

2021-04-01

# Task specific dystonia a patients' perspective

Butler, K

<http://hdl.handle.net/10026.1/17572>

---

10.1016/j.jht.2021.04.005

Journal of Hand Therapy

Elsevier BV

---

*All content in PEARL is protected by copyright law. Author manuscripts are made available in accordance with publisher policies. Please cite only the published version using the details provided on the item record or document. In the absence of an open licence (e.g. Creative Commons), permissions for further reuse of content should be sought from the publisher or author.*

# Task specific dystonia – a patients’ perspective

Katherine Butler<sup>1,2,3</sup>, Karin Rosenkranz<sup>4</sup>, Jennifer Freeman<sup>1</sup>

1. Faculty of Health, University of Plymouth, Peninsula Allied Health Centre, Plymouth, PL6 8BH
2. University College London, Gower Street, London, WC1E 6BT
3. London Hand Therapy, King Edwards VII’s Hospital, London, W1G 6AA
4. Ruhr-University Bochum, Department of Psychiatry and Psychotherapie, Lübbecke, Germany

## Declarations

### Competing interests

The authors declare that they have no conflict of interest.

### Ethical standards

This study was approved by the appropriate ethics committee and has therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. All persons gave their informed consent prior to inclusion in the study. Details that might disclose the identity of the participants in the study have been omitted.

### Acknowledgements

British Association of Hand Therapy assisted with part funding for this project.

The participants in this study who are affected by this complex condition (from whom we always learn.)

## Address for correspondence

Katherine Butler, London Hand Therapy

King Edward VII’s Hospital

5-10 Beaumont Street

London W1G 6AA [katherinebutler@londonhandtherapy.co.uk](mailto:katherinebutler@londonhandtherapy.co.uk)

## **Abstract**

### **Study design**

Descriptive survey

### **Introduction**

Task specific dystonia (TSD) is a subtype of dystonia with no cure and significant limitations on treatments. Few studies have investigated the outcomes of rehabilitative therapy from the patient's perspective.

### **Purpose of the study**

This study explored the interventions that patients have utilized and their perceived effectiveness in treating and managing their TSD, specifically musicians' dystonia (MD) and writer's dystonia (WD). Symptoms and the effect of TSD on the perceived performance of every day and specific tasks, and possible reasons why the condition developed, were also investigated.

### **Methods**

Patients diagnosed with TSD, treated at a private hand therapy unit, who had consented to being contacted for research purposes, were emailed a link to an online survey (or posted if email was not available), administered via KwikSurveys. The survey consisted of four subsections: personal information, general medical history, dystonia medical history and dystonia treatment history.

### **Results**

Invitations were sent to 105 patients of whom 90% (n=95/105) responded. Results for both the MD and WD groups were similar. There was a significant association between the two groups as to what they viewed may have led to the development of TSD (a change in technique;  $p<0.001$ ) and the most effective treatments (massage  $p<0.043$ , modifications to the instrument  $p<0.002$ ; ultrasound therapy  $p<0.013$ .) All reported that daily activities were affected by their condition.

### **Discussion**

Although full levels of task specific function (playing their instrument or writing) were not usually regained, participants with both MD and WD perceived improvement in symptoms with rehabilitation intervention. Three treatments were perceived to be most effective in achieving this, suggesting that these should be considered for inclusion within treatment plans.

### **Conclusions**

From the perspective of people with TSD, a range of rehabilitation interventions are effective in enhancing symptom management, providing further evidence to support their use.

## Key words

Musicians' dystonia, writers' dystonia, task specific dystonia, rehabilitation therapies, hand therapy

## Introduction

Task specific dystonia (TSD) is a subtype of dystonia which is characterized by an abnormal pattern of muscle activity often associated with involuntary, sustained, patterned and co-contraction of opposing muscles that can cause twisting-movements or abnormal postures. It is often task specific and frequently only occurs during the performance of skilled tasks such as writing (writers' dystonia, WD) or playing a musical instrument (musicians' dystonia, MD). It is a condition associated with occupations, most commonly affecting professional musicians. Approximately 1% of professional musicians are affected, which is a rate of about 10 times greater than that of non-musicians.<sup>1</sup> It is reported that up to 62% of musicians affected by this condition are unable to continue their performance careers.<sup>2</sup>

At present, there is no cure for dystonia, and many of the treatments available have significant limitations.<sup>3</sup> Current treatments include oral medication, botulinum toxin injections, surgery, rehabilitative therapies, ergonomic changes to the instrument and supportive approaches.<sup>4,5</sup> Existing medical treatments such as oral medications (e.g. trihexyphenidyl) and botulinum toxin injections are limited in their long-term efficiency.<sup>6</sup> Rehabilitative therapies aim to re-shape sensory and motor representations in the brain with the hope of regaining control of the affected movement.

