would be significant differences in FAM scores and metronome speeds achieved between testing occasions over time for both pieces.

METHODS

The study was approved by the Research Ethics Committee of the School of Health and Social Care at Glasgow Cale-

donian University. All participants provided written informed consent.

Subjects

Eight instrumental musicians, affected by FHD (Fig. 1), volunteered to participate in the study. Six were professional musicians, two were amateur musicians. There was only one female in the group. Table 1 summarises the subjects' characteristics.

Each subject met the inclusion criteria for the study^{34,41}: 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 syndromes; they were not undertaking any other medical treatment or therapy for their dystonia.

Additionally, each subject met all the following clinical inclusion criteria⁴²: 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

Neurological tests were carried out on both hands to ascertain normal sensory function^{16,32,34,41,43,44} (Table 2), using worksheets to record accuracy. A screen was used to blindfold subjects to the tests.^{41,45,46}

Threshold tests included temperature, light touch, and pinprick.^{34,44,45} For temperature, 10 random areas were tested for each hand, palmar and dorsal aspects, with test tubes. Light touch and identification of any area of anaesthetics were tested with cotton wool. Pinprick, for pain, was tested using a grid dividing the palmar aspect of the hand into 20 zones (14 zones represented the phalanges, 6 zones for the palm).⁴⁵ Functional tests included the static two-point dis-

TABLE 1. Subject Characteristics

Subjects	Instrument	Gender	Age* (yrs)	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‡	Accordion	Male	30	Right wrist, D2, D3, D4	Extension	2005	N/A

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

†Compliance: percentage of days when the protocol was practised during the 12 month- period.

‡These subjects did not complete the study: subject 3, despite making good progress, was excluded after month 10 after receiving a botulinum toxin injection; subject 7 withdrew after month 6 and subject 8 after month 2 due to lack of progress.

crimination test.^{29,31,38} Only the palmar aspects of the fingertips were tested⁴⁵ with a two-point discriminator disk (Touch-Test, North Coast Medical Inc., Morgan Hill, CA, USA). The researcher alternated randomly between one and two points (5 mm), making sure each fingertip had been stimulated with one and two points. Light pressure was applied to the fingertip in a longitudinal orientation. A score of 7 or more out of 10 correct responses was considered normal.⁴⁵

Sensory perceptual tests, measuring the ability to discriminate tactile stimuli with the hands, can be used as a clinical estimate of cortical somatosensory processing.⁴¹ The tactile

finger recognition (agnosia), tactile form recognition (stereognosis), and fingertip number writing (graphaesthesia) tests were included in the present study.^{32,34,41,42,44} The Halstead-Reitan neuropsychological test battery⁴⁶ was preferred to the Sensory Integration and Praxis Tests (SIPT)^{32,41-43} for administering these tests. This was because the SIPT was originally developed to assess child development, has limited normative data on adults, and has reduced sensitivity for detecting differences in adults due to ceiling effects.^{41,48,49} On the other hand, the Halstead-Reitan test battery was designed to test adults, contains normative data on adults, and has been

TABLE 2. Neurological Screening Tests for the Dystonic Hand

Test	Subject 1	Subject 2	Subject 3	Subject 4	Subject 5	Subject 6	Subject 7	Subject 8
Reflexes*	Normal	Normal	Normal	Normal	Normal	Normal	Normal	Normal
Temperature: hot/cold†	10/10	10/10	10/10	10/10	10/10	10/10	9/10	9/10
Light touch	Normal	Normal	Normal	Normal	Normal	Normal	Normal	Normal
Areas of anaesthesia	None	None	None	None	None	None	None	None
ROM all digits‡	↓ D3-4 ^a	Normal	Normal	↓ D3-4 ^a	Normal	Normal	Normal	↓ D3-4 ^a
Power§	5/5	5/5	5/5	4/5 ^b	5/5	5/5	5/5	4/5 ^c
†Pinprick: sharp/dull	39/40	39/40	39/40	37/40	37/40	39/40	39/40	40/40
Joint position: up/down†	5/5	5/5	5/5	5/5	5/5	5/5	5/5	5/5
Static two-point discrimination¶	9/10	9/10	10/10	8/10	10/10	10/10	9/10	9/10
Tactile finger recognition (agnosia)†	20/20	20/20	20/20	20/20	20/20	20/20	20/20	20/20
Tactile form recognition (stereognosis)†	8/8	8/8	7/8	8/8	8/8	8/8	8/8	8/8
(time to complete tests, in sec)¶	(10.10)	(14.67)	(11.04)	(10.12)	(12.44)	(11.75)	(13.56)	(17.19)
Fingertip number writing (graphaesthesia)†	20/20	18/20	18/20	20/20	20/20	20/20	17/20	20/20

*Biceps, brachioradialis, triceps.

