

Sensory–motor rehabilitation therapy for task-specific focal hand dystonia: A feasibility study

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Katherine Butler^{1,2} , Anna Sadnicka³, Jennifer Freeman¹,
Anne-Marthe Meppelink⁴, Isabel Pareés⁵, Jon Marsden¹ and
Mark J Edwards⁶

Abstract

Introduction: Medical treatments have limited long-term effect in task-specific dystonia. Whilst evidence supports the use of rehabilitation, no randomised controlled trials have been undertaken. This small-scale study determined the feasibility of implementing and evaluating a mixed sensory–motor task-specific dystonia rehabilitative programme.

Methods: Participants with writer's or musician's dystonia were recruited from a movement disorder and hand therapy clinic. Feasibility measures were recruitment rate, retention, session attendance, adherence to exercises. Self-report measures at 0, 3 and 6 months included: Arm Dystonia Disability Scale (ADDS), Tubiana and Chamagne Scale (TCS), Brief Illness Perception Questionnaire (BIPQ), Health Status (EQ-5D 5L), Clinical Global Impression Scale (CGI). Task performance was video-recorded at baseline and 6 months. At 6 months, interviews explored participant experiences of the intervention.

Results: Fifteen patients were recruited over 6 months (rate 2.5/month, musician's dystonia = 8, writer's dystonia = 7). Complete data sets were collected for twelve people (80%). The programme comprised a maximum six sessions plus daily home exercises. Nine completed the home programme at $\geq 75\%$. No adverse events were reported. Effect sizes at 3 and 6 months, respectively, were ADDS 3-month = 0.28, 6-month = 0.23; TCS 3-month = 0.13, 6-month = 0.53; BIPQ 3-month = 0.38, 6-month = 0.71; EQ-5D-5L 3-month = 0.34, 6-month = 0.59; video analysis at 6 months = 0.78. Eleven (92%) improved on the CGI. Interviews supported acceptability of the intervention.

Conclusions: This intervention was feasible to deliver with high retention, adherence and acceptability. Improvements occurred across measures. This informs the development of future randomised controlled trials.

Keywords

Musicians' dystonia, writers' dystonia, sensory–motor rehabilitation therapy, hand therapy

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Introduction

Task-specific dystonia (TSD) is a subtype of dystonia in which an abnormal posture occurs during the performance of a specific, usually highly skilled task, such as playing a musical instrument (musicians' dystonia, MD) or writing (writers' dystonia, WD). It can be very disabling, especially for professional musicians, with up to 62% of affected patients unable to continue their performance careers.¹ The pathophysiology and aetiology of TSD is not completely understood. It is thought likely to be related to sensorimotor system alterations potentially caused by interaction between the continued repetitive practice of a highly skilled movement in the face of imposed intrinsic (e.g. fatigue or injury)

¹Faculty of Health and Human Science, Plymouth University, Plymouth, UK

²London Hand Therapy, Wimpole Street, London, UK

³Sobell Department of Motor Neuroscience and Movement Disorders, UCL Institute of Neurology, London, UK

⁴Department of Neurology, University Medical Centre Groningen, Groningen, Netherlands

⁵Department of Neurology, Hospital Ruber International, Madrid, Spain

⁶Institute of Molecular and Clinical Sciences, St George's University of London, London, UK

Corresponding author:

Katherine Butler, Faculty of Health and Human Science, Plymouth University, Plymouth PL6 8BH, UK.

Email: katherinebutler@londonhandtherapy.co.uk

or extrinsic (alterations in technique or mechanical demands) changes. Correspondingly, there is experimental evidence for both motor and sensory dysfunction (such as altered inhibition within motor cortical areas and alterations in the delineation of the sensory homunculus representing the affected part)² as well as a wider cognitive context that includes an abnormal attentional focus during performance, anxiety and perfectionism.²

Existing medical treatments such as oral medications (e.g. trihexyphenidyl) and botulinum toxin injections are limited in their long-term efficacy.³ There is an increasing interest in using specific rehabilitative techniques which include: sensory re-education,⁴⁻⁸ sensory motor retraining,⁹⁻¹³ mirror therapy¹⁴ and slow-down exercise treatment.¹⁵ However, randomised controlled trials (RCTs) of these interventions have not yet been undertaken.

Given that there is pathophysiological evidence for mixed sensorimotor dysfunction in TSD, our hypothesis is that a mixed sensory-motor rehabilitation approach would be effective in improving function. There is need for a full-scale blinded RCT to investigate the efficacy of such an intervention in improving the function and quality of life of persons with TSD. However, beforehand a feasibility study is warranted to ensure the operational components are appropriate. Thus, we had two main aims. The first aim was to assess the feasibility (patient acceptability and adherence) of providing a mixed (supervised and home-based programme) of sensory-motor rehabilitative activities currently offered as "usual care" in an outpatient hand therapy practice. The second aim was to evaluate operational elements of the trial design related to participant recruitment, retention and suitability of the assessment process and outcome measures. To achieve these aims this study had several objectives (Table 1).

Table 1. Study objectives.