A literature search of PubMed (to February 2021) revealed several studies that outline available rehabilitative therapies for treating patients with TSD.<sup>3</sup> Other studies explore precipitating or contributory factors that may have led to the development of musicians' dystonia.<sup>7,1</sup> No studies were found which explored the effectiveness of treatments from the perspective of WD patients, with four studies investigating this from the perspective of professional musicians. Schuele and Lederman surveyed 21 string players with musicians' dystonia about their treatment attempts, which included nerve decompression, physical therapy, retraining, anticholinergic medication, botulinum toxin injections and orthoses. The authors concluded the benefit of treatment was limited as more than half the patients' careers were ended due to dystonia.<sup>8</sup> Vugt et al retrospectively explored the response to treatment in 54 pianists affected by musicians' dystonia where about 50% objectively (and 80% subjectively) of patients improved with task performance following participation in a variety of intervention strategies (retraining therapy, relaxation techniques and change in teacher).<sup>9</sup> Along similar lines, Jabusch and Altenmüller surveyed 144 instrumentalists affected with musicians' dystonia asking them to subjectively rate cumulative treatment responses to individual therapies. An alleviation of symptoms was reported by 54% with the most effective treatments (in descending

order) being ergonomic changes, unmonitored technical exercises, pedagogical retraining, botulinum toxin injections and trihexiphenidyl.<sup>10</sup> A feasibility study of participants with TSD<sup>11</sup> of a rehabilitation program that included a mix of sensory and motor treatments, demonstrated the intervention was feasible to deliver with high retention, adherence and acceptability; and provided supportive evidence of positive treatment outcomes across a range of measures, at three and six month follow-up.

Overall, these studies show that around 50% of musicians affected by dystonia seem to be helped by a variety of treatment techniques that commonly include retraining therapy, physical therapy, and ergonomic changes.

Considering this limited evidence, the purpose of this survey was to investigate, from the perspective of people with TSD:

1. What they view may have led to the development of TSD
2. Which treatment approaches (medical and rehabilitative) they have utilized
3. Their perceived effectiveness of these treatments
4. The impact of their symptoms on their ability to function in everyday activities
5. The impact of their symptoms on their ability to perform specific activities (writing and playing their musical instrument)
6. Whether there was an association between the two groups regarding the above

## **Patients and methods**

Patients diagnosed with TSD, treated at a private hand therapy unit in London between 2006 and 2016, and who had consented to being contacted for research purposes, were invited to participate in this ethically approved study (University of Plymouth Faculty of Health and Human Sciences Research Ethics Committee, 15/16-560).

### **The survey instrument**

A search of the literature highlighted that no relevant standardized questionnaires were available for use. Consequently, two draft survey questionnaires (musicians' dystonia and writer's dystonia) were developed in line with established methods.<sup>12</sup> The questionnaire content was based on a combination of information from the scientific literature and discussions with multidisciplinary health care team members with a good working knowledge in managing musicians' and writers' dystonia. The draft questionnaires, designed using the online software tool KwikSurvey (kwisurveys.com), were piloted on eight individuals (four with musicians' dystonia and four with writers' dystonia) who covered the range of people intended for inclusion in the survey. Specific feedback was sought in relation to ease

of completion, the wording and potential ambiguity or bias of questions, relevance, and length of the questionnaire, and whether there were any redundant or missing items. Based on this pilot, only minor amendments were required to the wording of some questions. Each questionnaire comprised several sub-sections: personal information (3 items), general medical history (4 items), dystonia medical history (16 items), dystonia treatment history (10 items) and for the MD questionnaire a musical history subsection was also included (13 items). In total there were 48 questions in the MD survey and 25 in the WD survey.

## **Data collection**

An invitation to participate in the survey was sent to the 102 potential participants via email, which included a link to the online questionnaire. In three cases, where the patient did not have an email address, a paper version of the questionnaire was sent by post. Potential participants were requested to complete the questionnaire within 10 days with a reminder email sent at the 10-day mark.

Participation was voluntary and anonymous.

## **Data Analysis**

Data was exported from KwikSurveys to Microsoft Excel 2016 for analysis. Data sets, where participants had completed at least 45% of the survey, were analyzed. Descriptive statistics were used to outline the participant characteristics and the responses from each question (% and frequency counts). Associations between the two groups in terms of their symptoms, perceived treatment effectiveness, events that may have led to dystonia development, and the impact of symptoms on every day functional tasks and specialist tasks (playing musical instrument and writing) were analyzed using the Pearson chi-square test and Fisher's exact test.

## **Results**

### **Participant characteristics**

In total, 105 patients with TSD registered on the database were sent, and were recorded to receive, the survey invitation. Of these 71% (n=75) were diagnosed with MD and 29% WD (n=30). One MD survey was not returned. The response rate was 90% (n=95/105). The returned surveys were completed in entirety by 68% (n=50/74) of MD and 67% (n=20/30) of WD patients. Table 1 details the demographic and diagnostic characteristics for participants who completed at least 45% or more of the survey.

### **Co-morbidities**

Of the MD responders 76% (n=42/55) reported experiencing several co-morbidities, with the five most common being: depression 21% (n=9/42), anxiety disorder 19% (n=8/42), osteoarthritis 14%

(n=6/42), high blood pressure 12% (n=5/42) and repetitive strain injury (RSI) 12% (n=5/42.) For the WD responders, co-morbidities were reported for 67% participants (n=14/21), with the five most common being: osteoarthritis 21% (n=3/14), depression 14% (n=2/14), anxiety disorder 14% (n=2/14), hypermobility 14% (n=2/14) and high blood pressure 14% (n=2/14.)

## **Symptom manifestation**

### *Initial symptoms*

The two most common symptoms reported at diagnosis of TSD were involuntary movements (MD 92%, n=46/50; WD 81%, n=17/21) and lack of co-ordination (MD 82%, n=41/50; WD 86%, n=18/21).

When comparing symptom manifestation at initial diagnosis between the two groups significant associations occurred for pain ( $p<0.005$ ) and weakness ( $p<0.023$ .) Pain was described by 38% (n=19/50) of MD and 71% (n=15/21) of WD participants and weakness was described by 52% (n=26/50) of MD and 81% (n=17/21) of WD participants.

