

Dystonia in musicians an unrecognized cause of career loss.

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ABSTRACT: Musician's dystonia is a task-specific focal dystonia affecting approximately 1% of all professional musicians. Dystonia develops in all types of instruments (keyboards, strings, woodwind, brass and percussions), presenting as focal dystonia of the hand or as embouchure dystonia. There is an association between the musical instrument and the localization of dystonia (piano and guitar players are affected in the right hand, violinists and violists in the left hand). Genetic predisposition and a variety of triggering factors, such as trauma, compressed nerve, emotional stress and mainly overuse of the hand due to marked increase in practice time, are implicated for the development of the focal dystonia. Treatment of musician's dystonia is currently symptomatic and includes drugs, botulinum toxin injections, pedagogical training, relaxation techniques, ergonomic changes and psychotherapy. The results of the treatment are more or less unsatisfactory and a proportion of professional musicians are forced to change career.

Key Words: Musicians, Dystonia, Music instruments.

INTRODUCTION

Dystonia is an involuntary hyperkinetic movement disorder **characterized by patterned, sustained or repetitive** muscle contractions of opposing muscles, causing twisting movements and abnormal postures^{1,2}. Dystonic movements are almost always aggravated during voluntary movement. They tend to be increased with fatigue, stress and emotional state, **while relaxation**, sleep and tactile or proprioceptive sensory tricks suppress them. According to body distribution of the abnormal movements dystonia is classified into focal, segmental, multifocal, generalized and hemidystonia. Task-specific action dystonia refers to the appearing of abnormal movements with a special action such as writing, typing, playing an instrument e.t.c.

Musician's dystonia or musician's cramp is a task-specific dystonia presented as a painless muscular incoordination or loss of voluntary motor control in any part of the body that is engaged in repetitive, highly skilled tasks while a musician is playing the instrument^{3,4}.

EPIDEMIOLOGY AND DEMOGRAPHICS

Musicians use their hands continuously controlling the strength, pressure, amplitude and kinetics of independent fingers and hand movements. The rapidity and precision of these highly skilled movements is without parallel in any other profession. Furthermore, professional musicians push themselves to physical and mental extremes. These characteristics may explain the fact that focal dystonia is more common in musicians than other occupational dystonia. As many as 1 out of 200 musicians may be affected by dystonia during their career and at performing arts medical centers 8% to 14% of musicians seeking medical advice are diagnosed with dystonia^{5,6}. Symptoms usually begin in the mid to late 30s and early 40s, that is at the peak of musicians' careers; however, there is a wide range of disease onset from age 16 to 75^{3,5,6-11}. There is a preponderance of male (2/1 to 5/1) musicians in all studies.

The majority of musicians with dystonia are professionals (solists or playing in orchestras) who per-

form very frequently. Small percentage of patients are music teachers, students and amateurs^{3,9}. Furthermore, dystonia is more prevalent in classical musicians. It is rare in pop and jazz musicians who are more free to improvise^{3,7,8},

TYPE OF INSTRUMENT AND LOCALIZATION OF DYSTONIA

No type of instrument seems to be associated with immunity against the development of dystonia. The location of dystonia varies according to the instrument the musician plays. The hand and fingers are involved in keyboard (piano, organ, harpsichord, accordion), woodwind (clarinet, flute, oboe, saxophone, bassoon, bagpipe), plucked strings (guitar, bass, banjo, harp, mandolin), bowed strings (violin, viola, cello), brass (trumpet, French horn) and percussion (drums, tabla) instruments^{3,5,6,9}. Embouchure (the pattern of lip, jaw and tongue muscles used to control the flow of air into a mouthpiece) dystonia is reported in brass and woodwind players^{3,12}. Musicians with instruments requiring high levels of spatial sensorimotor precision were more often affected by dystonia. So in the study of Jabusch & Altenmuller (2006)³ the distribution of instrument groups among 144 musicians with dystonia was as follows: 28% keyboard instrumentalists (34 piano players, 5 organ and 1 harpsichord), 26% were woodwind players (14 flute players, 13 clarinet, 5 saxophone, 3 oboe and 1 bassoon), 20% were playing plucked strings (mostly guitar), 15% were bowed string players (16 violin players, 3 viola, 1 cello and 1 double-bass) and 11% were brass players (8 trombone players, 4 trumpet, 3 horn and 1 tuba).