†Number of accurate answers out of total possible score.

‡ROM, range of movement. Gross test carried out.

§Oxford scale: 0 to 5.

¶||Tested at 5mm with a two-point discriminator disk.

¶Total time, in seconds, to complete the series of tests twice for that hand.

^aD3/D4 Abduction ROM was reduced compared to non-affected side.

^bD3/D4 abduction power was reduced to 4/5.

^cD2/D3 and D4/D5 abduction power were reduced to 4/5.



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

extensively validated.⁵⁰ Tests were carried out following the detailed administration procedures designed by the authors.⁴⁶

Reflexes and gross range of movement of the fingers (flexion and extension of all joints, abduction, adduction) and thumb (extension, flexion, abduction, opposition) were tested. Muscle power was tested using the Oxford scale⁵¹ for the dorsal interossei (abduction) and flexor digitorum profundus muscles of the fingers, grip strength, the thumb extensor, flexor and abductor muscles. The joint position test was also carried out for one joint (chosen randomly) per digit, as described by Lindsay and Bone.⁵²

Constraint-induced Therapy

The concepts of sensory motor retuning (SMR), as described by Candia et al.,³⁴ were applied in this study. Splints were used to immobilise the wrist and fingers, depending on the pattern of the dystonia (Fig. 1, Table 1).^{34,38,40} The finger that trained was not splinted, while one or several adjacent fingers were splinted to reduce co-contraction.^{34,38,40} Determining which fingers would need to be splinted was done by immobilising in turn each finger participating in the abnormal movement pattern while the musician was playing with the remaining digits. In all cases, immobilising one or two digits enabled a freer and more independent movement pattern of the main dystonic finger with reduced co-contractions (Fig. 2).³⁴

Retraining involved playing specific finger combinations, aimed at normalising movement patterns for the main dystonic finger, with a splint in situ immobilising the selected adjacent fingers. The splints were made of a thermoplastic material (Ezeform, Sammons Preston Rolyan, Cedarburg, WI, USA) that could be moulded to the subject's hand while holding a normal playing posture (Fig. 2). This allowed retraining to occur in a task-specific posture.^{2,7,9,21}

The specific finger exercises for subject 3 are shown in Table 3. The first week of retraining involved intensive constraint-induced therapy practice, for 2 hrs/day. Each subject would play each sequence continuously for 10 minutes, with a 2-minute rest between sequences; five sequences therefore

were performed in 1 hour.³⁴ The sequences were designed by the main author (PB). A 10-minute rest was given after the first series was completed with one splint. Another series of finger exercises was carried out with the same splint, or another splint where applicable (Table 3). A metronome (Zen-on metrina, quartz metronome, Seiko Instruments & Electronics Ltd, Japan), previously tested for accuracy and precision, was used at all times. Speed was modified every 2 minutes during the performance of each sequence in order to challenge the subjects and make the motor control retraining more specific (Table 3).^{34,53} The maximum speed was chosen so that the subject would still be able to perform the finger sequences without any abnormal movement pattern occurring. Speeds therefore would be different for each subject and even according to the finger being splinted.

Contrary to Candia et al.,³⁴ the wind players were asked to play continuously during a specific sequence, alternated by a sequence without blowing in the instrument. Although this was tiring, retraining had to be as task specific as possible⁵³ and therefore take account of finger-mouth coordination as a possible source of dystonic patterns.^{34,36}

Subjects were monitored by the researcher at day 1 and day 8 of the intensive period. Following this period, they were asked to practise these finger sequences daily with their splints for 30 to 60 minutes.³⁴ They were encouraged to modify (up or down) the metronome speeds, as long as they could manage without the development of abnormal movement patterns. Subjects were then re-assessed every 2 months for 12 months.

Compliance was ascertained by using a calendar worksheet, requiring subjects to tick each day after having done their constraint-induced protocol (Table 1).