### *Current symptoms (at the time of survey completion)*

The two most common symptoms experienced at the time the participants completed the survey were involuntary movements 94% (n=47/50) and clumsiness 68% (n=34/50) for MD and lack of co-ordination 67% (n=14/21) and involuntary movements 67% (n=14/21) for WD.

When comparing the groups for current symptoms significant associations occurred for involuntary movements ( $p<0.006$ ) and tremor ( $p<0.007$ ). Involuntary movements were described by 94% (n=47/50) MD and 67% (n=14/21) WD and tremor described by 14% (n=7/50) of MD and 48% (n=10/21) of WD patients. Table 2 outlines symptom comparisons between groups at time of diagnosis and when completing the survey “current symptoms.”

### *Affected part of upper limb*

The upper limb area that was most affected for both MD and WD was the finger 96% (n=48/50) for MD and 95.2% (n=20/21) for WD, thumb 22% (n=11/50) for MD and 66.7% (n=14/21) for WD and wrist 10% (n=5/50) for MD and 66.7% (n=14/21) for WD. Significant associations between the two groups occurred for the thumb and wrist ( $p<0.001$ ), but not for other body parts.

### *Affected digits*

For the MD group the digits affected, in descending order of frequency were ring (60% n=30/50), middle (52% n=26/50), index (42% n=21/50), small (36% n=18/50) and thumb (20% n=10/50). In the WD group these were middle and index fingers (each 76.2% n=16/21), ring and small (each 61.9%

n=13/21) and thumb (57.1% n=12/21.) Significant associations between the two groups occurred for the thumb ( $p < 0.002$ ), index ( $p < 0.001$ ) and small ( $p < 0.045$ ) fingers.

## **Medical management**

The medical management of MD and WD was categorized into three broad bands: medication, botulinum toxin injections and surgery. The medical treatments participants have received in the past (before survey completion) are outlined in Table 3.

At the time of survey completion most participants in both groups (71% n=39/55 for MD and 76% n=16/21 for WD) had never used any medication to alleviate symptoms.

Botulinum toxin injections were received by 28% (n=14/50) of MD participants with 36% (n=5/14) saying that they found them effective and 43% (n=6/14) saying they were having them on an ongoing basis. The ideal time frame between injections for 57% (n=8/14) of the participants was 0-3 months with 21% (n=3/14) having received more than 10 injections. Injections were discontinued by 57% (n=8/14) of the MD participants and the reasons given (more than one response was permitted) were: 43% (n=6/14) said they found them ineffective, 21% (n=3/14) said the symptoms increased in severity once the effect of the botulinum toxin wore off and 14% (n=2/14) said they did not like the idea of being reliant on injections.

Of the WD participants 60% (n=12/20) had received botulinum toxin injections with 58% (n=7/12) of these participants saying they found them effective but only 17% (n=2/12) reporting to have them on an ongoing basis. The ideal time frame between injections for 50% (n=6/12) of these participants was 3-6 months with 50% (n=6/12) having received up to three injections. Of the 83% (n=10/12) of WD participants who discontinued the injections (more than one response was permitted) 58% (n=7/12) said they did not like the feeling of weakness or loss of function and 25% (n=3/12) said they found them ineffective.

For both groups other reasons for discontinuing the injections were described by them in free text, and included: they were too painful, did not like the idea of being reliant on injections, condition improved significantly and did not need them anymore, wanted to find the cause of the problem rather than treat the symptoms and that it was too difficult to accurately target the specific muscles affected by the dystonia.

Surgery had been performed on 10% (n=5/50) of MD participants; three of whom had trigger finger release and two a carpal tunnel decompression. Of these five participants, four said it had no effect on their TSD symptoms, and one said that their TSD symptoms were worse. No WD participants reported having had surgery.



## **Musical history (in MD participants)**

The instrument most represented was the guitar 49% (n=27/55) followed by the piano 35% (n=19/55), wind players (flute, saxophone, Irish uilleann pipes) 11% (n=6/55) and string players (violin and cello) 5% (n=3/55).

The musical style that was mainly played before the onset of dystonia was: classical 84% (n=46/55), jazz 29% (n=16/55), rock 20% (n=11/55) and other (folk, indie, world, musicals, blues) 22% (n=12/55.) The musical style that was mainly played after the onset of dystonia, and at the time the survey was completed was: classical 69% (n=38/55), jazz 27% (n=15/55), rock 16% (n=9/55) and other (folk, indie, world, musicals, blues) 25% (n=14/55) with 11% (n=6/55) unable to play at all.

Professional players made up 67% (n=37/55) with amateurs comprising the remaining 33% (n=18/55.) Of the total group, before the onset of dystonia 45% (n=25/55) found themselves in the role of a soloist, 25% (n=14/55) ensemble player, 15% (n=8/55) music teacher and 15% (n=8/55) accompanists. Of the total group at the time of completing the survey 29% (n=16/55) played in the role of a soloist, 20% (n=11/55) ensemble player, 25% (n=14/55) music teacher and 13% (n=7/55) accompanists and 13% (n=7/55) were unable to play at all.

For 64% (n=35/55) the age of starting to play their instrument was  $\leq 14$  years with the average length of playing per day being 0-30 minutes for 36% (n=20/55), 30-60 minutes for 22% (n=12/55), 1-3 hours for 35% (n=19/55) and more than 3 hours for 7% (n=4/55.) Only 40% (n=22/55) played a second instrument with 36% (n=8/22) of these participants playing the piano and 18% (n=4/22) the guitar.