There is an association between the instrument and the localization of the focal dystonia^{3,9,10,11}. Keyboard musicians and plucked string players were primarily affected in the right hand, that is the hand with higher workload. Violinists and violists who have a higher workload and complexity of movements in the left hand show dystonia in the left hand. Woodwind players may show dystonia of the hand (the right hand in clarinetists and flutists and the left in bagpipers), or the lip. Brass players are affected by embouchure dystonia. Finally percussionists and brass players show hand dystonia without clear lateralization. Dystonia in both hands was seen in keyboard and woodwind instruments since these instruments require similar movement patterns in both hands.

SYMPTOMS AND PHENOMENOLOGY OF DYSTONIC MOVEMENTS

The task-specific dystonia begins insidiously, although it may have an abrupt onset, on occasion triggered by a traumatic physical or emotional event. Often the onset of symptoms coincide with a period of intense musical activity or a change in technique. The initial symptoms include fatigue, a sense of loss of control, stiffness or cramping of muscles and abnormal movements (curling or straightening of the finger) during playing fast and demanding pieces^{9,10,11}. Pain, diminished speed and tremor are less common symptoms. The patient tries to correct the finger from remaining extended or flexed but this aggravates dystonia while the compensatory movements of the adjacent fingers which tend to move in the opposite direction deteriorate the musical performance even more. Symptoms may be disguised in slow passages and become apparent in very rapid passages. In cases with embouchure dystonia the patients are complaining of inability to initiate tone or control the sound with a certain range, lip fatigue and lip tremor¹².

Usually there is a long delay between onset of symptoms and correct diagnosis. Musicians are reluctant to seek medical attention, ascribing their difficulty to faulty practice and physicians do not consider dystonia in the differential diagnosis of the musician's motor dysfunction.

Phenomenologically characteristic patterns of dystonic movements are commonly seen, such as flexion of the 4th and 5th fingers in pianists, flexion of the 2nd, 3rd and 4th finger in guitarists, extension of the 3rd and 5th finger in clarinetists, flexion of the 4th and 5th finger of the bowed arm in violinists and shoulders in cymbal players^{6,9}. Conti et al (2008)⁵ reported the dystonic phenotype in 960 musicians with hand dystonia. In patients in whom only one finger was involved finger 3 was most commonly affected (37%), followed by finger 2 (23%). When multiple fingers were involved the most common combination was 4 and 5 fingers (32%), followed by combination 3 and 4 fingers (17%) and 3, 4 and 5 fingers (17%). The predominant dystonic movement was flexion of one or more fingers (54%), followed by extension (13%) and then other dystonic movements. In keyboard players involvement of finger 4 together with finger 5 was more common (33%), in plucked strings isolated finger 3

(18), in bowed string players isolated finger 4 and wrist/forearm/arm was equally involved (21% each), in woodwinds combination 4,5 (24%), in percussionists combination of fingers 3,4,5 or wrist/forearm/arm (27% each) and in brass players isolated finger 3 or combination 2,3 (25% each). The most common phenotype in embouchure dystonia was task-specific tremor of the lips. Other phenotypes include lateral pulling of one or both lips, elevation of one or both corners of the mouth, protrusion of both upper and lower lips, involuntary lip closure, involuntary movements of the jaw and velopharyngeal incompetence^{6,12}.