Motor Control Retraining

After completion of the first week of intensive constraint-induced therapy, the second aspect of the protocol, motor control retraining, was added. This did not involve using splints, but was based on slow-down exercise (SDE) retraining, as described by Sakai.³⁷ Subjects selected two pieces of

TABLE 3. Constraint-Induced Therapy Protocol for Subject 3, Flute Player

Splinted Finger	Finger Combinations	Initial Speed (bpm)*
D5	D4-D3-D2-D1-D2-D3 blowing	50-54-58-63-40 2 notes per beat
	D3-D4-D2-D1-D4-D2 blowing	
	D4-D3-D4-D2-D4-D1 blowing	
D4	D2-D3-D5-D3-D1-D5 blowing	44-48-52-56-40 2 notes per beat
	D3-D2-D5-D2-D3-D5 blowing	
	D3-D5-D2-D5-D1-D5 blowing	
	D5-D2-D3-D5-D1-D5 blowing	

*Metronome speed in beats per minute.

music with the main author (an easy piece or excerpt and a more challenging one), both of which were used for data collection^{16,37} and had to trigger the abnormal dystonic movements. For each piece, the researcher, using a metronome, reduced the speed to the level at which subjects could play with no noticeable abnormal (or minimal abnormal) movements. This level became the “baseline” speed at which subjects were asked to practise their pieces. Subjects were instructed to use a metronome when practising the two pieces at this “baseline” speed to encourage normal rhythm and movement pattern.³⁷ They were also encouraged to use a mirror for visual feedback of normal movement.³²

Subjects were allowed to increase the metronome speed by one or two increments on a weekly basis, as long as they could manage to play with no or minimal abnormal movements at the new speed. The date and speed were recorded on a worksheet. If unsuccessful, subjects would continue practising at the initial speed set out previously.

They were asked to practise SDE retraining for 30 min/day, in addition to the 30 to 60 minutes of constraint-induced therapy. For the wind players, SDE was always done while blowing into the instrument, for reasons given earlier.³⁶ Compliance was monitored with the same calendar worksheet, as previously described (Table 1).

Free Playing

Subjects were encouraged to practise any other piece, exercise, or musical excerpt using these SDE retraining principles up to 30 min/day, according to available time, in order to introduce task variations and optimise cortical plasticity and motor control.⁵³ They were also allowed to practise any musical piece at normal speed for a few minutes in order to maintain motivation and compliance.^{34,37}

Data Collection and Follow-up

All outcome measures were taken at day 1, day 8, and then every 2 months for 12 months. Constraint-induced therapy

retraining started on day 1, after the first data collection. The main researcher (PB) carried out all the tests.

At each testing session, participants were rated by the researcher once using two ordinal dystonia scales, the Tubiana and Chamagne scale (TCS)⁵⁴ and the arm dystonia disability scale (ADDS)³⁹ (Table 4). Although it has not been validated, the TCS was chosen because it had the highest range of values (0 to 5) and was written specifically to assess musicians. The Burke-Fahn-Marsden scale (BFM) had been studied and considered as valid (Spearman's $\rho > 0.89$, $p < 0.01$); it showed good intra-rater (Spearman's $\rho = 0.98$) and inter-rater (Spearman's $\rho > 0.85$) reliability ($p < 0.01$).⁵⁵ Moreover, the same authors³⁹ developed additional scales to allow a more specific and sensitive assessment of focal dystonias. The arm dystonia disability scale (ADDS) is one of those, which includes a subsection dealing with playing a musical instrument, and therefore was used in the present study.

The frequency of abnormal movements scale (FAM), developed by Spector and Brandfonbrener,¹⁶ 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 an objective rater.¹⁶ The FAM score is the number of abnormal movements per second of instrumental playing. The FAM has shown high intra- and inter-rater reliability (intraclass correlation, ICC = 0.92, Spearman's $\rho = 0.87$), high internal consistency (Cronbach's $\alpha = 0.96$), is responsive to change ($p < 0.06$), and proved superior to the BFM and ADDS with regard to inter-rater correlation, concordance, and internal consistency.¹⁶

A digital video camera (Lumix DMC-FX3, Panasonic, Osaka, Japan) was used to videotape subjects playing the two musical pieces. An easy and a medium-difficulty piece were chosen in the event that subjects may not be able to play the more challenging piece without stopping.¹⁶ Subjects were asked to play continuously for at least 3 minutes. Each piece was recorded twice, in order to assess subject “inter-occasion” reliability at each testing session. A 3-minute window of playing was used for data analysis. A tempo was chosen for each piece and was considered by the subject as being the “normal” tempo for that piece. The same tempi were used throughout the 12-month period at each follow-up session to enable standardisation.

SDE scores were determined as previously described.