## **Views as to what may have led to the development of TSD**

Participants were given a selection of eight (with the ninth being “other”) potential events that they considered may have led to the development of TSD. Table 4 outlines these for both groups. There were no significant associations for these events between the two groups, apart from “a change in technique” ( $p < 0.001$ ).

Interestingly when the musicians were asked if there was a piece or playing technique that may have led to them developing dystonia 51% (n=28/55) identified that there was. With tremolo (32%, n=9/28), a specific piece (29%, n=8/28), obsessive practice (25%, n=7/28) and poor technique (14%, n=4/28) being reported as more than likely leading to their development of MD.

## **Rehabilitative treatment approaches utilized and perceived effectiveness**

Table 5 details the rehabilitation interventions participants had tried. Regarding these different interventions, there was a significant association between the two groups for altering playing/writing

technique ( $p<0.027$ ) and modifications to the musical or writing instrument/implement ( $p<0.002$ ). Table 6 outlines participants perceived effectiveness of rehabilitation interventions where potential responses were “ineffective”, “neutral” or “effective.” With respect to this, significant associations between the two groups were seen for massage ( $p<0.043$ ), modification to the instrument ( $p<0.002$ ) and the use of ultrasound therapy ( $p<0.013$ ).

### **Perceived impact of symptoms on daily function**

In terms of tasks of daily living, the five most common difficulties for MD participants were: typing 58% ( $n=29/50$ ), thinking about playing a musical instrument 40% ( $n=20/50$ ), writing 34% ( $n=34/50$ ), fastening buttons 30% ( $n=15/50$ ) and drawing 24% ( $n=12/50$ ). For WD these were: drawing 81% ( $n=17/21$ ), typing 62% ( $n=13/21$ ), thinking about writing 52% ( $n=11/21$ ), using cutlery/chopsticks 52% ( $n=11/21$ ), brushing teeth 45% ( $n=9/21$ ), fastening buttons 45% ( $n=9/21$ ) and applying makeup 57% ( $n=8/21$ ). Table 7 outlines the top five tasks of daily living affected by TSD for both groups. Significant associations between the two groups for these activities were seen for brushing teeth ( $p<0.013$ ), drawing ( $p<0.001$ ), thinking about writing ( $p<0.008$ ) and using cutlery/chopsticks ( $p<0.032$ ); with all impacting the daily activity of WD participants more than MD participants

### **Perceived impact of symptoms on ability to perform specific activities (writing and playing their musical instrument)**

Participants were asked to rank on a scale of 0-5 their ability to play their musical instrument on the Tubiana and Chamagne scale<sup>13</sup> where 0 indicates inability to play and 5 indicates return to concert performances. This scale was modified for use in the WD questionnaire, with 0 indicating the inability to write and 5 the ability to write with no limitations.

The vast majority reported that, before the onset of TSD they had no limitations (level 5) with playing/ writing (MD, 84% ( $n=42/50$ ); WD, 95.2% ( $n=20/21$ )). In contrast, their ability to play/write at the time of survey completion was significantly more limited (MD, 38% ( $n=19/50$ ) at level 4; WD, 28.6% ( $n=6/21$ ) at level one,  $p<0.025$ ). Level 4 on the MD scale indicates ‘being able to play almost normally but avoiding long passages due to fear of incoordination, pain, or tiredness in my arm’ and level 1 on the WD scale indicates ‘being able to write several words but having to stop regularly to rest.’

## **Discussion**

We report on findings of a survey where patients affected by MD and WD were asked to consider what may have led to them developing dystonia, consequences on specific and other activities, and

the effectiveness of treatments they have tried. The response rate was excellent, with 90% of participants starting the survey, and approximately two thirds completing it all.

The sample characteristics of this study were mostly in line with epidemiological studies of this condition. MD has a higher relative prevalence within professional musicians with estimates that 1% will be affected within their lifetime<sup>1</sup> in comparison to 0.01% of the general population being affected by WD.<sup>14</sup> As in other publications,<sup>15</sup> our sample was comprised of a higher percentage (84% n=46/55) of men affected by MD. For the WD group our sample comprised more woman (62% n=13/21) which contrasts with the literature that states there is a slight male preponderance.<sup>16</sup>

Our ratio of 9:1 right to left handedness of survey participants reflects that found in the general population where about 90% of humans, independent of cultural background, are right-handed.<sup>17</sup> The survey findings reflect this, with the right hand being where most MD and WD participants experienced dystonic movements in the fingers that are most utilized (ring and middle fingers for MD and index and middle for WD.)

It has been identified that there may be several predisposing factors constituting a vulnerability or predisposition to developing MD including instrument played (musicians who play piano, guitar and woodwind instruments may be at particular risk)<sup>18</sup> and a positive family history.<sup>1</sup> Guitarists and pianists were the two most represented instruments in the MD survey, with 9% (n= 5/55) of MD and no WD participants reporting family members with neurological conditions. Therefore, our results are in line with papers that report only 6% of either writer's or musician's dystonia patients' have dystonia in their families<sup>7</sup> rather than those that have found 20-25%<sup>19</sup> or 36%<sup>20</sup> of patients with TSD have a family member that is also affected.

