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The development and use of an anatomy-based retraining program (MusAARP) to assess and treat focal hand dystonia in musicians–A pilot study



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# Bronwen Ackermann, PhD, MPH, BAppSc(PT)<sup>a,b,\*</sup>, Eckart Altenmüller, PhD, MD, MMus<sup>a</sup>

<sup>a</sup> Institut für Musikphysiologie und Musiker-Medizin, Hochschule für Musik, Theater und Medien, Hannover, Germany
<sup>b</sup> School of Medical Sciences, Faculty of Medicine and Health, The University of Sydney, NSW, Australia

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# ABSTRACT

*Background:* Movement dysfunctions are commonly reported in musicians, and in extreme cases may result in a persisting loss of motor control. This condition, whereby motor control of the hand during previously highly trained movements on the instrument is lost, is termed focal hand dystonia. It is widely considered to be a consequence of prolonged repetitive daily practice, often in combination with exposure to a range of other risk factors. Current literature recommends retraining as a promising treatment intervention, although only scant scientific evidence exists on which components should be included in a retraining program, and how these may be best administered.

*Methods:* A progressive muscle activation and movement exercise program was devised by one of the authors applying a series of anatomy-based off-instrument movement tasks. This series of fine motor control exercises, was used to both assess and retrain focal hand dystonia in a population of musicians. The standardized approach aimed to provide a systematic method of retraining musically relevant muscular synergies that could later be applied to the instrument, while still allowing individual modifications. Retraining sessions were mostly run online as a consequence of the coronavirus pandemic, although some early sessions were also able to be undertaken face to face. Both qualitative and quantitative measures were used in this case series to evaluate program efficacy, due to the typical heterogeneity of the fo-cal hand dystonia participants. This included: blinded external neurological evaluation of video footage using the Tubiana grading system, written subjective feedback, exercise progressions, and performance outcomes.

*Results:* Pilot testing of 4 patients indicated the utility of the program over approximately a 12- month time period. All subjects improved, 2 of whom have returned to pre-dystonia performance levels. These patients reported the importance of patience and persistence with daily exercise sessions in their recovery.

*Conclusion:* Using off-instrument playing-relevant exercises to enhance fine motor control and muscle activation can be effective in retraining focal hand dystonia, regardless of additional treatments or level of performance. They should be regularly practiced and progressed in order for effects to be best progressed to instrumental applications. Further research may elucidate whether there are optimal outcomes with single or particular combinations of treatment approaches.

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# Introduction

International studies consistently indicate that musicians across their lifespan will suffer from extremely high rates of performancerelated health disorders, affecting up to 89.5% of professional or-

Musician's dystonia (MD), also known as focal dystonia in musicians or musician's cramp, is a task-specific movement disorder which presents as persistent muscular incoordination or loss of

 $<sup>^{\</sup>ast}$  Corresponding author. School of Medical Sciences, Faculty of Medicine and Health, The University of Sydney, NSW, Australia.

E-mail address: Bronwen.Ackermann@sydney.edu.au (B. Ackermann).

chestral musicians worldwide.<sup>1</sup> Overuse injuries, movement disorders such as musician's dystonia, as well as music performance anxiety are all commonly reported conditions, and in many cases these conditions may be associated.<sup>2,3</sup>

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voluntary motor control of extensively trained movements when musicians play their instruments. This commonly affects hand (focal hand dystonia) or orofacial (embouchure dystonia) movements in instrumentalist musicians. Musician's dystonia can be highly disabling and terminates musical careers in about 30% of cases.<sup>3,4</sup> Besides pharmacological interventions with anticholinergic medication or with local injections of Botulinum-toxin, retraining is considered to be a promising treatment for focal hand dystonia.<sup>5-7</sup> However, there is very little evidence to identify a standardized approach to this – for example, how to structure the retraining, which components to include, and the best way to implement such an approach.

Sensorimotor dysfunctions in musician's dystonia may occur during specific complex movements not only in playing-related tasks, but also in activities similar to the instrumental performance. For example, pianist's dystonia with involuntary flexion of a specific finger may also occur during typing on a computerkeyboard. This suggests that there is an opportunity to retraining dysfunctional movement synergies off the instrument.<sup>8,9</sup> Furthermore, clinicians with extensive experience working with patients with focal hand dystonia recommend that a wholistic approach to treatment which is incorporating postural correction and exercises to regain muscle balance prior to on-instrument retraining, is required.<sup>10</sup> In the relatively limited literature on retraining approaches, combinations of elements such as mirror therapy, sensory feedback, constraint-induced therapy, sensory motor retuning, differential learning and slow-down movement have been applied to motor control of the hand to facilitate regaining functional musical hand movements.<sup>6,11-13</sup> However, there are no standardized fine motor control hand retraining programs available that are accessible for health professionals treating this condition, particularly in relation to off-instrument actions that do not require specific musical expertise.