Data Analysis

Video segments were transferred to a computer for analysis by the main author. Each video was scored twice, with a 1-week interval, to avoid possible memory bias when assessing intra-rater reliability.

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).^{16,35}

TABLE 4. Ordinal Dystonia Scales Used to Monitor Effects of Treatment

TCS	Stage Definition	ADD5	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 scale⁵⁴; ADD5, arm dystonia disability scale.³⁹

The Spector and Brandfonbrener¹⁶ procedure for counting abnormal movements was, however, modified within this study. For example, it was felt that if the subject maintained his dystonic finger flexed for long periods without attempting to reset it to a normal playing position, this should be seen as an abnormal movement pattern. It therefore was decided to approximate the number of abnormal movements for this finger to the number of metronome beats for which it would stay flexed, until the subject attempted to reset his finger to a normal position.

Statistical Analysis

SPSS software version 16 was used for descriptive and inferential analysis (SPSS Inc., Chicago, IL). Differences between affected and non-affected hands for some of the sensory perceptual tests were assessed using either paired *t*-tests or paired Wilcoxon test.

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); between-sessions comparison (eight testing sessions over 12 months). Intra-rater and inter-occasion reliability were tested using an intraclass correlation model: ICC Model (2,1). A two-factor parametric repeated measures analysis of variance (ANOVA) was subsequently carried out, with the number of abnormal movements (AM) per second (AM/sec) as the dependent variable. Diagnostics were performed, after the model was fitted, to confirm normality and homoscedasticity.³⁵

For the DES scores, one-factor repeated measures parametric ANOVA models were used, and normality and homoscedasticity were confirmed.³⁵

For the metronome speed scores, a two-factor parametric repeated measures ANOVA was carried out, and once again appropriate diagnostics were performed.³⁵

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

Neurological Screening

Table 2 presents the neurological test results for the dystonic hand only. Results were within normal limits for all tests and for both hands when considering normative data available.^{45,46}

Moreover, statistical analysis was carried out for some of the sensory perceptual tests in order to determine whether there were significant differences between the hand affected by FHD and the nonaffected hand for all subjects. For agnosia, all subjects obtained maximum scores for both hands, and testing was therefore not required. For stereognosis, a paired *t*-test was used (data were normally distributed) to compare the time taken to complete the series of tests for each hand (Table 2). There were no significant differences between the affected hand and the nonaffected hand ($t = 1.18$, $p = 0.278$). For graphaesthesia, a paired Wilcoxon signed-ranks test (data were not normally distributed) was performed to compare scores obtained between the affected hand and the nonaffected hand, and no significant differences were found ($W = 1$, $p = 0.210$).

Intra-rater and Inter-occasion Reliability Tests for FAM Scores

Each video clip was scored twice to evaluate intra-rater reliability. Results from the ICC Model (2,1) showed excellent reliability, 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). Therefore, the mean of the two scores for each clip was used when assessing inter-occasion reliability (within-subject and within-day variation), since each piece was recorded twice.

For the inter-occasion reliability test, the ICC Model (2,1) revealed generally good results, but the 95% CI were wider (Table 5). In two situations (month 6, medium piece; month 10, medium piece), the ICC values were not significant (Table 5). However, a paired *t*-test revealed that no statistical change took place between occasions for these two situations.

TABLE 5. Inter-Occasion Reliability Test

Testing Session	Piece	Pairs	ICC	p-value	95% CI
Day 1	Easy	8	0.976	< 0.001	0.895–0.995
	Medium	5	0.990	< 0.001	0.795–0.999
Day 8	Easy	8	0.907	< 0.001	0.630–0.980
	Medium	5	0.995	< 0.001	0.950–0.999
Month 2	Easy	8	0.974	< 0.001	0.886–0.995
	Medium	5	0.960	0.002	0.681–0.996
Month 4	Easy	7	0.984	< 0.001	0.910–0.997
	Medium	5	0.907	0.009	0.341–0.990
Month 6	Easy	7	0.981	< 0.001	0.818–0.997
	Medium	5	0.739	0.058	0.000–0.970
Month 8	Easy	6	0.996	< 0.001	0.972–0.999
	Medium	4	0.955	0.003	0.520–0.997
Month 10	Easy	6	0.966	< 0.001	0.785–0.995
	Medium	4	0.782	0.063	0.000–0.984
Month 12	Easy	5	0.778	0.044	0.000–0.975
	Medium	3	0.995	0.003	0.919–1.000

ICC, intraclass correlation; 95% CI, 95% confidence interval.