In line with other studies many of our MD participants started playing their instrument from a young age (before 14 years).<sup>21</sup> Depending on the age at which the instrumental playing was started, structural and functional alterations in the sensorimotor cortex of professional musicians have been described.<sup>22</sup> A vulnerability has been shown in musicians who start playing intensively and at a young age.<sup>23</sup> However, there are mixed opinions in the literature with reports that musicians who start practising after the age of 10 years are at much higher risk of developing dystonia.<sup>24,25</sup>

Both depression and anxiety were within the top five most reported co-morbidities within our survey respondents. A few studies have indicated that there is an association with higher levels of anxiety<sup>26</sup> in patients who develop WD and traits of perfectionism, neuroticism, anxiety and phobias in musicians suffering with MD<sup>27</sup> Reported levels of anxiety and depression in the general adult UK population vary widely from 3.3% depression and 5.9% anxiety<sup>28</sup> to 19.7%<sup>29</sup> so it is difficult to make comparisons with our survey participants. The relationship between anxiety and depression and the development of TSD is an important area of future research.

In each group, about 80% of participants could identify a specific event they felt may have led to the development of TSD. These included a change in technique, an increase in practice time for MD and an increase in need to write for WD participants. With respect to WD, in line with the views of other authors<sup>26</sup> it appears particularly challenging to predict the risk of WD given the numerous variables which may contribute to this regarding job description, working pressure and ergonomics. However, the results of our survey identify that MD tremolo, a specific piece, obsessive practice, and poor technique were perceived by MD participants as more than likely leading to the development of symptoms and a significant life event or an increase in need to write in the WD group.

Regarding treatment approaches used, the MD participants reported a change of technique, slow down exercise therapy, focusing on using large free movements and orthoses were helpful treatments that they had tried as part of a rehabilitation program. In the WD group modification to pen, massage, change of technique and ultrasound therapy were reported as being effective. There was a significant association between the two groups with respect to their perceived benefit of massage, modifications to the instrument and ultrasound therapy. A recent study by Cyganska<sup>30</sup> investigated the effects of massage and exercise on musculoskeletal pain of young musicians. Whilst this study does not relate specifically to MD it showed that massage and exercise (focusing on strength and movement for the whole body) should be used regularly for treating and reducing pain levels. A number of small scale studies have investigated rehabilitation interventions for TSD, providing evidence to support their effectiveness: slow down exercise therapy,<sup>31</sup> sensory re-education<sup>32</sup> and orthoses (constraint induced movement therapy).<sup>33</sup> Butler et al<sup>11</sup> found that combining individual treatment techniques seems to assist most people affected by TSD. Behavioral therapies and interdisciplinary strategies combining pharmacological and pedagogical methods are promising, but the different approaches need to be evaluated, and long-term effects are still unknown.<sup>34</sup>

Initially symptoms may relate purely to the act of playing a musical instrument or writing but symptoms tend to spread.<sup>35</sup> Our study showed that a wide variety of daily activities were affected by MD and WD and this is in line with others that show that writing or typing<sup>36</sup> in addition to several everyday activities<sup>34</sup> are affected by the task-specific symptoms.

### **Strengths and limitations of the study**

Given the rarity of TSD the existing sample set of 105 is relatively large. This convenience sample, however, does have limitations, given that it was comprised of patients who had attended a single, private hand therapy clinic. Response bias is therefore possible due to self-selection by the interest of individuals. Furthermore, the MD survey questionnaire was long, which may have accounted for the drop off. However, the sample characteristics suggest that, at least in terms of demographic and diagnostic features, it is like others that have been reported in the literature.

Of importance, the very nature of surveys mean that the results only reflect the participants perceptions; the effectiveness of interventions should be interpreted within this context. We also do not know how long the treatment options were used, which would have provided valuable information. Rigorously designed randomized controlled studies are required to investigate the short- and long-term effectiveness of specific interventions for this patient group.

## **Conclusion**

The findings of this survey help to characterize participants with TSD and describe their perceptions as to what may have led to dystonia developing (a change in technique), which rehabilitative treatment approaches were most utilized (altering playing/writing technique and modifications to musical or writing instrument/implement) and most effective (massage, modification to the instrument/implement and ultrasound therapy) and the manner in which symptoms may affect functional daily activities (brushing teeth, drawing, thinking about writing and using chopsticks) and specific activities of playing their musical instrument and writing.

Evidence for which medical and rehabilitation therapies are effective in treating and managing TSD is limited with minimal guidance on treatment selection, intensity, and efficacy and significant limitations in outcome measures. Further research in this area is needed.

Very few musicians return to normal motor control using the currently available therapies and whilst rehabilitative interventions assist in managing the condition, they do not cure it. Prevention should be the focus and with this it seems reasonable to pay specific attention to all young musicians learning to play “at risk instruments” so they hopefully avoid developing dystonia. Promotion of healthy work habits and advising varying task performance whilst working is paramount.

## HIGHLIGHTS

- A descriptive survey which explores the interventions that patients have utilized and their perceived effectiveness in treating and managing their TSD, specifically musicians' dystonia (MD) and writer's dystonia (WD). Symptoms and the effect of TSD on the perceived performance of every day and specific tasks, and possible reasons why the condition developed, are also investigated.
- Results for both the MD and WD groups were similar. With significant association between the two groups as to what they viewed may have led to the development of TSD and the most effective treatments. Participants perceived three treatments to be most effective, suggesting these should be considered for inclusion within treatment plans.
- Although full levels of task specific function (playing their instrument or writing) were not usually regained, participants with both MD and WD perceived improvement in symptoms with a combination of rehabilitative interventions. Initially symptoms may relate purely to the act of playing a musical instrument or writing but symptoms tend to spread, affecting a wide variety of daily activities.
- It is reported that up to 62% of musicians affected by this condition are unable to continue their performance careers, with few returning to normal motor control using the currently available therapies. Whilst rehabilitative interventions assist in managing the condition, they do not cure it. Prevention should be the focus, so people hopefully avoid developing this condition. Promotion of healthy work habits and advising varying task performance whilst working is paramount.