Aim: to address the call to develop a clinical guideline to retrain hand movements in patients with focal hand dystonia<sup>5</sup> by developing and testing a standardized and progressive off-instrument movement retraining protocol for the treatment of focal hand dystonia in musicians.

# Method

# Formative evaluation stage in protocol development

To design the protocol, existing literature was reviewed relating to retraining methods, applied anatomy and fundamental kinetic chain and biomechanical principles. During music performance, kinetic chain actions involve movement patterns which require multijoint coordination, with larger proximal muscular groups supporting distal actions. For example, the drumming movements of professional drummers are based on a kinetic chain requiring scapula and trunk stabilization, upper-arm and forearm coordination resulting in a whiplash like acceleration of the wrist.<sup>14</sup> With no existing off-instrument retraining methods found, applied kinetic chain principles were combined with clinical experience of experts treating focal hand dystonia to develop a novel retraining protocol over a period of several months. This utilized theory and carefully considered practical implementation as well as available support systems to produce a realistic and flexible clinical approach.<sup>15</sup> The protocol was designed to progress from fundamental action components, usually starting with individual intrinsic hand muscle actions, then following an 'add-on' approach to developing progressively more complex synergistic actions, ultimately integrating a kinetic chain approach to movement incorporating proprioceptive inputs to support the return to more functional arm and hand actions on the instrument<sup>16,17</sup>. All participants received postural

correction and scapula stabilization instructions, as well as elbow and forearm mobilizing warm-up exercises in addition to the fine motor hand retraining protocol with the goal to not only retrain the hand, but to integrate the arm and body as a whole.<sup>10</sup> The MusAARP protocol for the distal upper limb was essentially divided into planes of motion that were based on single intrinsic movements progressing into musically applied anatomy synergies that can be easily modified to suit different instrument playing actions (see Appendix 1).

Existing literature recommends that a multifaceted approach will best meet patients' needs due to the heterogeneity of these patients and their work demands.<sup>18</sup> Accordingly, some participants were offered complementary interventions, in these cases low dose medications, at the discretion of the movement disorders specialist and neurologist.

Ethics were approved by the ethics committee of the Hannover Medical School (MHH E2020. 7324). Data confidentiality was granted and procedures were performed according to the general rules of the German Research Foundation for Good Scientific Practice and the declaration of Helsinki.

#### Process evaluation stage -pilot study case series

To develop further refinements and implementation methods to the protocol, a pilot case series was conducted.

#### Participant inclusion criteria

Following medical screening by an expert neurologist, potential participants who were diagnosed with focal hand dystonia and were active musicians and thus met the inclusion criteria were offered the opportunity to participate in the study using the musically applied anatomy retraining program (MusAARP). Exclusion criteria were a) additional neuropsychiatric disorder, b) age younger than 20 or older than 65 c) inability to follow the training due to time constrictions.

#### Medical Screening Protocol

The medical screening protocol included a comprehensive subjective examination, gathering relevant current, past and family history of hand disorders, co-morbidities and any potential underlying psychological factors. The objective examination included an extensive baseline musculoskeletal and neurological examination, with key tests summarized in Appendix 2.

#### Video evaluation

A video was taken of their playing on their instrument as able as a baseline reference of their dystonia, and subsequent videos at approximately the 6 and 12 month follow-ups. This method has been found effective in previous dystonia retraining trials.<sup>6</sup>

#### Tubiana rating scale

The Tubiana rating scale<sup>19</sup> is a music-specific dystonia evaluation Likert scale, scoring from 0 through to 5.

Score definitions were:

Score 0: Unable to play.

