

# Focal Dystonia is a Movement Disorder Manifesting among Musicians: A Single Case Study

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**Abstract** – Focal dystonia among musicians manifests as involuntary movements during musical performance, which may potentially ruin the musicians' professional careers. Based on the preceding studies concerning pathophysiology of focal dystonia, the authors assumed that focal dystonia is mainly triggered by abnormal sensory input caused by dysmyotonia or shortened soft tissues, thereby decreasing the inhibitory system activities in the cerebral cortex. This report depicts how a focal dystonia patient, a professional cellist, went through a six-month course of regular outpatient treatments, in which the patient's abnormal soft tissues were intervened. After the course, combined with the patient's self-maintenance routines monitored through interviews, both the pain and abnormal muscle tension were reduced, improving symptoms of focal dystonia.

**Keywords** – Focal Dystonia, Musicians' Cramp, Musician's Dystonia, Occupational Therapy.

## I. INTRODUCTION

Focal dystonia is a movement disorder manifesting among musicians. Specifically, it is ataxia or voluntary movement control disorder during instrumental performance [1,2]. The symptom, for example, includes a cellist whose middle finger involuntarily inflecting while trying to press a string with his/her adjacent index finger. Focal dystonia is reported to be seen in about 1% of the entire musician population [2]. Because the involuntary movements often manifest during performance, it could ruin the musicians' professional career [3]. Some reports explain the pathophysiology of focal dystonia as degenerated basal ganglia and dysfunction of the thalamic afferent pathway [4, 5], but it has become clearer that the abnormal sensory input from the afferent pathway, caused by hours of practice without taking heed of pains and muscle fatigue, is the main factor of the symptom [6]. According to a report, the chronic abnormal sensory input excessively suppresses the excitement signals to the muscle tension inhibitory system in the cerebral cortex, resulting in increased muscle tension in the affected area [7]. Studies on the factors involved in the onset of focal dystonia suggested links with certain psychological dispositions, such as perfectionism and strong anxiety [2, 8, 9], while other studies imply extreme amounts of practice hours compared to other musicians [10] and continuous, grueling practice without any maintenance routine to look after the shortening soft tissues and pain in the affected fingers as potential triggers for focal dystonia [2, 11]. In terms of gender difference, male musicians are found to be twice more susceptible to the disorder than female counterparts [12], while another report points out hereditary factors [13]. Focal dystonia may be treated with drugs, such as trihexyphenidyl [14] and Botox [15], but these medications only offer symptomatic therapy through the relaxation of regions affected with involuntary movements. Therefore, treatments corresponding to pathophysiological factors are required to essentially improve symptoms. For a professional musician, neglecting practice or avoiding correct fingering is impossible, and they cannot escape from the psychological pressure at concerts either. For these reasons, the first author focused on the importance of alleviating the abnormal muscle tension and shortening of soft tissues, the two factors directly contributing to abnormal sensory input, as a measure to improve focal dystonic symptoms. As there have been no previous studies focusing on muscle tension and/or shortening of the area affected by focal dystonia other than reports on drug

therapy, this study conducted intervention aiming at continuously adjusting muscle tension and improving the shortening of the affected area and verified its effect on the symptom, which are presented herein together with discussions.

## II. METHOD

### A. About the Patient

The patient is a woman in her thirties and a professional cellist. She has been practicing cello for more than twenty years. Presently, she holds solo concerts but also performs as an orchestra member several times a year. About two years ago, her ring finger started to inflect whenever she used the middle finger to press a string. After the onset, she continued to perform by using a fingering which excluded the use of the middle finger, but this solution disabled her from playing pieces requiring highly advanced techniques. The symptom manifested only during performances. Focal dystonia was not observed in any activities of daily life. The manifestation of symptom was often preceded by pain in the fingers during performance. The pain was felt in the left thenar area, the muscle bellies of the wrist flexor and the finger flexor. The patient consulted with a doctor a year ago, where she was diagnosed as focal dystonia. The patient was referred to the first author through a mutual acquaintance. At the initial evaluation, the interphalangeal joint of the ring finger inflected up to about 60% of the total active motion whenever she used her middle finger to press a string, making it difficult for her to use the middle finger in performance. Hoffmann's reflex and Tromner's reflex were positive on both hands. The muscle tension of the wrist and finger flexors was mildly increased. While the patient conducted stretching exercise of the upper limbs and fingers before practice and stage performances, she did not perform maintenance after practices or performances.