It was concluded that the large variability between subjects, in a small sample with some missing data for these situations, caused methodological problems with regards to the ICC model with wider CI than expected. Since the vast majority of results were good, the model was considered robust and the mean of the two occasions was therefore used for the analysis of variance ANOVA.

FAM Scores: Comparison over Time

Some data were missing for three subjects who did not complete the study period. Despite making good progress, Subject 3 received a botulinum toxin injection after month 10 and was excluded after that point. Subjects 7 and 8 decided to withdraw after month 6 and month 2, respectively, due to lack of progress. Furthermore, three subjects were unable to play the more difficult piece due to the severity of their dystonia.

However, the results from the two-factor repeated measures ANOVA showed a very significant trend: the number of abnormal movements per second of instrumental playing decreased with time for both pieces combined ($F = 6.32$, $df = 7$, $p < 0.001$) (Fig. 3), indicating a significant improvement with normalisation of movement patterns over the 12-month period. Since these results were very significant, Tukey's post-hoc test was carried out to determine which time periods were statistically significant over the 12-month period. Results showed significant differences between day 1 and any period from month 8 and between day 8 and any period from month 8. This would suggest a late effect of therapy.

FAM Scores: Comparison Between Easy and Medium-Difficulty Pieces

The results showed a statistically significant difference between the two pieces ($F = 6.36$, $df = 1$, $p = 0.014$) (Fig. 3).

The number of abnormal movements 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 ($F = 0.48$, $df = 7$, $p = 0.844$). 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, subjects generally found the medium-difficulty piece harder to play, and this is reflected by the larger number of abnormal movements per second. After 6 months, this difference virtually disappeared (Fig. 3).

Dystonia Evaluation Scale Scores

The results of the one-factor repeated measures ANOVA carried out for each dystonia evaluation scale revealed a significant improvement in scores for both the Tubiana and Chamagne scale ($F = 4.96$, $df = 7$, $p < 0.001$) and the Arm Dystonia Disability Scale ($F = 3.60$, $df = 7$, $p = 0.004$) over the treatment period (Fig. 4).

Tukey's post-hoc tests for the Tubiana and Chamagne scale showed statistically significant differences between day 1 and any period from month 10, between day 8 and any period from month 10, between month 2 and month 12, between month 4 and month 12, and between month 6 and month 12.

Tukey's post-hoc tests for the Arm Dystonia Disability Scale showed statistically significant differences between day 1 and month 12 and between day 8 and month 12. These post-hoc tests would suggest a late effect of therapy.

Metronome Speed Scores

The results from the two-factor repeated measures ANOVA showed a very significant trend: the metronome speed

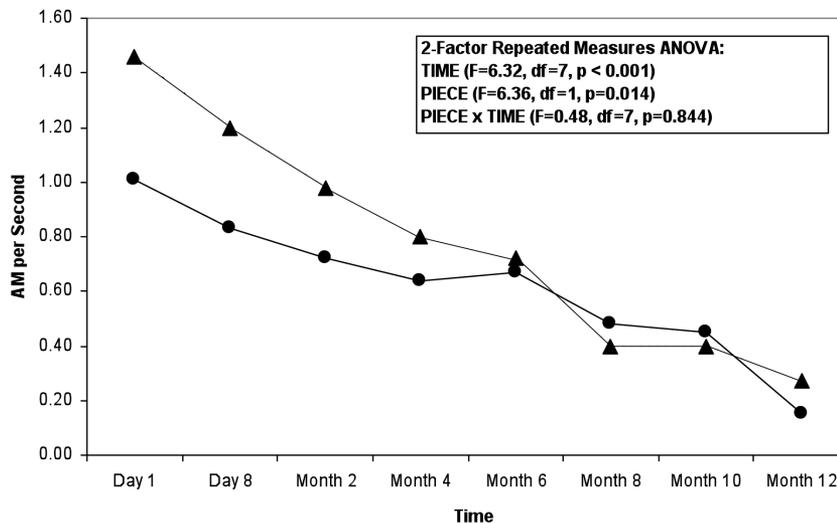


FIGURE 3. Frequency of abnormal movements (AM) scale: mean values for each piece and all subjects. Easy piece, circles; medium-difficulty piece, triangles.

achieved by subjects without occurrence of abnormal movements increased significantly over time for both pieces combined ($F = 20.73$, $df = 7$, $p < 0.001$) (Fig. 5). Tukey's post-hoc test showed significant differences between day 1 and each other adjacent testing time point. This continued up to month 6, being significantly different from both month 10 and month 12.