A characteristic and unique feature of dystonic movements is that they can be diminished or alleviated by tactile or proprioceptive sensory trick (such as touching the chin or the face in patients with torticollis, touching the lips in patients with orolingual dystonia). Characteristically, there is a report of a musician with embouchure dystonia who had considerable symptom relief after cooling the dystonic muscles¹³. Such sensory tricks are only occasionally seen in musicians (for example putting a piece of plastic between the teeth in embouchure dystonia)^{9,10,12}. An explanation for this relatively infrequent use of sensory tricks is that they are not possible during musical performance. According to Lederman¹⁰ there is lack of options for such maneuvers given the need for using both hands to play the instrument. Finally, an interesting point is the fact that although muscle fatigue in non-dystonic musicians worsens subsequent musical performance, in dystonic musicians fatiguing muscle contraction improved motor performance¹⁴.

PREDISPOSING FACTORS AND PATHOPHYSIOLOGY

Recent works propose that a combination of triggering factors and genetic predisposition are important in the development of musicians' dystonia. The epidemiology of musicians' dystonia suggests a genetic component in the pathogenesis of the disorder. Although only 10% of dystonic musicians have relatives with dystonia, detailed examination showed much higher prevalence of dystonia among family members of musicians with dystonia¹⁵. However, no GAG deletion in the DYT1 gene was found in 28 musicians with dystonia^{15,16}.

Potential triggers of dystonia were noticed in about

53% and 67% of the patients¹¹. Peripheral trauma has been described as potential trigger for dystonia. Dystonia may follow closely some major or minor trauma (fractures, burns, infections, injury to the lip, loss of tooth e.t.c.)^{3,5,6,9,12,17}. In some cases, compressed or pinched nerve can be identified (especially ulnar nerve compression at the elbow) preceding the appearance of dystonia^{17,18}. Overuse can also trigger dystonia. There are reports of acute onset of dystonia after a marked increase in practice time, increase in difficulty of repertoire and change in instrument mechanics^{3,9-11}. Furthermore, local pain and intensified sensory input are potential triggers of dystonia. Psychological conditions such as perfectionism and anxiety were found to be related to dystonia; anxiety has been described as an aggravating factor during onset of musician's dystonia^{3,19}.

Musicians developed remarkable skills through long, regular practice often beginning in early childhood. Using functional MRI, researchers reported that the advanced training of pianists allows them to control finger movements through reduced activation of secondary motor areas compared to non-musicians²⁰⁻²⁴. Lotze et al (2003)²⁵, comparing brain activity during performance between professional and amateur musicians, found that, although both groups showed cortical activity in primary sensory and primary motor areas that represented the hand, the activity in professionals was much more tightly focused spatially and in the primary motor cortex was more intense and confined to the contralateral side of the performing hand.

The pathophysiology of musician's dystonia is still obscure. There is growing evidence that focal dystonia is not a malfunction of peripheral motor control but has its origin in the central nervous system. Besides alterations of the basal ganglia circuits, several studies provided evidence for a disturbed central processing of sensory information, abnormalities in sensorimotor integration and distorted cortical representation of motor function^{3,6,21}. Based on the theory of aberrant neuroplasticity evidence is accumulating in support of the sensorimotor learning hypothesis^{26,27}. According to this theory focal hand dystonia (writer's cramp, musician's dystonia) reflects a maladaptive response of the brain to repetitive performance of stereotyped

movements. It postulates that the usual processes of reorganization that accompany learning of new tasks can be pushed to an extreme where they begin to interfere with task performance rather than improving it. So in monkeys that develop dystonia after being trained to carry out repetitive hand movements there is marked degradation of the sensory map of the hand²⁷. The receptive fields of cortical neurons receiving information from the fingers can be enlarged by a factor of 10-20. This causes considerable overlap between the cortical representation of different fingers and between the front and the back of the hand. Investigations in musicians with focal dystonia revealed similar maladaptive changes in cortical mapping. There is a breakdown of the normally sharply delineated representational zones of the digits and the finger representations of the dystonic hand are much closer together and may either overlap or be in random order²⁸. Finally, neurophysiological studies with repetitive transcranial magnetic stimulation revealed that musicians had enhanced long term potentiation/depression-like plasticity and a steeper recruitment of corticospinal excitatory and intracortical inhibitory projections, suggesting higher than normal gain of regulating plasticity and excitability²⁹. This finding was attributed to a increased synapses within the motor area, caused by long term extensive musical training.