### B. Evaluation

In order to verify the effect of intervention, the study evaluated the pain, state anxiety about performance and trait anxiety, and the movement of the afflicted finger during performances before and after the intervention with the methods described as below. The pain was evaluated with Visual Analogue Scale (VAS). The study targeted the left thenar and the muscle bellies of wrist and finger flexors, for which the patient expressed pain at the initial evaluation, and evaluated the onset of pain during performances. For the evaluation of anxiety about performance and constitutional tendency towards anxiety before and after intervention, the study used State-Trait Anxiety Inventory (STAI) [16]. STAI is a questionnaire consisting of a set of 20 questions for state anxiety and another set of 20 questions for trait anxiety. Each question is answerable by four scales: 1. Not true at all; 2. True to a certain extent; 3. More or less true; and 4. 100% true. The four scales are scored as 1, 2, 3 and 4 points respectively, and the total score for each set of questions for state anxiety and trait anxiety becomes the result. The lowest total score is 20 points, while the highest is 80 points. A higher total score signifies stronger anxiety. Furthermore, the study used Modified Ashworth Scale (MAS) [17] to evaluate the muscle tension of the wrist and finger flexors before and after the intervention. The movement of the affected finger during performance before and after intervention was taped to visually analyze the motions. The first author responsible for the evaluation of the case is a string player in an amateur orchestra with basic knowledge on how to play cello. The author had also provided occupational therapy for more than ten focal dystonia patients in the past.

### C. Intervention

The patient agreed to six months of evaluation period. The intervention and evaluation were conducted by the first author, who treated the case as a hospital outpatient. The initial intervention session was about how to conduct self-maintenance of the soft tissues. Also, the session included instructions on how to use breathing as a way of avoiding excessive tension during performance, as well as consultations on better finger combinations that are less harmful to fingers. Furthermore, the patient was asked to itemize daily life activities from wake time till bed time on a weekday and in a weekend to identify the average lifestyle.

Then, a maintenance program with customized menus for before and after daily practices and stage performances was prepared and instructed. The purpose of the maintenance program is to avert abnormal sensory input to the central nervous system by alleviating abnormal muscle tone or shortening of soft tissues. The program consisted of exercise and stretch of scapular arches and shoulder joints, forearm and wrist stretching, finger and intermetacarpal stretching, massaging of interdigital space, pain test and cooling of interphalangeal joints, etc. In addition, the first author conducted 40 minutes of manipulative therapy and 20 minutes of monitoring interview every week. Apart from checking how the instructions were followed and getting feedbacks on the progress, mental support for the patient was also offered during the monitoring interviews by carefully listening to the patient's anxiety, frustration and agony she felt towards the prospect of recovery as a professional. Furthermore, the first author provided a manipulative therapy on concert days before and after the performances, as the affected finger went through long hours of stress, which could not be sufficiently taken care of through self-maintenance only.

The manipulative therapy included the fascial release of the shortened soft tissues and muscle tension suppression mobilization of digital flexor muscles.

#### *D. Research Ethics*

The study was conducted under the approval of Shonan University of Medical Sciences Research Ethics Committee (Approval No.: 17-001). Upon receiving oral and written explanations by the first author, the patient consented to the study.

### **III. INTERVENTION RESULT**

The patient underwent six months of intervention menus, which included 29 outpatient treatments and interviews and five sessions of treatment before and after stage performances. The patient fully complied with the instructed maintenance program every day and successfully improved the breathing during performance by following the instructed method. At the interview conducted two months after the initial intervention, she was already able to gradually include the affected finger in the fingering.



Fig. 1. Ring finger movement before and after intervention approach to soft tissues during performance.