## Bibliography

1. Altenmuller E, Jabusch H. Focal dystonia in musicians: phenomenology, pathophysiology, triggering factors, and treatment. *Medical Problems of Performing Artists*. March 2010;25(1):3-9.
2. Peterson DA, Berque P, Jabusch HC, Altenmuller E, Frucht SJ. Rating scales for musicians' dystonia. *Neurology*. August 2013;81(6):589-598.
3. Altenmuller E, Lee A, Ioannou CI. Musician's dystonia. *The musicians hand - A clinical guide*. London: JP Medical Ltd.; 2018.

4. Stahl CM, Frucht SJ. Focal task specific dystonia: a review and update. *Journal of Neurology*. 2017;264(7):1536-1541.
5. Paulig J, Jabusch HC, Grosbach M, Boulet L, Altenmuller E. Sensory trick phenomenon improves motor control in pianists with dystonia: prognostic value of glove-effect. *Frontiers in Psychology*. 2014;5:1-7.
6. Karp BI, Cole RA, Cohen LG, Grill S, Lou JS, Hallett M. Long-term botulinum toxin treatment of focal hand dystonia. *Neurology*. 1994;44(1):70-6.
7. Brandfonbrener AG. Musicians with focal dystonia: A report of 58 cases seen during a ten-year period at a performing arts medicine clinic. *Medical Problems of Performing Artists*. 1995;10(4):121-127.
8. Schuele S, Lederman RJ. Long-term outcome of focal hand dystonia in string instrumentalists. *Movement Disorders*. January 2004;19(1):43-48.
9. Vugt FT, Boulet L, Jabusch HC, Altenmueller E. Musician's dystonia in pianists: Long-term evaluation of retraining and other therapies. *Parkinsonism and Related Disorders*. January 2014;20(1):8-12.
10. Jabusch H, Zschucke D, Schmidt A, Schuele S, Altenmuller E. Focal dystonia in musicians: Treatment strategies and long-term outcome in 144 patients. *Movement Disorders*. 2005;20:1623-1626.
11. Butler K, Sadnicka A, Freeman J, et al. Sensory-motor rehabilitation therapy for task-specific focal hand dystonia: A feasibility study. *Hand Therapy*. 2018;23(2):53-63.
12. Bowling A. *Research Methods in Health: Investigating Health and Health Services*. Buckingham: Open University Press; 2000.
13. Tubiana R. Musician's focal dystonia. *Hand Clinics*. 2003;19:303-308.
14. Nutt J, Muentner MD, Melton IJ. Epidemiology of dystonias in Rochester, Minnesota. *Advances in Neurology*. 1988;50:361-365.
15. Berque P, Gray H, Harkness C, McFadyen A. A combination of constraint-induced therapy and motor control retraining in the treatment of focal hand dystonia in musicians. *Medical Problems of Performing Artists*. 2010:149-161.

16. Payne J. Writer's Cramp. Focal Hand Dystonia Information. November 6, 2014.  
<https://patient.info/doctor/writers-cramp>. Accessed June 18, 2020.
17. Corballis MC. From mouth to hand: gesture, speech, and the evolution of right-handedness. *Behavioural and Brain Sciences*. 2003;26:199-208.
18. Zeuner KE, Molloy FM. Abnormal reorganisation in focal hand dystonia - sensory and motor training programs to retrain cortical function. *NeuroRehabilitation*. 2008;23:43-53.
19. Waddy HM. A genetic study of idiopathic focal dystonias. *Annals of Neurology*. 1991;29(3):320-324.
20. Schmidt A, Jabusch H, Altenmuller E. Etiology of musician's dystonia: Familial or environmental? *Neurology*. 2009:1248-1254.
21. Rosenkranz K, Williamon A, Butler K, Cordivari C, Lees AJ, Rothwell JC. Pathophysiological differences between musician's dystonia and writer's cramp. *Brain*. April 2005;128(4):918-931.
22. Rosenkranz K, Butler K, Williamon A, Rothwell JC. Regaining motor control in musician's dystonia by restoring sensorimotor organisation. *Journal of Neuroscience*. 2009;29:14627-14636.
23. Rozanski VE, Rehfuss E, Botzel K, Nowak D. Task-Specific Dystonia in Professional Musicians. *Deutsches Arzteblatt*. December 2015;112:51-52.
24. Altenmuller E, Ioannou CI, Lee A. Apollo's curse: neurological causes of motor impairments in musicians. *Progress in brain research*. 2015;217:89-106.
25. Ioannou CI, Furuya S, Altenmuller E. The impact of stress on motor performance in skilled musicians suffering from focal dystonia: Physiological and psychological characteristics. *Neuropsychologia*. 2016;85:226-236.
26. Pritchard MH. Writer's cramp: is focal dystonia the best explanation? *Journal of The Royal Society of Medicine Short Reports*. July 2013;4(7):1-7.
27. Steinlechner S, Jabusch HC, Altenmuller E, et al. Personality profiles are different in musician's dystonia and other isolated focal dystonias. *Psychiatry Research*. 2018;266:26-29.