PROGRESSION OF DYSTONIA

Following the onset of symptoms dystonia may remain limited to the limb initially affected or to the lip and after a few years tends to stabilize. However, in a proportion of patients dystonia may spread from one digit to an adjacent one and then to the entire hand and sometimes may involve the whole arm. In about 1/3 of the patients dystonia progresses to involve other activities (writing, cutting, speaking eating e.t.c.)^{9,11,12}. In the study of Rosset-Llobet et al (2007)³⁰ 53.5% of patients reported secondary motor disturbances in activities other than playing their main instrument, with the onset delayed in some cases by up to 12 years from the awareness of dystonic symptoms.

EVALUATION OF A MUSICIAN WITH DYSTONIA

Examination of the musician suspected for focal dystonia includes the following: 1) **History:** We must

not expect the patient to describe the dystonic movements. The musician will complain for difficulty in controlling speed and dexterity, stiffness or slowing of the finger, causing him to play wrong notes. 2) **Inspection:** The greater part of the diagnosis rests on the examination of the musician while playing, revealing the dystonic movements. 3) **Neurological examination,** with special attention to the presence of other dystonic movements or a peripheral neuropathy. 4) **Neurophysiological examination,** searching for evidence of associated nerve entrapment. 5) **Rating scales:** Currently there is no standard method for evaluation musician's dystonia. Many different scales have been used that are subjective or objective. Such scales are: Dystonia Evaluation Scale, Self-Rating Scale, Six-Step Self-Rating Score, Arm Dystonia Disability Scale and Tubiana-Chamagne Scale³¹. 6) **Automated methods,** such as Dexterity and Displacement Device, MIDI-Based Scale Analysis and Three-Dimensional Movement Analysis, which are objective and have the advantage of reduced rater bias³³. The MIDI-Based Analysis and the Three-Dimensional Movement Analysis are only available for pianists^{32,33}. Embouchure video stroboscopy is used for the evaluation of patients with embouchure dystonia⁹.

TREATMENT

Treatment of musician's dystonia is long and difficult, because pathogenesis-targeted therapy is not available and because the symptomatic treatment provides inconsistent or unsatisfactory results. This is particularly difficult for professional musicians who demand from themselves perfect motor function. Current symptomatic treatment includes medication with trihexyphenidyl and other drugs, injection of botulinum toxin, ergonomic changes, pedagogical training, relaxation techniques (including biofeedback, physical and occupational therapy) and psychotherapy^{3,10,11,34}. After treatment with trihexyphenidyl an improvement was reported in 1/3 of musicians with hand dystonia but the applicability of the drug was further limited due to side effects³⁴. Embouchure dystonia in general does not respond to anticholinergics³⁴. In some cases baclofen, tetrabenazine, carbamazepine, muscle relaxants sometimes combined with propranolol or primidone have been helpful⁶. Botulinum toxin injections have become the primary treatment option for