The result of Visual Analogue Scale (VAS) before and after the intervention showed alleviated pain both in the left thenar (from 48mm to 24mm) and in the muscle bellies of wrist and finger flexors (from 42mm to 17mm). STAI scores only showed a limited improvement in trait anxiety (42 points before intervention to 40 points after intervention), but the state anxiety, measured while the patient recalled a stage performance, improved from 54 points before intervention to 20 points after intervention. MAS for the wrist flexors and MAS for finger flexors respectively improved from 1+ to 1. The movement of the affected ring finger during performance, which initially inflected involuntarily every time the middle finger was used, almost completely ceased to show any involuntary inflection after six months of intervention, and the dystonic symptom became unnoticeable. (See Figure).

#### IV. DISCUSSIONS

While the onset mechanism of focal dystonia remained unclear, studies are gradually identifying combinations of multiple factors contributing to the abnormal suppression of nervous excitement, which is supposed to suppress muscle tension in the overlapping localized areas controlling fingers and in the cerebral cortex. Among several reasonable factors, this study assumed that the overuse of body parts through strenuous practice triggering the constant abnormality of soft tissues and creating abnormal sensory information input as the main factor of the onset mechanism of focal dystonia and intervened in its symptoms. The intervention successfully alleviated the dystonic symptoms and achieved a result which suggests transformation of abnormal muscle tension to that of normal one. According to previous studies, both patients of focal dystonia and patients of chronic pain suppress the control system involved in the muscle tension suppression mechanism during the adjustment process of muscle tension in the cerebral cortex [18-20], suggesting that the symptom of focal dystonia is mainly triggered by the frequent onset of pain. The patient in this study experienced pain in the affected finger which preceded the manifestation of focal dystonia symptom. As the prodromal symptom matches the finding in a report that 9% of patients with focal dystonia experience pain prior to the onset of dystonic symptoms [21], the patient in this study was suggested to fall under this patient category. The role of pain as a trigger of focal dystonia is undeniable, and the pain may be caused as a result of continuous involuntary movements and performance in a modified posture. According to previous studies, the average age of onset of focal dystonia is 33 [22, 23], and the patient in this study also experienced the initial manifestation at the syndrome's peak age of onset. For professional performers, thirties are when their horizon as a successful musician significantly expands. Therefore, the psychological shock brought about by the onset is imaginable. Because the frustration and desperation may adversely affect the recovery process, provision of mental support, in addition to physical support, may be important in intervention efforts.

#### V. CONCLUSION

Therapeutic intervention on abnormal soft tissue was performed regularly on a cellist outpatient presenting symptoms of focal dystonia for six months, and interviews were conducted to monitor the patient's self-maintenance. As a result, aching decreased and abnormal muscle tone was reduced, thus demonstrating improvement in the focal dystonia symptoms.

#### VI. LIMITATIONS AND ISSUES

This study focused on the soft tissue abnormality and verified the effect of intervention. However, the pathological development of central nervous system responsible for the onset of focal dystonia remain unsolved.

Also, the method introduced in this report might not be proved effective for all focal dystonia. Because all onset factors mentioned in preceding studies so far remain hypotheses, clarification of pathology is indispensable for a valid verification of the effective intervention. At the same time, it is important to accumulate clinical evidences until the pathological explanations are sufficiently supported.

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### Conflict of Interest

None declared.