- Score 1: Plays several notes but stops because of blockage or lack of facility.
- Score 2: Plays short sequences without rapidity and with unsteady fingering.
- Score 3: Plays easy pieces, without rapidity and with unsteady fingering.

| Table | 1 |
|-------|---|
|-------|---|

Changes in Tubiana rating scale pre and 12 months post retraining program

| Neurologist | Participant<br>number<br>pre/post | Stage 0<br>"Unable to<br>Play" | Stage 1"Can play<br>several notes but has<br>to stop because of<br>blockage or lack of<br>faculty" | Stage 2"Can play<br>short sequences<br>without rapidity<br>and with unsteady<br>fingering" | Stage 3"Can play<br>easy pieces but<br>unable to perform<br>more challenging<br>pieces" | Stage 4"Can play<br>almost normally but<br>difficult passages<br>avoided for fear of<br>motor problems | Stage 5"Can<br>play normally" |
|-------------|-----------------------------------|--------------------------------|--|--|---|--|-------------------------------|
| N1          | P1 pre                            |                                |  |  | $\checkmark$  |  |                               |
|             | P1 Post                           |                                |  |  |   | $\checkmark$   |                               |
|             | P2 pre                            |                                |  | $\checkmark$   |   |  |                               |
|             | P2 post                           |                                |  |  |   | $\checkmark$   |                               |
|             | P3 Pre                            |                                |  | $\checkmark$   |   |  |                               |
|             | P3 Post                           |                                |  |  | $\checkmark$  |  |                               |
|             | P4 pre                            |                                |  |  | $\checkmark$  |  |                               |
|             | P4 Post                           |                                |  |  |   | $\checkmark$   |                               |
| N2          | P1 pre                            |                                |  |  | Х   |  |                               |
|             | P1 Post                           |                                |  |  |   |  | Х                             |
|             | P2 pre                            |                                |  |  | Х   |  |                               |
|             | P2 post                           |                                |  |  |   |  | Х                             |
|             | P3 Pre                            |                                |  |  | Х   |  |                               |
|             | P3 Post                           |                                |  |  |   | Х  |                               |
|             | P4 pre                            |                                |  | Х  |   |  |                               |
|             | P4 Post                           |                                |  |  |   |  | Х                             |

Score 4: Plays almost normally: difficult passages are avoided for fear of motor problems.

Score 5: returns to concert performances.

Score 3; was considered the stage to commence on-instrument exercises.

For the purpose of the pilot trial, four participants were included who represented a spectrum of the typical types of patients seen with FHD. These patients had attended the clinic of the second author for diagnosis of their condition, and once diagnosed with focal hand dystonia and meeting the inclusion criteria, were offered a chance to participate in the retraining trial

#### Participant feedback

Participants were asked to write a narrative of their experiences with the retraining program. Additionally, they wrote short feedback on each repeat visits, along with verbal feedback to the retraining therapist.

#### Evaluation of the Retraining effects

Blinded video analysis was conducted on video footage from approximately 12 months apart, by two different expert neurologists using the Tubiana scale rating (see Table 1).

#### MusAARP implementation and progressions

All sessions of assessment and retraining were conducted by the primary author. Musician participants were asked to demonstrate tasks from the off-instrument protocol and were asked to do a series of daily exercises from the program that effectively challenged them, but that they could effectively work on to regain control of the dystonic and compensatory finger actions. Each musician is able to be individually assigned to a level based on their ability to perform the particular tasks in the protocol, that are progressed depending on the ability of the musicians to develop a high level of competency of this task (ie, rapid speed, eyes closed, done in instrumental playing positions). The exercises chosen are thus able to be individualized to a musicians' major presenting symptoms, while still following a standardized format. These exercises were progressed on return visits, eventually into ranges more closely representing playing positions. Additionally, posture correction and an upper limb warm up was included for all patients. It had been the intention to run sessions fortnightly for the first 3 months and then monthly, however, due to the coronavirus pandemic, sessions were much less frequent and follow-up appointments mostly ceased for 4 months in the middle of the trial (participants continued exercises on their own) and then ultimately became online only. This certainly was felt to not be ideal by the researchers, but nonetheless participants were satisfied to be able to continue with supervised retraining.

#### Pilot trial participant characteristics and outcomes

#### Participant 1 (P1)

A left-handed healthy 30-year-old male professional concert pianist presented with worsening right hand movement problems for about 18 months, triggered by a very high workload preparing a series of concert programs. The main presenting findings were involuntary flexion of the right third (middle) and fourth (ring) finger with compensatory hyperextension of the right fifth (little) finger during piano performance, with some symptoms also reported at the computer keyboard. On initial consultation, the anticholinergic medication trihexyphenidyl (up to 4mg/day) was prescribed and the patient referred to the retraining program. Sessions were completed after 3 face to face and two online retraining sessions over 6-months. He reported an improvement of 75% with the combination of medication and retraining exercises, and has returned to recording concert repertoire, and did not seek further treatment.