28. Mind for better mental health. 2020. Available at: <https://www.mind.org.uk/information-support/types-of-mental-health-problems/statistics-and-facts-about-mental-health/how-common-are-mental-health-problems/>. Accessed June 18, 2020.
29. Mental Health Foundation. 2020. Available at: <https://www.mentalhealth.org.uk/statistics/mental-health-statistics-depression>. Accessed June 18, 2020.
30. Cyganska A, Truszczynska-Baszak A, Tomaszewski P. Impact of exercise and chair massage on musculoskeletal pain of young musicians. *International Journal of Environment Research and Public Health*. 2020;17:1-10.
31. Sakai N. Slow-down exercise for the treatment of focal hand dystonia in pianists. *Medical Problems of Performing Artists*. 2006;21:25-28.
32. Byl NN, Archer ES, McKenzie A. Focal hand dystonia: effectiveness of a home program of fitness and learning-based sensorimotor and memory training. *Journal of Hand Therapy*. 2009;22:183-197.
33. Candia V, Schafer T, Taub E, et al. Sensory motor retuning: a behavioural treatment for focal hand dystonia of pianists and guitarists. *Archives of Physical Medicine Rehabilitation*. 2002;83:1342-1348.
34. Jabusch H, Altenmuller E. Focal Dystonia in musicians: from phenomenology to therapy. *Advances in Cognitive Psychology*. 2006;2:207-220.
35. Torres-Russotto D, Perlmutter JS. Task-specific dystonias: a review. *Annals of the New York Academy of Sciences*. 2008;1142:179-99.
36. Lederman R. Focal dystonia in instrumentalists: Clinical features. *Medical Problems of Performing Artists*. December 1991:132-136.

## Tables

**Table 1: Demographic and diagnostic characteristics for participants who completed 45% or more of the survey**

	<b>Musicians' dystonia</b>	<b>Writer's dystonia</b>	<b>All</b>
<b>Gender</b>	n=55 (%)	n=21 (%)	n=76 (%)
Male	46 (84)	8 (38)	54 (71)
Female	9 (16)	13 (62)	22 (29)
<b>Hand dominance</b>	n=55 (%)	n=21 (%)	n=76(%)
Right	48 (87)	17 (81)	65 (86)
Left	6 (11)	4 (19)	10 (13)
Ambidextrous	1 (2)	0	1 (1)
<b>Age (at time of survey completion)</b>	n=55(%)	n=21(%)	n=76(%)
<30	2 (4)	1 (5)	3 (4)
31-50	22 (40)	9 (43)	31 (41)
>50	31 (56)	11 (52)	42 (55)
<b>Family members with a neurological condition</b>	n=55 (%)	n=21 (%)	n=76(%)
No	53 (96)	21 (100)	74 (97)
Yes	2 (4)	0 (0)	2 (3)
<b>Time from first symptoms until diagnosis (years)</b>	n=50 (%)	n=21 (%)	n=71 (%)
0-6 months	18 (36)	4 (19)	22 (31)
6-12 months	9 (18)	4 (19)	13 (18)
1-2 years	12 (24)	7 (33)	19 (27)
2-3 years	1 (2)	3 (14)	4 (6)
3-4 years	3 (6)	0 (0)	3 (4)
>4 years	7 (14)	3 (14)	10 (14)
<b>Number of years had dystonic symptoms (years)</b>	n=50 (%)	n=21 (%)	n=71(%)
<b>0-4</b>			
<b>4-9</b>	7(14)	3 (14)	10(14)
<b>&gt;10</b>	20(40)	0 (0)	20(28)
	23(46)	18 (86)	41(58)

**Table 2: Symptom comparisons between groups at time of initial diagnosis and when completing the survey “current symptoms”**

	<b>MD (n=50)</b>	<b>WD (n=21)</b>	<b>p-value</b>
	<b>n (%)</b>	<b>n (%)</b>	
<b>Initial</b>			
Clumsiness	33 (66.0)	12 (57.0)	0.480
Decreased sensation	15 (30.0)	10 (48.0)	0.156
Involuntary movements	46 (92.0)	17 (81.0)	0.224
Lack of co-ordination	41 (82.0)	18 (86.0)	1.000
Pain	19 (38.0)	15 (71.0)	0.005*
Tremor	9 (18.0)	12 (57.0)	0.100
Weakness	26 (52.0)	17 (81.0)	0.023*
<b>Current</b>			
Clumsiness	34 (68.0)	13 (62.0)	0.620
Decreased sensation	15 (30.0)	6 (29.0)	0.904
Involuntary movements	47 (94.0)	14 (67.0)	0.006*
Lack of co-ordination	30 (60.0)	14 (67.0)	0.597
Pain	13 (26.0)	10 (48.0)	0.076
Tremor	7 (14.0)	10 (48.0)	0.007*
Weakness	25 (50.0)	12 (57.0)	0.582
<b>p-value (b)</b>	0.897	0.977	

Data were analyzed with Chi-square test and Fisher’s exact test, p-value (a) was analyzed between group and p-value (b) was analyzed within group.

\* Statistically significant at the level of 0.05.