- To investigate the appropriateness of the eligibility criteria and determine the participant recruitment rate
- To investigate the acceptability and feasibility of this physical rehabilitation programme by determining attendance at face-to-face sessions, adherence to the home exercise programme and participant opinion of the effectiveness of individual therapy components
- To assess the suitability of outcome measures proposed for use in a future randomised controlled trial
- To obtain baseline estimates of scores (and standard deviations) on the proposed outcome measures
- To provide preliminary data on the clinical effectiveness of the rehabilitation programme
- To estimate the sample size required for a powered clinical trial to evaluate the effectiveness of this approach

Methods

This was a small-scale, single-group feasibility study. The intervention was delivered as part of routine clinical practice, at an outpatient hand therapy clinic, and the assessments were approved by the local ethics committee (North West London Research Ethics Committee 1. Ref: 11/LO/0307) and carried out at a specialist neurological hospital. Informed written consent was obtained from all participants. Figure 1 provides an overview of the study design and participant pathway.

Participants

The study recruited consecutively eligible participants over a 6-month period, from a movement disorder clinic at the National Hospital for Neurology and Neurosurgery and a private hand therapy clinic, London Hand Therapy. Participants were eligible for inclusion if they were: aged ≥ 18 years of age, with a diagnosis of TSD affecting writing or musical performance made independently by two neurologists specialising in movement disorders (MJE, AS, AM, IP) and symptoms causing sufficient impairment for the patient to request treatment. Exclusion criteria were the presence of another neurological (e.g. peripheral neuropathy) or musculoskeletal condition that could impair hand function, ongoing treatment with botulinum toxin injections into any muscles of the affected upper limb and the receipt of hand therapy or physiotherapy in the last 12 months. Of those who met the inclusion and exclusion criteria, all accepted to participate in the study apart from three people who were declined by the research team due to funding limitations.

Intervention: sensory-motor rehabilitation therapy

The intervention was designed so that it could be implemented in publicly funded clinical practice and included a self-directed home exercise program framed within clinical therapist encounters. For this feasibility study a specialist hand therapist with more than 18 years of experience treating TSD (KB) carried out the mixed sensory-motor rehabilitative therapy. The intervention combined four previously published specific treatment approaches to TSD and five generic hand therapy treatments. A summary of treatment approaches can be seen in Table 2. The treatment sessions were standardised, apart from minor changes such as accommodating positions to make them more comfortable, and notes were taken at each treatment session for all individuals.

The intention was for participants to receive a maximum of six therapy sessions over a 6-month period:

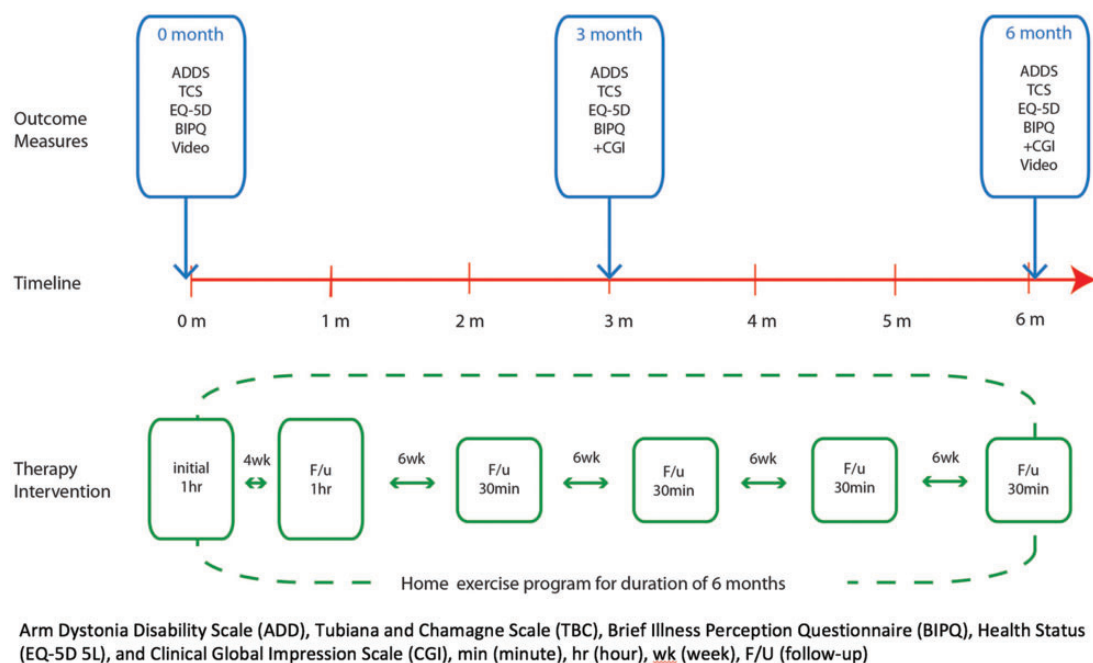


Figure 1. Study design.

initial appointment (60 min), first follow-up appointment 1 month after initial appointment (60 min) and then each further follow-up appointment at six weekly intervals (30 min each). Participants were requested to undertake a home exercise programme which was integral to this rehabilitation programme. They were asked to complete a daily log by hand or electronically. The recommended amount of each exercise was indicated on the written home exercise log and patients were asked to accurately record the time they spent undertaking the exercises.

Feasibility outcomes

Feasibility measures were recruitment rate over the 6-month period, retention, attendance at therapy sessions and adherence to the home exercise programme (quantified using the patient-reported daily log). There is no guidance regarding the minimum intensity and frequency of rehabilitation required to gain an effect; however, from a theoretical perspective it is more likely that an effect will be gained if adherence levels are high. With this in mind, together with clinical experience and feedback from patients as to what was a “reasonable” rate of adherence, we decided a priori that acceptable adherence was attendance at $\geq 75\%$ therapy sessions and completion of $\geq 75\%$ or more of prescribed home exercises. Adverse events were to be recorded in the clinical notes by the clinician and/or in the home exercise log by the patient. Participants were asked their opinion of

the effectiveness of individual therapy components on a three-point Likert scale (anchored with effective and not effective).