musician's dystonia. Schule et al (2005)³⁵ reported that 69% of the 84 musicians with hand dystonia experienced improvement after injections and 36% had long-term benefit in their performance ability. According to the authors botulinum toxin injections in hand muscles (intrinsic hand muscles, forearm flexors and forearm extensors) appear to be recommendable only in instrumentalists with limited demand on lateral finger motion (woodwind players and the right hand in guitarists). The most injected muscles in the series of patients of Jabuch and Altenmuller were flexor digitorum superficialis, flexor digitorum profundus, flexor carpi radialis, flexor pollicis longus, extensor digitorum, extensor indicis and interosseus palmaris. However the increased risk of muscle weakness after botulinum toxin injections is unacceptable for some musicians. Botulinum toxin treatment for embouchure dystonia is disappointing ;there is either no improvement or partial improvement that lasts only few days. Two musicians out of nine with embouchure dystonia had some improvement in their dystonia^{12,35}. Mechanical prevention of dystonic movements includes attaching splints to the affected fingers, modifications of the instrument to circumvent dystonic movements and support systems when the dystonic hand has to carry the instrument while playing. Pedagogical retraining comprise a variety of supervised behavioral approaches. Jabusch et al (2005)³⁴ evaluated the long-term outcome of 144 musicians with dystonia treated with different treatment strategies. An alleviation of the symptoms was found in 54% of all musicians (63%, 56%, 50%, 49% and 33% of the patients who had been treated with ergonomic changes, technical exercises, pedagogical retraining, botulinum toxin and trihexyphenidyl respectively). An extensive rehabilitation program based on identifying maladaptive postural and technical factors and working with the musicians to overcome them was described by Tubiana and Chamagne^{8,10}. Priori et al (2001)³⁶ reported a treatment method including immobilization of the dystonic forearm and hand for 4-5 weeks followed by a rehabilitation program. Candia et al³⁷ employed a behavioral procedure, the method of sensory motor retuning, for the treatment of focal hand dystonia in musicians. The non-dystonic fingers were immobilized by splints while the dystonic finger performed systematic training with the musical instrument. The

authors have found both clinical improvement and reorganization of the central somatosensory neural networks^{37,38}.

In general, treatment of focal dystonia continues to be disappointing. The response to different types of treatment strategies varies between none to mild or moderate improvement, often insufficient to maintain the high level of performance. Some professional performing musicians are forced to change career path and instrumental students to change profession. Anyhow with the available treatment a considerable percentage of patients manage to stay in their profession although with substantial compromises.

Δυστονία σε μουσικούς: μία αδιάγνωστη αιτία διακοπής της καριέρας.

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ΠΕΡΙΛΗΨΗ: Η δυστονία στους μουσικούς είναι μία εστιακή δυστονία εξειδικευμένου έργου που προσβάλλει περίπου το 1% των επαγγελματιών μουσικών. Η εστιακή δυστονία εμφανίζεται στους μουσικούς όλων των μουσικών οργάνων (κλειδοκύμβαλα, έγχορδα, κρουστά, πνευστά) υπό την μορφή δυστονίας των άνω άκρων ή δυστονίας του επιστομίου. Η εντόπιση της εστιακής δυστονίας εξαρτάται από το είδος του μουσικού οργάνου, π.χ. δεξιό χέρι για τους πιανίστες και κιθαρίστες, αριστερό χέρι για τους βιολιστές. Στην παθογένεια της δυστονίας εμπλέκονται η γενετική προδιάθεση και πολλοί εκλυτικοί παράγοντες, όπως τραύμα, πίεση νεύρου, συναισθηματική φόρτιση και ιδιαίτερα η παρατεταμένη χρήση του μουσικού οργάνου. Η αξιολόγηση της εστιακής δυστονίας στους μουσικούς περιλαμβάνει υποκειμενικές και αντικειμενικές κλίμακες αξιολόγησης καθώς και αυτοματοποιημένους τρόπους (στους πιανίστες). Η θεραπεία της νόσου (φαρμακευτική αγωγή, ενέσεις αλλαντικής τοξίνης, εργονομικές μεταβολές, συμπεριφορική προσέγγιση κ.ά.) είναι μόνον συμπτωματική και τα αποτελέσματα όχι ιδιαίτερα ικανοποιητικά, έτσι αρκετοί επαγγελματίες μουσικοί οδηγούνται σε αλλαγή καριέρας.

Λέξεις Κλειδιά: Δυστονία, Μουσικοί, Μουσικά όργανα.

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