The results showed no statistically significant difference between the two pieces ($F = 0.36$, $df = 1$, $p = 0.553$), and no significant interaction between the factors "time" and "piece", as shown in Figure 5 ($F = 0.74$, $df = 7$, $p = 0.639$). Indeed, the steady improvement over time is very similar for both pieces, and would explain the lack of a significant interaction effect.

DISCUSSION

Effects of the Treatment Protocol on FHD: FAM Scores

The present study revealed that a retraining protocol combining constraint-induced therapy (SMR) and motor control retraining (SDE) led to significant improvements in FAM scores over time for both the easy and medium-difficulty pieces of music (Fig. 3). These results demonstrate a trend toward normalisation of movement patterns when tailored retraining takes place in a task-specific environment⁵³ (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 abnormal movements per second of playing between the two pieces virtually disappeared after 6 months of therapy. These findings are in keeping with the theory that, in the presence of maladaptive and use-dependent cortical reorganisation,^{2,4,12,23,28,56} 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.^{2,4,12,34,56}

Of important note, the results of the Tukey's post-hoc test suggested that retraining had to be carried out for at least 8 months before significant changes in FAM scores were noted. These findings were made possible by the study design (repeated measures design), since all subjects were followed over a 12-month period and were tested at similar time intervals. This notion of a minimum period of retraining has not been reported before in musicians with FHD. Indeed, in the studies by Candia et al.³⁴ and Sakai³⁷, the follow-up period for the final measurement was not standardised and varied greatly between subjects.

Although this was not the main aim of their study, Spector and Brandfonbrener¹⁶ analysed 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. The authors attributed this lack of improvement to either a deficiency in therapy with decreased subject compliance, between-subject variations, or a defect in the FAM method itself. Their results contrast with the present study findings, and several explanations are possible.

Firstly, there was no indication on how Spector and Brandfonbrener monitored compliance between week 1 and month 6. In contrast, the present study used a calendar worksheet that was handed back to the researcher at each testing session, to monitor subject compliance closely. A score was derived, expressed as a percentage of days of practice over 365 days, and was found to be very high (Table 1). Secondly, the endpoint of their study was after 6 months of retraining, whereas the present results suggest that a minimum duration of retraining of 8 months may be required before noticing statistically significant differences in FAM scores. In summary, when considering the results of the present study, the lack of improvement observed by Spector and Brandfon-

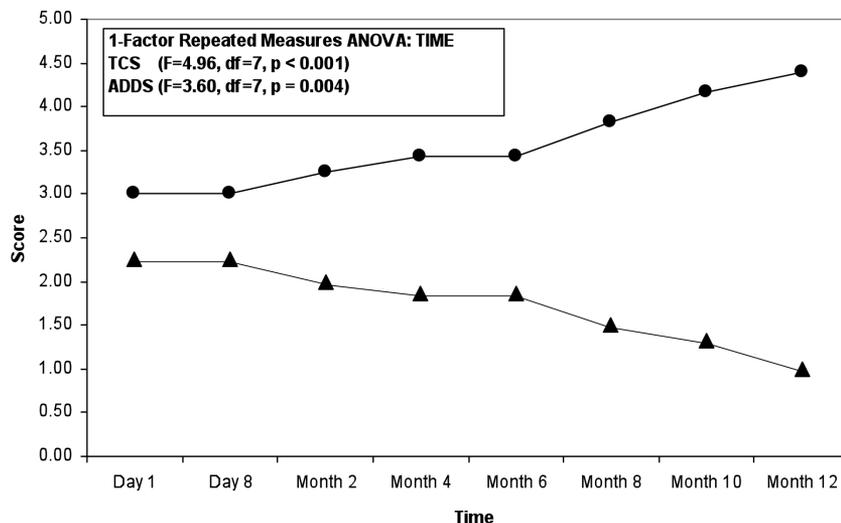


FIGURE 4. Mean values for ordinal dystonia evaluation scale scores. TCS, Tubiana and Chamagne dystonia scale, circles; ADDS, arm dystonia disability scale, triangles.

brener¹⁶ may be due to a lack of subject compliance and a short retraining period rather than a defect in the FAM method. This concept of a “minimum” duration of retraining has already been mentioned by Zeuner et al.³⁸ in their study on 10 patients with writer’s cramp, and in the absence of changes in their neurophysiological measures, the authors concluded that 4 weeks of retraining was not long enough to produce excitability changes or reorganisation of the motor cortex.