Clinical outcomes

The following standardised and validated patient-reported questionnaires were collected at baseline, 3 and 6 months into the intervention:

- (1) The Arm Dystonia Disability Scale (ADDs) rates the severity of impairment in hand function across daily tasks such as writing, playing a musical instrument and handling utensils. It is designed to quantify disability on a scale of 0–100% (100% = no disability).^{21–24}
- (2) The Tubiana and Chamagne Scale (TCS) rates the severity of impact of dystonia on musical performance from 0 to 5 (5 = return to concert performance).^{25–29}
- (3) The Brief Illness Perception Scale (BIPQ) is a measure of the cognitive and emotional representation of illness. It ranges the threat of an illness from 0 to 80 and a higher score reflects a more threatening view of the illness.³⁰
- (4) Health Status measured by EQ-5D 5L includes descriptive assessment across five dimensions (mobility, self-care, usual activities, pain/discomfort, anxiety/depression) and a visual analogue rating of total health. These are then converted into an index value from 0 to 1 (1 = complete health).³¹

Table 2. Summary of treatment approaches.

Treatment approach	Aim	Description and equipment	When and how much
Sensory re-education ⁴⁻⁸	Retraining and improving sensory discrimination	Exercises focus on palpating and visualising the weight, size and texture of an object, matching paired dominoes or learning to read braille <i>Equipment:</i> Common household objects (e.g. a nut, bolt, coin or paper clip), dominoes and learn to read braille books	Patient advised to complete 10 min of focused exercise twice a day
Sensory-motor retuning (SMR – orthotic) ⁹⁻¹³	Increasing hand representation in the sensory-motor cortex	Movement patterns are altered by use of an orthosis that aims to rebalance the hand by blocking the compensating movement and facilitating use and “retraining” of the dystonic part. <i>Equipment:</i> A bespoke thermoplastic orthosis that blocks the compensating movement (e.g. finger MCPJ hyperextension or wrist flexion)	Orthosis to be worn for at least 30 min per day either on its own or in combination with mirror therapy or slow-down exercise treatment
Mirror therapy ¹⁴	Re-establishing motor control to assist in retraining movement patterns	Instant visual feedback of an unimpaired movement mirrored on the impaired hand/limb can facilitate re-establishment of motor control by retraining basic movement patterns in a slow and precise manner. The patient visualises the feeling and image of the dystonic side carrying out the specific task of writing or playing a musical instrument <i>Equipment:</i> A mirror, musical or writing instrument	Patient advised to complete 10 min twice a day
Slow-down exercise treatment (SDET) ¹⁵	Retraining movement patterns	The task causing the dystonia is repeated at a speed where dystonic movements are not elicited with the hope that the dystonic movement pattern can eventually be “overwritten” The MD subjects play a piece very slowly and record the speed (using a metronome) in beats per minute. After two weeks, the speed is increased by 10% and if there are no increases in dystonic symptoms then incremental speed increases occur every fortnight. The focus is on precision and accuracy of movement patterns, not on playing speed. For the WD subjects, the subjects undertook slow writing retraining, using a loose relaxed tripod grip, large free movements coming from the shoulder and elbow and a variety of writing implements and surfaces. Slow writing in an exercise book or large sheets of paper that are on a high, low or sloped work surface are used to write letters such as Aa, Bb, Cc and shapes such as circles, loops and ellipse <i>Equipment:</i> MD: musical instrument and metronome; WD: variety of writing implements (e.g. pencil, pen, marker, paint brush) and writing surfaces (e.g. paper, white board, sloped writing board)	30 min/day
Soft tissue massage (STM)	To decrease myofascial pain (tender, taut bands and trigger points) ¹⁶	Patients affected by TSD may have myofascial pain due to the severity of or trying to compensate for the involuntary movements. STM administered by hand therapist in treatment sessions	Ten minutes of STM administered per treatment session
Ultrasound therapy (UST)	To decrease levels of myofascial pain and trigger points	UST used in combination with STM has been shown to have an immediate effect when treating latent myofascial trigger points. ^{17,18} Administered by hand therapist in treatment sessions	Ten minutes of UST administered per treatment session

(continued)

Table 2. Continued

Treatment approach	Aim	Description and equipment	When and how much
Forearm muscles stretches	To decrease myofascial tension and pain	Forearm flexor and extensor muscle stretches ¹⁶ one repetition of each stretch five times a day with a 30-s hold in each position at the end of range (but not going into hypermobile range). Exercises were taught within the therapy sessions and incorporated into the HEP	One forearm flexor and one forearm extensor muscle stretch five times a day with a 30-s hold at the end of range (taking care not to go into hypermobile ranges)
Shoulder exercises	Increase freedom of movement in the shoulder girdle	Tension in the shoulder girdle is often a secondary symptom of TSD. Compensating movement patterns can be employed in an attempt to stabilise the affected body part. Scapulo-humeral rhythm exercises ¹⁹ require the subject to lean over and in a relaxed manner, using inertia, to carry the arm forward and backwards, side-to-side and in a circular motion. These exercises were taught within the therapy sessions and incorporated into the HEP	Five minutes twice a day of the paired shoulder exercise positions (forward and backwards, side-to-side and circular motions)
Hand strengthening exercises (putty)	To increase proprioceptive awareness and strength of intrinsic hand muscles	Whilst TSD is not due to muscular weakness the affected hand can show signs of weakness or altered proprioception due to the involuntary motions, altered functional hand movements and decreased ability to perform specific tasks. The functional value of strengthening exercises in muscles that are affected by a myopathic process has been supported ²⁰ These exercises were taught within the therapy sessions and incorporated into the HEP	5 min twice a day