#### Participant 2 (P2)

A 27-year-old right-handed female flute student completing a Masters in flute performance, with no prior or family history of problems, reported a sudden appearance of left finger movement problems for 6 months triggered by a sudden increase in playing in preparation for final performance exams. She presented with task-specific involuntary flexion of the fourth and fifth fingers. No medications or other treatments were prescribed, other than attending the retraining program. Sessions were scheduled fortnightly face to face for 3 months, then 5 monthly online sessions. After 8 months she returned to flute playing and recital preparation.

# Participant 3(P3)

Twenty-nine-year-old female professional orchestral violinist, right-handed, no family history of problems, with a past history of 10 years focal tremor in the left middle finger, and 'tendonitis' between fourth and fifth rays of the left hand 3 years, treated mostly by rest. She reported ongoing sensory disturbances there, such as increased cold sensitivity. For 2 and half years she reported an increasing involuntary flexion of the fourth and fifth fingers of the left hand, following a period of an increased workload on the violin. On initial assessment, she was prescribed trihexyphenidyl up to 4 mg/day and was offered to undergo the retraining program. About 6 months into retraining, the participant was noted to be breath holding so meditation and focus on breath preplaying was recommended. During the retraining pause, she tried applying principles from books, and felt she regressed until retraining resumed. Retraining sessions were scheduled fortnightly for 6 months, a 4-month pause, then online for 6months.

Sessions: 16 retraining sessions to date.

#### Participant 4 (P4)

Avid amateur Bagpiper, right-handed, healthy, Carpal Tunnel Surgery left hand in 2019, no other physical trauma, no dystonia in the family. Since about 2016, slowly increasing dysfunction of the left index finger, also stiffness in the left middle and ring finger. Diagnosed with dystonia November 26th 2019, after 13 months he feels his playing is returning to good and is able to have bagpipe lessons with advanced players to continue his desired playing program.

### Results

The protocol and sequence of individual muscle activation to synergistic tasks was successfully applied by the lead researcher. All exercises were able to be progressed and some were refined – mostly needing additional intermediate simpler synergies added – as participant skill increased. Exercises could be easily applied into ranges of motion and into relevant movement synergies at the instrument as a method to initially transfer synergies into playing actions (at Stage 3 onwards of the Tubiana scale). The slowest progress was noted in the musician who took the least time off her orchestral playing, which may have acted to slow down the integration of new habits into playing technique.

Independent video analysis by the 2 experts yielded overall improvements over 12 months (Table 1).

Narrative thematic analysis of written participant feedback.

#### Off-instrument approach to exercises

All patients found the off-instrument exercises helpful as a component of their treatment, especially in the early stages of retraining while dystonic and compensatory movements on the instrument were more severe. For example, P3 stated that ": "I couldn't feel my hand shape anymore....when I played violin the whole left hand was very uncomfortable..." P4 commented that: "Fingers and sometimes the whole hand slipped off the chanter and I exerted a lot of will power and tried to force my hand to stay in a playable position. My fingers nonetheless did not close holes properly, were cramped and were extremely slow... the little finger and thus the ring finger pulled away."

Doing off-instrument exercises provided access to relearning movements in a less stressful manner. P3 reported of her experiences - "I learned many different things about how to move my finger, arm and body correctly and efficiently....Like to control every finger to move independently from the base knuckle in all directions, to feel the rotation from the forearm,"

For P4 – "I found doing the exercises off the instrument to be essential to regaining control of movements.... Doing the exercises regularly increases suppleness and speed and precision of movement significantly, independent of whether one has focal dystonia or not." P1 commented that "I notice that I benefit a lot from the different perspectives from which the exercises and problems are approached."

#### Mental practice

One player found that he could successfully mentally practice the movement synergies, commenting that "I feel that it is important to be mentally aware of the exercises even when not doing them." (P1)

For another P3, it was observed that she was breath holding while doing the exercises as well as playing the guitar, and she was encouraged to try meditation prior to doing her program and playing. She remarked that "I began to meditate everyday 2 times before I do exercises. After I felt that my arm and hand were very soft, and slowly this softness could last longer."