The results of the present study are in keeping with the findings of Candia et al.³⁴ However, in their study, the wind players (two flautists and one oboist) did not improve. One of the reasons the authors gave for this failure is that wind players were only asked to blow occasionally into their instruments and that finger-mouth coordination was therefore not addressed in the study protocol.³⁶ Performing the finger exercises while simultaneously blowing may take account of finger-mouth coordination in FHD subjects and facilitate an increased differentiation between digit and mouth cortical representations.^{34,36} The present study protocol took account of the findings of Hirata et al.³⁶ by asking the wind players to blow for long periods of time during SMR and during SDE. This may explain the progress of the majority of the wind players over time (Table 1). These comments would also apply the study by Spector and Brandfonbrener,¹⁶ since their subjects were not blowing into their instruments or using their bows during SMR.

Furthermore, the three wind players in the study by Candia et al.³⁴ 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, comments made by Zeuner et al.³⁸ on treatment duration, and the findings of the present study suggesting a minimum of 8 months of retraining, these are very short periods to make assumptions of treatment failure. However, since the present study protocol was a combination of constraint-induced therapy and motor con-

trol retraining, it is difficult to establish which aspect of treatment had the more beneficial effect.

Comparison of the FAM Scores and Dystonia Evaluation Scale Scores

The present study indicated that the retraining protocol led to significant improvements in subjective ordinal dystonia scale scores for both the Tubiana and Chamagne scale (TCS) and the arm dystonia disability scale (ADDS) over time (Fig. 4), thereby confirming the results obtained from the FAM scale and the trend toward normalisation of movement patterns. These findings are in keeping with the improvement in dystonia evaluation scale scores obtained by Candia et al.³⁴ and Sakai³⁷ in their studies on musicians. However, as already noted, their protocols were not standardised with regards to the follow-up period, contrary to the present study. This is also in keeping with the studies by Zeuner et al.^{38,40} on writer’s cramp, who used a similar repeated-measures design for their studies but with a very short retraining period.

Interestingly, the results from the Tukey’s post-hoc tests also revealed differences between the two ordinal scales used in the present study. For each scale, the post-hoc tests showed that significant changes took place near the end of the study period (month 10 and month 12), indicating a very late effect of therapy. Moreover, the results from these tests suggest that the ADDS would seem less sensitive to change than the TCS, and this may be explained by the fact that the TCS has a larger range of values (0 to 5) than the ADDS (0 to 3) (Table 4). However, there should be some caution with these results since, contrary to the ADDS, the TCS has not been validated. Furthermore, no intra or inter-rater reliability tests were carried out on these ordinal dystonia scales in the present study.

Although the findings from the dystonia evaluation scale scores confirmed the notion of a late effect of therapy

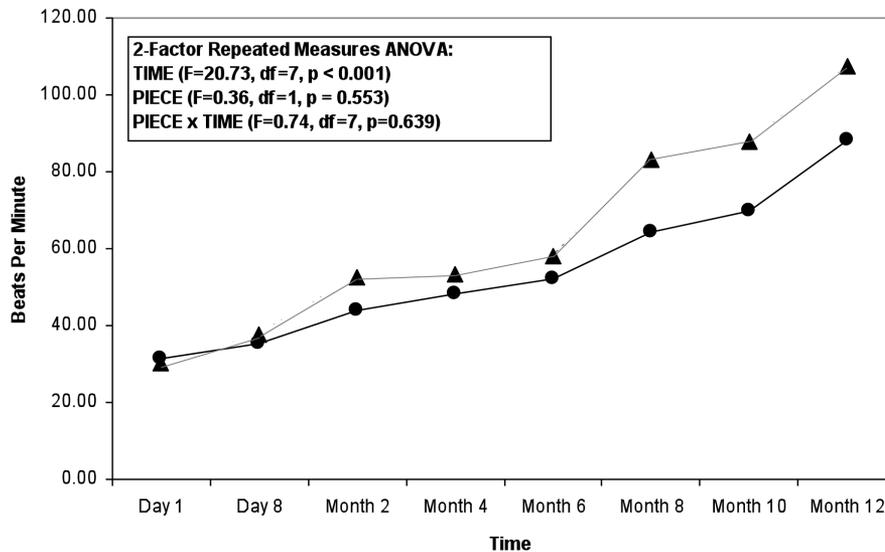


FIGURE 5. Metronome speed scores: mean values for each piece and all subjects. Easy piece, circles; medium-difficulty piece, triangles.

already obtained from the FAM scores, Tukey's post-hoc tests revealed that the FAM scale was more sensitive to change, as suggested by Spector and Brandfonbrener,¹⁶ showing significant changes taking place from 8 months of therapy (Fig. 3). Furthermore, the present findings seem to confirm what was hypothesised by Spector and Brandfonbrener,¹⁶ 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 scales are also in keeping with Zeuner et al.,³⁸ who commented on the difficulty of relying on subjective self-assessment scales and on the ADDS to rate FHD.