(5) Clinical Global Impression Scale (CGI).^{32–34} This rates change from baseline impairment on a seven-point scale: very much improved, much improved, minimally improved, no change, minimally worse, much worse, very much worse. This was rated at 3 and 6 months.

At baseline and 6 months a standardised video recording was undertaken to capture a clinician-rated assessment of change. Musicians were asked to play a piece that exacerbated their dystonia for 2 min and a mezzo forte legato scale of two octaves 15 times at a speed of eight notes per second (metronome paced). For the free playing the tempo was defined at the baseline assessment and the piece was repeated at the same tempo at 6 months. Participants with writing dystonia were video-recorded writing a standard sentence 10 times, writing five lines of interconnected 'I' with 10 letters per line. Two neurologists (MJE, LR) blinded to the treatment session rated videos according to the following scale: no impairment (1), mild impairment (2), moderate impairment (3), severe impairment (4). Scores were generated for both the predetermined

sequence (interconnected 'I' or scale) and the free task production (writing sentence or piece).

Qualitative interviews

All participants were invited to undertake a brief semi-structured interview to ask them about their experience of engaging in the hand therapy intervention and home exercise programme. These brief, audio-recorded interviews were undertaken by the primary researcher (KB). They were transcribed verbatim, coded and thematically analysed.

Data analysis

This was a feasibility study, and hence inferential statistical analysis and hypothesis testing of the outcome measures is not appropriate and thus was not undertaken.³⁵ Both the patient characteristics and change in outcomes were investigated using descriptive statistics and the calculation of effect sizes (between baseline and 3 months; and baseline and 6 months) according to Cohen's *d*.³⁶ An effect size (ES) of 0.8 is considered large, an effect size of 0.5 is considered medium and

effect size of 0.2 is small.³⁷ Statistical analysis was run using SPSS (version 24).

Results

Over the 6-month recruitment period 15 patients met the inclusion criteria and were recruited, eight with MD and seven with WD.

Feasibility outcomes

The mean recruitment rate was 2.5 per month. In terms of retention, 12 (80%) of the recruited subjects completed the study. Table 3 provides a summary of the sample's demographic and diagnostic characteristics. Two patients dropped out of the study due to a lack of motivation (one before the commencement of the therapy and another after two therapy sessions as they stated that they did not want to complete the home exercise programme). A third patient stopped the study after 10 weeks due to developing a shoulder tendinopathy on the non-dystonic side, this was not related to the intervention or participation in the trial. For those completing the study, there was a 97% attendance rate at therapy sessions. Of the 72 therapy sessions offered (12 subjects with the possibility of six sessions each), only two sessions were not attended due to significant personal events (family funeral, patient in labour). Nine of the 12 participants completed the home exercise programme at $\geq 75\%$ (all writing dystonia patients, four (57%) with MD). Two of the 12 (16.67%) reported finding the home exercise programme too arduous and too intense to complete

(one from each patient group). No adverse events were reported.

Clinical outcome measures

Table 4 summarises the outcome measure results, at each time point, for the 12 participants who completed the study. The effect sizes demonstrated that there were small improvements from baseline in the patient-reported ADDS at the two assessment points. There were small-to-medium patient-reported improvements in the Tubiana Champagne Questionnaire, the BIPQ and perceived health status (EQ-5D 5L score); with scores on each of these measures continuing to improve over the timeline of the study. The clinician-rated video analysis showed moderate improvements over time with sensory-motor rehabilitation therapy.

Qualitative interviews

All 12 participants who completed the study engaged in the interviews. The techniques reported by participants to be effective, neutral or not effective are shown in Figure 2. Patients affected by MD found the following treatments effective: sensory motor retuning (86%), slow-down exercise therapy (71%) and mirror therapy, shoulder range of motion and hand-strengthening exercises (43% each). Patients affected by WD found the following treatments effective: hand-strengthening exercises (80%) and sensory-motor retuning, shoulder range of motion and slow-down exercise therapy (60% each). None of the participants reported either soft tissue massage or ultrasound therapy to be effective.

Table 3. Clinical characteristics for recruited participants ($n = 12$).