**Table 3: Medical treatments participants have received in the past (before survey completion)**

	<b>Musicians’ dystonia</b>	<b>Writer’s dystonia</b>	<b>All</b>
<b>Taking medications</b>	n=55 (%)	n=21 (%)	n=76 (%)
No	39 (71)	15 (71)	54 (71)
Yes	16 (29)	6 (29)	22 (29)
<b>Botulinum toxin in the past</b>	n=50(%)	n=20(%)	n=70(%)
No	36 (72)	8 (40)	44 (63)
Yes	14 (28)	12 (60)	26 (37)
<b>Surgery in the past</b>	n=50(%)	n=20(%)	n=70(%)
No	45 (90)	20 (100)	65 (93)
Yes	5 (10)		5 (7)
CTR	2 (40)		
Trigger finger release	3 (60)		

**Table 4: Participant views of what may have led to the development of TSD**

	MD (n=50)		WD (n=20)		p-value
	Ranking	n (%)	Ranking	n (%)	
A change in technique	1	30 (60.0)	5	3 (15.0)	0.001*
An increase in practice time or the need to write	2	21 (42.0)	1	9 (45.0)	0.819
An anxiety provoking event	3	10 (20.0)	4	4 (20.0)	1.000
Nothing that I can think of	4	9 (18.0)	4	4 (20.0)	1.000
A significant life event	4	9 (18.0)	2	6 (30.0)	0.337
An injury or pain in the affected area	5	5 (10.0)	3	5 (25.0)	0.135
A new instrument	5	5 (10.0)	-	0 (0.0)	0.312
Hypermobility (e.g. flexible or lax joints)	6	4 (8.0)	5	3 (15.0)	0.399
A change in your workplace or work role (MD only)	7	2 (4.0)	-	-	-
Regularly playing in a confined space or writing on a small piece of paper/notebook	8	1 (2.0)	6	1 (5.0)	0.493
Writing through many layers of paper (WD only)	-	-	6	1 (5.0)	-

Data were analyzed with Chi-square test and Fisher's exact test.

\* Statistical significant at the level of 0.05.

**Table 5: Rehabilitation interventions participants had tried**

	<b>MD (n=50)</b>	<b>WD (n=20)</b>	<b>p-value</b>
	n (%)	n (%)	
Rehabilitative			
Acupuncture	13 (26.0)	9 (45.0)	0.122
Alexander Technique	16 (32.0)	3 (15.0)	0.148
Altering playing/writing technique	46 (92.0)	14 (70.0)	0.027*
Biofeedback therapy	6 (12.0)	3 (15.0)	0.708
Counselling or psychotherapy	13 (26.0)	6 (30.0)	0.734
Custom made orthoses	38 (76.0)	15 (75.0)	1.000
Enforced rest	28 (56.0)	9 (45.0)	0.405
Focusing on using larger freer movements that come from the shoulder rather than the wrist or hand	30 (60.0)	16 (80.0)	0.111
Massage	28 (56.0)	13 (65.0)	0.490
Mirror therapy	24 (48.0)	9 (45.0)	0.820
Modifications to musical or writing instrument/implement	13 (26.0)	13 (65.0)	0.002*
Playing slowly with gentle increases in tempo as tolerated/writing slowly using large letters	45 (90.0)	16 (80.0)	0.265
Proprioceptive training	7 (14.0)	3 (15.0)	1.000
Ready-made orthoses	22 (44.0)	10 (50.0)	0.649
Sensory re-education	42 (84.0)	17 (85.0)	1.000
Sensory tricks	44 (88.0)	17 (85.0)	0.708
Stretching exercises	42 (84.0)	15 (75.0)	0.498
Therapeutic putty	41 (82.0)	14 (70.0)	0.337
Ultrasound therapy	22 (44.0)	10 (50.0)	0.649

Data were analyzed with Chi-square test and Fisher's exact test.

\* Statistical significant at the level of 0.05.

**Table 6: Participants perceived effectiveness of rehabilitation interventions**

	<b>MD (n=50)</b>	<b>WD (n=20)</b>	<b>p-value</b>
	<b>n of effective (%)</b>	<b>n of effective (%)</b>	
Acupuncture	0 (0.0)	2 (22.0)	0.156
Alexander Technique	2 (13.0)	1 (33.0)	0.422
Altering your writing/playing technique	25(50.0)	9 (45.0)	0.705
Biofeedback therapy	2 (33.0)	1 (33.0)	1.000
Counselling or psychotherapy	3 (23.0)	2 (33.0)	1.000
Custom made orthoses	13 (34.0)	7 (47.0)	0.399
Enforced rest	7 (25.0)	4 (44.0)	0.404
Using larger freer movements that come from the shoulder	12 (24.0)	8 (40.0)	0.181
Massage	9 (32.0)	9 (69.0)	0.043*
Mirror therapy	6 (25.0)	4 (44.0)	0.400
Modifications to instrument	2 (15.0)	10 (77.0)	0.002*
Playing/writing slowly with gentle increases in speed/difficulty as tolerated	24 (48.0)	8(40.0)	0.544
Proprioceptive training	1 (14.0)	1 (33.0)	1.000
Ready-made orthoses	8 (36.0)	5 (50.0)	0.699
Sensory re-education	10 (24.0)	7 (41.0)	0.214
Sensory tricks	11 (25.0)	4 (24.0)	1.000
Stretching exercises	12 (29.0)	7 (47.0)	0.202
Therapeutic putty	6 (15.0)	5 (36.0)	0.124
Ultrasound therapy	3 (14.0)	6 (60.0)	0.013*

Data were analyzed with Chi-square test and Fisher's exact test.

\* Statistically significant at the level of 0.05.

**Table 7: Top daily living tasks affected by TSD**

<b>Activities</b>	<b>n (%)</b>
<b>MD</b>	<b>50 (100)</b>
Typing	29 (58.0)
Thinking about playing a musical instrument	20 (40.0)
Writing	17 (34.0)
Fastening buttons	15 (30.0)
Drawing	12 (24.0)
<b>WD</b>	<b>21 (100)</b>
Drawing	17 (81.0)
Typing	13 (61.9)
Thinking about writing	11 (52.4)
Using cutlery/chopsticks	11 (52.4)
Brushing teeth	9 (45.0)
Fastening buttons	9 (45.0)
Applying makeup	8 (57.1)