Finally, in the present study, the FAM scale showed excellent intra-rater reliability and good inter-occasion reliability within a robust repeated-measures design and could therefore be considered as a useful objective clinical outcome measure for musicians with FHD. It is, however, worth noting that the FAM scale measures frequency of abnormal movements, and not severity of the spasms (Fig. 1), thereby strengthening the need to have an appropriate range of measures to assess both the quantitative and qualitative aspects of the condition.

Metronome Speed Scores

The present study revealed a significant improvement in metronome speed achieved over time without occurrence of abnormal movements, for both pieces (Fig. 5). These results are in keeping with the trend observed by Sakai,³⁷ 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, in contrast to the present study. The findings of the present study seem to indicate that "change in metronome speed" could prove a

sensitive tool to measure progress in musicians' FHD. However, there should be some caution with these results since no intra- or inter-rater reliability tests were carried out on the metronome speed data.

Whereas the post-hoc test revealed a late effect of therapy for the FAM scores, the results of the Tukey's post-hoc test for the metronome speed scores showed that significant changes occurred gradually over the 12-month period (Fig. 5). This difference may be explained by the fact that the metronome speed scores did not have a finite endpoint, contrary to the FAM scores, which were converging towards "0." Indeed, subjects 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, and three subjects managed to reach or go beyond the "normal" tempi for their test pieces.

Finally, the results suggested that improvement was similar for each piece, as indicated by the trend observed in Figure 5, and the lack of significant interaction between the factors "time" and "piece."

Limitations of the Study

Firstly, the study design meant that there was no control group for this study, since recruitment of participants was limited to the patients with dystonia available during the study recruitment period. In view of the results obtained by Zeuner et al.,⁴⁰ who concluded that nonspecific retraining was as effective as task-specific motor retraining in subjects affected by writer's cramp, future studies on musicians' FHD should consider control groups in order to take account of possible threats to internal validity.

Secondly, the sample was small and this caused some methodological difficulties with regard to the inter-occasion reliability test (Table 5), due to missing data especially for the medium-difficulty piece. Similar problems had been encoun-

tered by Spector and Brandfonbrener in their study,¹⁶ and the authors had to exclude the difficult excerpt from data analysis since several subjects were unable to play it without stopping. Although the trend shown between the easy piece and the medium-difficulty piece was interesting (Fig. 3), using only one easy piece may be preferable in future studies on musicians' FHD to avoid methodological issues.

Finally, 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.

CONCLUSION

The present study demonstrated that a combined behavioural therapy using constraint-induced therapy and motor control retraining could yield a significant decrease in the frequency of abnormal movements, and a significant increase in metronome speed achieved without occurrence of abnormal movements, over a 12-month period in musicians with FHD. This possibly could suggest that normalisation of movement patterns and recovery of fine motor control in musicians affected by FHD occur through normalisation of the cortical representational maps.

The study findings also suggest that this combined type of retraining needs to be carried out for at least 8 months before significant changes in FAM scores are noted. This trend was confirmed by the significant improvements in ordinal dystonia evaluation scale scores obtained toward the end of the study period. A minimum of 12 months of specific retraining may therefore be recommended.

These trends may be explained by the combination of two retraining strategies, task-specificity of the protocol, close monitoring of compliance, a rigorously monitored treatment protocol with regular and standardised follow-ups for all participants, and the use of outcome measures which are responsive to change. Future studies could endeavour to ascertain whether the observed tendencies can truly be explained by task-specificity of the treatment protocol, using a randomized controlled study design where the control group receives a nonspecific retraining programme.

With regard to the outcome measures used, the present study validates the FAM scale as a useful objective outcome measure capable of providing quantification of the frequency of abnormal movements in musicians' FHD. Regarding the subjective scales, further work is needed in order to ascertain that the TCS is a valid tool and to compare it to the ADDS to determine whether the TCS is more sensitive to change.

Finally, owing to the neurological nature of FHD, long-term follow-up studies are required to determine whether this type of retraining programme can yield long-term benefits for musicians with FHD.

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