	Musician's dystonia ($n = 7$)	Writer's dystonia ($n = 5$)
Gender	4 Male/3 female	0 Male/5 female
Age: mean years (sd)	45 (13.9)	57.2 (3.5)
Age of dystonia onset: mean years (sd)	41.6 (11.9)	43.4 (8.0)
Time from symptom onset to diagnosis: mean years (sd)	2.9 (1.9)	5.0 (0.6)
Past use of botulinum toxin: n (%)	0 (0%)	5 (100%)
Instrument played:		
Piano	4 (58%)	N/A
Guitar	2 (28%)	
Flute	1 (14%)	
Style of music played:		
Classical	6 (76%)	N/A
Rock	1 (14%)	
Affected body part: n (%)		
Small finger	1 (14%)	2 (40%)
Middle finger	1 (14%)	1 (20%)
Small and ring fingers	4 (58%)	1 (20%)
Wrist and middle finger	1 (14%)	1 (20%)

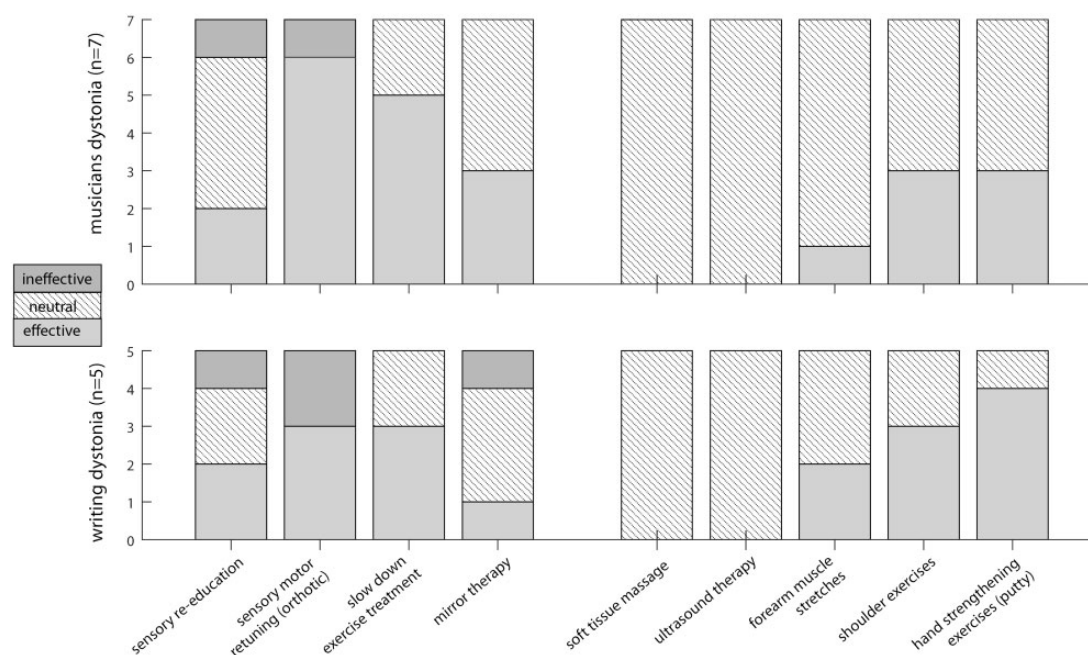
N/A: not applicable.

Data pertains to all participants who completed the study.

Table 4. Changes in clinical outcomes at baseline, 3 and 6 months ($n = 12$).

Scale (available scale range)	N	Baseline Mean (sd) Median (IQR)	3 Months Mean (sd) Median (IQR)	Effect size (Baseline– 3 months)	6 Months Mean (sd) Median (IQR)	Effect size (Baseline – 6 months)
Arm Dystonia Disability Scale (0–100%, higher score = less disability)	12	66.07 (13.85) 72.86 (52.50, 77.14)	70.0 (12.57) 77.14 (62.14, 77.14)	0.28	69.25 (11.65) 73.0 (57.0, 80.0)	0.23
Tubiana-Chamagne Scale ^a (0–5, higher score = less disability)	7	2.86 (1.07) 3.0 (2.0, 4.0)	3.0 (1.15) 3.0 (2.0, 4.0)	0.13	3.43 (0.98) 3.0 (3.0, 4.0)	0.53
Brief Illness Perception Questionnaire (0–80, higher score = more threatening view of the illness)	12	50.67 (8.97) 49.50 (41.75, 55.75)	47.25 (10.23) 45.0 (39.25, 55.50)	0.38	44.33 (10.39) 46.50 (38.50, 51.25)	0.71
Health State (EQ-5D 5L) (0–1, 1 = complete health)	12	0.79 (0.15) 0.79 (0.70, 0.90)	0.84 (0.13) 0.86 (0.85, 0.92)	0.34	0.88 (0.9) 0.91 (0.84, 0.94)	0.59
Video score (1–4, higher score = greater impairment)	12	2.5 (0.78) 2.75 (2.75, 3.19)	NA	NA	1.98 (0.81) 1.88 (1.06, 2.75)	0.78
Clinical Global Improvement Scale (0–7, where 0 = very much improved and 7 = very much worse)	12	NA	3.08 (0.51) 3.0 (3.0, 3.0)	NA	2.58 (0.99) 2.5 (2.0, 3.0)	0.97

NA = Not assessed.

^aScale only completed by musicians.**Figure 2.** Effectiveness of specific therapy techniques (participant reported).

Three main themes were identified: (i) The impact of dystonia, (ii) individuals respond differently to different treatment techniques and (iii) changes require persistence and can take time (Table 5).

Theme 1: the impact of dystonia. All the participants described the negative impact of dystonia on their lives, which resonates with the findings of other qualitative work.³⁸ In particular, the participants described

the specific physical impairments they experienced which impacted upon their ability to carry out tasks and professional activities. This marked impact on people's emotional well-being was striking but is not surprising given that these symptoms typically reach their peak at the high point of the musicians' professional career and can render them unable to play and indeed at times never able to return to their profession. This impact on emotional well-being is in line with

Table 5. Overview of qualitative results.