### REFERENCES

- [1] Fruchtn S, Fahn S, Greene P: The natural history of embouchure Dystonia. *Mov. Disord* , 2001, pp. 899-906..
- [2] Altenmüller E: Focal dystonia: advances in brain imaging and understanding of fine motor control in musicians. *Hand Clin* , 2003, pp. 523-538.
- [3] Bara-Jimenez W, Catalan M, Hallett M, Gerloff C: Abnormal somatosensory homunculus in dystonia of the hand. *Ann. Neurol*, 1998, pp. 828-831.
- [4] Lenz F, Byl N: Reorganization in the cutaneous core of the human thalamic principal somatic sensory nucleus (Ventral caudal) in patients with dystonia. *J Neurophysiol*, 1999, pp. 3204-3212.
- [5] Naumann M, Pirker W, Reiners K, Lange K, Becker G, et al: Imaging the pre- and postsynaptic side of striatal dopaminergic synapses in idiopathic cervical dystonia: a SPECT study using [123I] epidepride and [123I] beta-CIT. *Mov. Disord*, 1998, pp. 319-323.
- [6] Rosenkranz K, Williamon A, Butler K, Cordivari C, Lees A, et al: Pathophysiological differences between musician's dystonia and writer's cramp. *Brain*, 2005, pp. 918-931.
- [7] Hummel F, Andres F, Altenmüller E, Dichgans J, Gerloff C: Inhibitory control of acquired motor programmes in the human brain. *Brain*, 2002, pp. 404-420.
- [8] Lim V, Altenmüller E: Musicians' cramp: Instrumental and gender differences. *Med. Probl. Perform*, 2003, pp. 21-26.
- [9] Schmidt A, Jabusch H, Altenmüller E, Hagenah J, Brüggemann N, Hedrich K, et al: Dominantly transmitted focal dystonia in families of patients with musician's cramp. *Neurology*, 2006, pp. 691-693.
- [10] Jabusch H, Zschucke D, Schmidt A, Schuele S, Altenmüller E: Focal dystonia in musicians: treatment strategies and long-term outcome in 144 patients. *Mov. Disord* , 2005, pp.1623-1626.
- [11] Charness M, Ross M, Shefner J: Ulnar neuropathy and dystonic flexion of the fourth and fifth digits: clinical or relation in musicians. *Muscle Nerve*, 1996, pp. 431-437.
- [12] Lederman R: Focal dystonia in instrumentalists: clinical features, 1991, pp. 132-136.
- [13] Schmidt A, Jabusch H, Altenmüller E, Hagenah J, Brüggemann N, Hedrich K, et al: Dominantly transmitted focal dystonia in families of patients with musician's cramp. *Neurology*, 2006, pp. 691-693.
- [14] Jabusch H, Zschucke D, Schmidt A, Schuele S, Altenmüller E: Focal dystonia in musicians: treatment strategies and long-term outcome in 144 patients. *Mov. Disord*, 2005, pp. 1623-1626.
- [15] Zoons E, Dijkgraaf M, Dijk J, van Schaik I, Tijssen M: Botulinum toxin as treatment for focal dystonia: a systematic review of the pharmaco-therapeutic and pharmaco-economic value. *J Neurol*, 2012, pp. 2519-2526.
- [16] Spielberger C, Sydeman S: State-Trait Anxiety Inventory and State-Trait Anger Expression Inventory. Lawrence Erlbaum Associates, 1994, pp. 292-321.
- [17] Bohannon RW, Smith MB : Interrater reliability of a modified Ashworth scale of muscle spasticity. *Phys Ther* , 1987, pp. 206-207.
- [18] Tremblay F, et al: Effects of prolonged muscle stretch on re ex and voluntary muscle activations in children with spastic cerebral palsy. *Scandinavian Journal of Rehabilitation Medicine*, 1990, pp. 171-180.
- [19] Flor H, Braun C, Elbert T, Birbaumer N: Extensive reorganization of primary somatosensory cortex in chronic back pain patients. *Neurosci. Lett*, 1997, pp. 5-8.
- [20] Tinazzi M, Fiaschi A, Rosso T, Faccioli F, Grosslercher J, Aglioti S: Neuroplastic changes related to pain occur at multiple levels of the human somatosensory system: A somatosensory-evoked potentials study in patients with cervical radicular pain. *J. Neurosci* , 2000, pp. 9277-9283.
- [21] Jabusch H Müller S, Altenmüller E: Anxiety in musicians with focal dystonia and those with chronic pain. *Mov. Disord* , 2004, pp. 1169-1175.
- [22] Jankovic J, Shale H: Dystonia in musicians. *Semin. Neurol* , 1989, pp. 131-135.
- [23] Brandfonbrener A: Musicians with focal dystonia: A report of 58 cases seen during a ten-year period at a performing arts medicine clinic. *Med. Probl. Perform*, 1995, pp. 121-127.

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