#### Dosage of exercises

For these participants, it took approximately 12 months from beginning the exercises to regaining substantial playing improvements on the instrument, and this approximate to time frame was made clear to all participants at the outset. It is important to establish the gradual nature of retraining motor synergies using regular daily practice to be able to stay patient and positive. P2 supported this observation stating that "I do the exercises according to the instructions, punctually every day, step by step according to the instructions.... half a year isn't good enough, keep persisting for a year. You can't give up, you have to have a good attitude"

# Applications to playing

Once the participant had progressed well in their combined tasks, they could then apply these movements to the instrument in relevant playing contexts. P2 found that "I was aware of doing the training every day on time, so that I can consciously master the relaxation and coordination of the whole-body muscles and apply it to playing the flute" Applying these synergies into playing action can be challenging as often dysfunctional synergies had become the norm, and the musicians needed to be open to exploring new movement possibilities. P1 stated "For me it was important to realize that it takes some time and that in order to change you must 'abandon' the old." For P4, after 13 months he noticed "my playing is now consistently good, and sometimes already better than just good. Body posture, hand and finger position are all going well now, most importantly I now really move my fingers from the elbow/forearm which makes all the difference!"

#### Whole body approach to retraining

It was noted in all musicians that there was a disconnect between the posture and more proximal arm actions and the hand. The importance of a balanced postural base and good scapula control was emphasized. For some players, more general forms of bodywork and health activities were also felt to be beneficial in conjunction with the exercise program. For example, P1 wrote that "I also feel it is important to the body/brain to include exercises such as Qigong, sport and general exercises for well-being." P3 commented that "I am more relaxed than I used to be, feel more comfortable when I practice, and my posture is healthier."

# Discussion

The MusAARP designed to retrain movements and muscle synergies off the instrument was effective in improving the symptoms of all four participants, regardless of baseline status and whether other modalities, such as medications, were utilized. The formative evaluation process was time-consuming, but important in designing a carefully considered protocol, which is likely to have helped achieve good results in this small sample. The participants reported the importance of regular exercise participation and progressed effectively through the MusAARP series of exercise tasks, despite the added complexity of conducting at least half of the retraining sessions online.

Themes emerging from patient feedback highlighted the gradually returning sense of movement control and awareness of movements at the hand being linked to the rest of the arm and body. The importance of regaining postural balance and an integrated flow of action from a stable scapula through a mobile elbow joint complex to dextrous hand function is consistent with the proximal to distal biomechanical models often referred to as the kinetic chain in sports.<sup>16</sup> Establishing good function in affected segments and then re-integrating these components together into a performance-specific flow of action appeared to be an important component in this study, supporting previous recommendations by clinicians working in this field.<sup>10</sup> Three of the participants emphasized the importance of regaining a better feel of movements, consistent with recommendations to integrate proprioception in rehabilitation.<sup>17</sup>

It was important to progress exercises into relevant ranges of motion and positions in relation to their musical performance to increase motivation and perceived relevance of exercises. Additionally, the exercises could be applied in an early stage to the instrument to facilitate understanding of flow of these synergistic actions onto the instrument. This was the primary component of any oninstrument work – application of the learnt synergies and actions to the instrument. All participants were encouraged to use mental imagery to facilitate integration of new synergies, and to use this as well as relevant personal prompts to move towards an external focus of attention (rather than internal) once better control was achieved.

The retraining protocol achieved its desired goal, with all patients achieving first independent control of fingers and thumb, and a gradual re-integration of these actions into upper limb kinetic chain actions, ultimately applied to the instrument. This effective novel protocol aims to provide a guide for those working on retraining to highlight a systematic anatomically and systematically designed protocol, as a contribution to some other attempts to systematize approaches to retraining.<sup>6,12</sup> One of the great advantages of starting with "off-instrument retraining" is a psychological benefit: musicians suffering from dystonia frequently are very anxious and easily frustrated when attempting to perform movements on the instrument. Here, off-instrument exercises ensure progress, self-efficacy and lay a fertile ground for less frustrating and more motivating exercises with the instrument.

The goal is to now test this protocol on a larger group of subjects to further elucidate best implementation strategies and potentially whether multimodal strategies are different to retraining alone.

# Acknowledgments

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#### Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jht.2021.05.007.

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- # 1. The subject population was \_\_\_\_\_ musicians
  - a. 20
  - b. 14
  - c. 4
  - d. 2
- # 2. The exercise program is centered on
  - a. off-instrument movement tasks
  - b. progressive practice sessions starting with a minimal period of playing
  - c. PREs for the intrinsic muscles
  - d. visualization

- # 3. A favorable outcome would see improved
  - a. self esteem
  - b. grip strength
  - c. sensibility
  - d. fine motor control
- # 4. The Tubiana rating scale
  - a. incorporates 3 elements of the DASH
  - b. was developed in conjunction with the Cleveland Clinic
  - c. uses a 0-5 scoring system
  - d. has an ICC of .98
- # 5. 75% of the subjects returned to pre-dystonia playing levels a. true
  - b. false

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