Themes	Sub themes	Indicative quotes
The impact of dystonia	Work life	<i>"I cannot perform any more or demonstrate in lessons. Playing is hard work, it knocks me out, and I do not loosen up as the time progresses – it becomes painful."</i> (012, male, MD) <i>"My second and ring fingers can claw into my palm and my brain freezes for two minutes or so and I cannot write and I have to consciously think through how to write and this feels very bizarre."</i> (003, female, WD)
	Emotional well-being	<i>"I feel I have a hole inside me. I am completely gutted that I can't play the piano like I used to."</i> (001, male, MD)
Individuals respond differently to specific treatment techniques	Slow-down exercise therapy (writing retraining)	<i>"Using white board markers and chunky pens really helps when writing as does the Coban tape on my thumb, index finger and middle finger; all together it helps a lot."</i> (008, female, WD)
	Mirror therapy	<i>"When using the mirror, I realise how small the movements with the affected finger need to be and so I am practicing contrary motion looking at the unaffected hand and visualising the movements in the affected side."</i> (001, male, MD)
	Sensory re-education	<i>"Sensory re-education is boring and mirror therapy/or playing with my guitar strung up the other way is frustrating as it highlights the problems that the left hand has as the right hand can move so freely and easily."</i> (012, male, MD)
Changes require persistence and take time	Small changes	<i>"No big improvements but I am doing exercises every day and I think there is a slight improvement overall."</i> (011, female, MD)
	Larger changes	<i>"I've made big improvements. I have already been able to play certain passages that I have not played for 4-5 years with more ease . . . I played freely and felt almost entirely normal."</i> (003, male, MD)

the findings of other studies, which demonstrate a tendency towards perfectionism and anxiety in musicians developing focal dystonia.³⁹ Interestingly, subjects with WD often see the condition as "inconvenient" rather than career-threatening, likely because difficulties with writing can be compensated for more easily in employment compared to difficulties with playing a musical instrument for a professional musician.

Theme 2: Individuals respond differently to different techniques.

People's perception of their response to specific treatment techniques varied. This variation in response differed over the timeline of the study. For example, some participants initially found the exercises frustrating and with little improvement in their function, but reported noticing marked improvements over time.

Several participants commented that these rehabilitative treatment approaches were preferable to medical interventions such as Botulinum toxin: *"I had three sessions of Botox but I found it made my hand weak and not functional, I couldn't do buttons up or hold a key or write at all and therefore I have not had any more. I'm now using a Bic biro with a lot of Coban tape to build it up and it really helps the feeling of freedom and ease of writing – my hand still sometimes gets tense and I need to shake it out but my forearm does not get tense now."* (010, female, WD)

Theme 3: Changes require persistence and take time. Many of the participants reported that the changes in symptoms and function took time and required patience and persistence, and could sometimes be demoralising. For some the changes were small, whilst others noted larger improvements.

TSD is a complex condition that affects individuals in a variety of ways: emotionally, physically and in specific task performance such as writing or playing a musical instrument. Use of a variety of individual techniques in combination seems to assist most people, but this requires time and commitment from patients to actively engage in the rehabilitation process.

Discussion

Here we have reported data from a feasibility study of a specific sensory-motor rehabilitation therapy programme for people with TSD affecting writing and musical performance. This intervention was feasible to deliver with high retention, adherence and acceptability to patients. Improvements occurred across measures and were typically larger at the 6-month compared to 3-month assessment, which lends support to an on-going therapeutic effect of hand therapy over time.

All patients who met the inclusion criteria and were invited to take part in the study agreed to do so. Perhaps the high acceptance rate was partially due to

the hand therapy being funded by a grant and thus not incurring private patient fees. There are a range of published treatment approaches typically delivered alone in a high-intensity experimental setting that are arguably difficult to translate into routine clinical practice. The sensory-motor rehabilitation therapy programme we employed in this study is one we have used in routine clinical practice for many years, and is specifically designed to incorporate a range of previously described hand therapy techniques using a small number of face-to-face treatment sessions and a home exercise programme. The results of this feasibility study demonstrated that this intervention is acceptable to most participants. Adherence to attendance at the face-to-face therapy sessions and to the home exercise programme was above our 75% threshold in 75% of the participants. There were no adverse events reported related to the intervention.

One difficulty with designing treatment studies in patients with TSD is the lack of specific measures to assess symptom severity.¹ In this study we used a range of patient-reported and clinician-rated objective outcome measures. Of these, the most promising outcome measures in this study with respect to effect size were the patient-reported CGI and the clinician-rated video. At the end of the treatment, 50% of participants (2 WD, 4 MD) reported a good outcome (self-rating of much improved or very much improved), with a large ES on the CGI. A slight tailing off from perceived benefit was noted at 6 months in comparison to 3 months on the CGI. This suggests that either the patient reported CGI or the clinician-rated video of performance may be useful primary outcome measures in this patient group, with the BIPQ and health state as secondary outcome measures. These results demonstrate that in a future powered trial (alpha 0.05, 80% power), a sample size of 36 would be required if the primary outcome was the patient-reported CGI (6 month mean 3.08, 3 month mean 2.58, SD 0.51); 54 for the clinician-rated video score (6 month mean 1.89, baseline mean 2.50, SD 0.78); or 66 for the patient reported BIPQ.

When interviewed, all patients reported that dystonia had a negative impact on their lives emotionally and in work and functional activities. The treatments that individuals found useful were dependent on the person with many preferring therapeutic approaches rather than medical interventions. MD patients reported that sensory-motor retuning and slow-down exercise therapy were the two most effective treatments followed equally by mirror therapy, shoulder active range of motion exercises and hand-strengthening exercises. Hand-strengthening exercises were reported as being the most effective treatment in the WD group with sensory motor retuning, shoulder range of

motion exercises and slow-down exercise therapy all equally being reported as effective. Time and persistence was required to note changes with some only having small and others having larger improvements.

While this feasibility study was not designed to assess the efficacy of this intervention, these results do provide supportive evidence of a positive treatment outcome in some patients across a range of outcomes. Uniquely compared to previous studies in this area we adopted a mixed rehabilitative approach, combining several specific and general therapies based on individual assessment and response. This approach is supported both by the patient-reported Likert scales evaluating effectiveness of specific therapy techniques and the qualitative interview data; participants typically found more than one therapeutic approach to be beneficial. We believe that it is reasonable to propose that in a future clinical trial this mixed rehabilitative approach be used, rather than selecting a single approach for all patients.

Study limitations

Ideally in establishing feasibility of a future RCT, we would have included a control condition, to determine the willingness of patients to be randomised, and likely retention rates for this group. Also, our sample size was small and we relied on self-report to assess compliance with the home exercise program. The hand therapy intervention was implemented by a single therapist and whilst this meant that the treatments were standardised this could be viewed as a limitation due to possible bias and lack of generalisability. As the hand therapy intervention was funded by a grant, the acceptance rate may have been artificially high. This may have also had an impact on participants being more motivated to attend the therapy sessions and complete the home exercise programme and log. The follow-up was limited to 6 months; ideally, longer-term follow-up of these patients would be of interest, as the treatment timeframe for this condition is considerable and often management of the symptoms is the focus rather than cure. Finally, in addition to asking people about their experience of engaging in the hand therapy intervention and home exercise programme, it would have also been helpful to seek their views, and the views of the clinical researchers, about the study processes and design features (such as acceptability of the recruitment procedures and outcome measures) to inform the design of a future RCT.⁴⁰

Conclusions

This intervention was feasible to deliver with high retention, adherence and acceptability. Improvements

occurred across measures and were typically larger at the 6-month compared to the 3-month assessment, which lends support to an ongoing therapeutic effect of hand therapy over time. The findings provide data to help support the design and development of a future controlled trial for rehabilitation in TSD.

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Declaration of Conflicting Interests

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Ethical approval

Ethics approval was gained from University College London, NHS Foundation Trust, The National Hospital for Neurology and Neurosurgery (Reference 11/LO/0307). This study has therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent

All persons gave their informed written consent prior to inclusion in the study. Details that might disclose the identity of the subjects in the study have been omitted.

Guarantor

KB.

Contributorship

KB and MJE researched literature and conceived the study. KB, AS and MJE were involved in protocol development, gaining ethical approval and patient recruitment. KB, AS, A-M M, IP were involved in data collection. KB was involved in implementing the intervention. KB, JF and AS were involved in data analysis. KB wrote the first draft of the manuscript and MJE, AS, JF and JM amended this draft. All authors reviewed and edited the manuscript and approved the final version of the manuscript.

ORCID iD

Katherine Butler  <http://orcid.org/0000-0003-3386-8730>

References

- Peterson DA, Berque P, Jabusch H-C, et al. Rating scales for musicians' dystonia. *Neurology* 2013; 81: 589–598.
- Sadnicka A, Kassavetis P, Parees I, et al. Task-specific dystonia: pathophysiology and management. *J Neurol Neurosurg Psychiatry* 2016; 87: 968–974.
- Karp BI, Cole RA, Cohen LG, et al. Long-term botulinum toxin treatment of focal hand dystonia. *Neurology* 1994; 44: 70–76.
- Byl N, Merzenich MM and Jenkins WM. A primate genesis model of focal dystonia and repetitive strain injury: I. Learning induced dedifferentiation of the representation of the hand in the primary somatosensory cortex in adult monkeys. *Neurology* 1996; 47: 508–520.
- Byl N, Wilson F, Merzenich MM, et al. Sensory dysfunction associated with repetitive strain injuries of tendonitis and focal hand dystonia: a comparative study. *J Orthop Sports Phys Ther* 1996; 23: 234–244.
- Byl NN and McKenzie A. Treatment effectiveness for patients with a history of repetitive hand use and focal hand dystonia: a planned, prospective follow-up study. *J Hand Ther* 2000; 13: 289–299.
- Byl NN, McKenzie A and Nagarajan SS. Differences in somatosensory hand organization in a healthy flutist and a flutist with focal hand dystonia: a case report. *J Hand Ther* 2000; 13: 302–309.
- Byl N and Topp KS. Focal hand dystonia. *Phys Ther Case Rep* 1998; 1: 39–52.
- Taub E, Miller NE, Novack TA, et al. Technique to improve chronic deficit after stroke. *Arch Phys Med Rehabil* 1993; 74: 347–354.
- Taub E, Uswatte G and Pidikiti R. Constraint-induced movement therapy: a new family of techniques with broad application to physical rehabilitation - a clinical review. *J Rehabil Res Dev* 1999; 36: 1–21.
- Candia V, Elbert T, Altenmüller E, et al. Constraint-induced movement therapy for focal hand dystonia in musicians. *Lancet* 1999; 353: 42.
- Candia V, Schafer T, Taub E, et al. Sensory motor retuning: a behavioural treatment for focal hand dystonia of pianists and guitarists. *Arch Phys Med Rehabil* 2002; 83: 1342–1348.
- Candia V, Wienbruch C, Elbert T, et al. Effective behavioural treatment of focal hand dystonia in musicians alters somatosensory cortical organisation. *Proc Natl Acad Sci U S A* 2003. June 24; 100(13): 7942–6.
- Ramachandran VS and Hirstein W. The perception of phantom limbs. *Brain* 1998; 121: 160–163.
- Sakai N. Slow-down exercise for the treatment of focal hand dystonia in pianists. *Med Probl Perform Art* 2006; 21: 25–28.
- Simons DG, Travell JG, Simons LS., *Myofascial pain and dysfunction: The trigger point manual. Volume 1: Upper half of body*. 2nd ed. Baltimore: Lippincott, Williams and Wilkins, 1999, p.11–93
- Aguilera F, Montanez J, Pecos D, et al. Immediate effect of ultrasound treatment and ischaemic compression techniques for the treatment of trapezius latent myofascial trigger points in healthy subjects: a randomised controlled study. *Manipulative Physiol Ther* 2009; 32: 515–520.

18. Saime A, Sebnem D, Deniz E, et al. Comparison the efficacy of phonophoresis and ultrasound therapy in myofascial pain syndrome. *Rheumatol Int* 2011; 31: 1203–1208.
19. Tubiana R. Musician's focal dystonia. In: R Tubiana and PC Amadio (eds) *Medical problems of the instrumental musician*. London: Martin Dunitz, 2000, pp.329–342.
20. Erwin JH, Keller C, Anderson S, et al. Hand and wrist strengthening exercises during rehabilitation of a patient with hereditary distal myopathy. *Arch Phys Med Rehabil* 1991; 72: 701–702.
21. Fahn S. Assessment of the primary dystonias. In: Munsat TL (ed.) *Quantification of neurologic deficit*. Newton, MA: Butterworth-Heinemann, 1989, pp.187–195.
22. Walter U, Buttkus F, Benecke R, et al. Sonographic alteration of lenticular nucleus in focal task-specific dystonia of musicians. *Neurodegenerative Dis* 2012; 9: 99–103.
23. Schuele S, Jabusch H-C, Lederman RJ, et al. Botulinum toxin injections in the treatment of musician's dystonia. *Neurology* 2005; 64: 341–343.
24. Schabrun SM, Stinear CM, Byblow WD, et al. Normalizing motor cortex representations in focal hand dystonia. *Cereb Cortex* 2009; 19: 1968–1977.
25. Tubiana R. Musician's focal dystonia. In: R Tubiana and PC Amadio (eds) *Medical problems of the instrumental musician*. London: Martin Dunitz, 2000, pp.329–342.
26. Tubiana R. Musician's focal dystonia. *Hand Clin* 2003; 19: 308.
27. Priori A, Pesenti A, Cappellari A, et al. Limb immobilization for the treatment of focal occupational dystonia. *Neurology* 2001; 57: 405–409.
28. Ackermann BJ and Adams R. Finger movement discrimination in focal hand dystonia: case study of a cellist. *Med Probl Perform Art* 2005; 20: 77–81.
29. Berque P, Gray H, Harkness C, et al. A combination of constraint-induced therapy and motor control retraining in the treatment of focal hand dystonia in musicians. *Med Probl Perform Art* 2010; 25: 149–161.
30. Broadbent E, Petrie KJ, Main J, et al. The Brief Illness Perception Questionnaire (BIPQ). *J Psychosom Res* 2006; 60: 631–637.
31. Herdman M, Gudex C and Lloyd A. Development and preliminary testing of the new five-level version of the EQ-5D (EQ-5D-5L). *Qual Life Res* 2011; 20: 1727–1736.
32. Guy WECDEU. *Assessment manual for psychopharmacology – clinical global impression (CGI)*. Rockville: U.S. Department of Health, Education, and Welfare, 1976.
33. Carson AJ. The outcome of neurology outpatients with medically unexplained symptoms: a prospective cohort study. *J Neurol Neurosurg Psychiatry* 2003; 74: 897–890.
34. Sharpe M, Walker J, Williams C, et al. Guided self-help for functional (psychogenic) symptoms: a randomized controlled efficacy trial. *Neurology* 2011; 77: 562–572.
35. Eldridge SM, Chan CL, Campbell MJ, et al. CONSORT 2010 statement: extension to randomised pilot and feasibility trials. *BMJ* 2016; 355: i5239.
36. Stangroom, J. Social Science Statistics. <http://www.socscistatistics.com/effectsize> (2018, accessed 9 March 2018).
37. Sedgwick P. Effect sizes. *BMJ* 2012; 7370: 345.
38. Zosso A and Schoeb V. Musicians' social representations of health and illness: a qualitative case study about focal dystonia. *Work* 2012; 41: 53–59.
39. Altenmüller E and Jabusch H-C. Focal dystonia in musicians: phenomenology, etiology, and psychological trigger factors. *J Hand Ther* 2009; 22: 144–155.
40. Eldridge SM, et al. Defining feasibility and pilot studies in preparation for randomised controlled trials: development of a conceptual framework. *PLoS One* 2016; 11: e0150205.