Review

Focal Dystonia in Musicians

Phenomenology, Pathophysiology, Triggering Factors, and Treatment

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Musician's dystonia is a task-specific movement disorder that manifests itself as a loss of voluntary motor control in extensively trained movements. Approximately 1% of all professional musicians develop musician's dystonia, and in many cases, the disorder terminates the careers of affected musicians. The pathophysiology of the disorder is not completely clarified. Findings include 1) reduced inhibition at different levels of the central nervous system, 2) maladaptive plasticity and altered sensory perception, and 3) alterations in sensorimotor integration. Epidemiological data demonstrate a higher risk for those musicians who play instruments requiring maximal fine-motor skills. For instruments where workload differs across hands, focal dystonia appears more often in the more intensely used hand. In psychological studies, musicians with dystonia have more anxiety and perfectionist tendencies than healthy musicians. These findings strengthen the assumption that behavioral factors may be involved in the etiology of musician's dystonia. Preliminary findings also suggest a genetic contribution to focal task-specific dystonia with phenotypic variations including musician's dystonia. Treatment options include pharmacological interventions, such as trihexyphenidyl or botulinum toxin-A, as well as retraining programs and ergonomic changes in the instrument. Patient-tailored treatment strategies may significantly improve the situation of musicians with focal dystonia. Positive results after retraining and unmonitored technical exercises underline the benefit of an active involvement of patients in the treatment process. Only a minority of musicians, however, return to normal motor control using the currently available therapies. *Med Probl Perform Art* 2010; 25:3–9.

Focal dystonia in musicians, also known as musician's dystonia or musician's cramp, is a task-specific movement disorder that presents itself as muscular incoordination or loss of voluntary motor control of extensively trained movements while a musician is playing the instrument. For those who are affected, focal dystonia is highly disabling, and in many cases the disorder terminates musical careers.[1]

Musician's dystonia may be classified according to the task specifically involved. For example, embouchure dystonia may affect coordination of the lips, tongue, facial and cervical muscles, and breathing in brass and wind players, whereas pianist's cramp and violinist's cramp may affect the control of finger, hand, and arm movements.

Various symptoms can mark the beginning of the disorder: Subtle loss of control in fast passages, finger curling (Figure 1), lack of precision in forked fingerings in woodwind players, irregularity of trills, fingers sticking on the keys, involuntary flexion of the bowing thumb in strings, and impairment of control of the embouchure in woodwind and brass players in certain registers are among the most frequent descriptions patients give. At this stage, most musicians believe that the reduced precision of their movements is due to a technical problem or lack of practice. As a consequence, they intensify their efforts, but this reaction often exacerbates the problem.

According to recent estimates, 1% of all professional musicians are affected.[1] In contrast, in the general population, prevalence of focal dystonias, including writer's cramp, blepharospasm, and cervical dystonia, is estimated as 29.5/100,000 in the United States and 6.1/100,000 in Japan.[2,3] In comparison to other activities producing dystonic movements, such as writing, playing golf (the "yips"), or darts ("dartism"), classical musicians are at highest risk to develop focal dystonia.

Typically, musician's dystonia occurs without pain, although muscle aching can present after prolonged spasms. Lack of pain distinguishes it from repetitive strain injury or occupational fatigue syndrome. It is important to make this distinction, bearing in mind that prolonged pain syndromes may lead to symptomatic dystonia. The loss of muscular coordination is frequently accompanied by a co-contraction of antagonist muscle groups. For example, in pianist's cramp the co-activation of wrist flexor and wrist extensor muscles is frequently observed. In Figure 1, typical postures of dystonic finger and embouchure movements in musicians are shown.

RISK FACTORS: WHO DEVELOPS MUSICIAN'S DYSTONIA?

Generally, demographic data demonstrate a preponderance of male musicians developing musician's dystonia, with a male/female ratio of about 4:1.[4] Hereditary factors may play a role in the etiology of musician's dystonia, since a positive

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FIGURE 1. Typical patterns of dystonic posture in a pianist, violinist, flautist, and trombone player.

family history of dystonia is considerably more frequent in affected musicians than previously assumed. In a recent study, 28 families of musicians with dystonia were investigated, 14 of these with a previously known family history of dystonia and 14 with no known family history, based on index patients' reports. Neurological examination of family members revealed that 18 of 28 families investigated (64%) were multiplex families: 2 to 4 family members in one, two, or three generations were affected with dystonia. The findings were compatible with autosomal-dominant inheritance in at least 12 families.[5]

The mean age at onset of dystonic symptoms is generally the mid 30s. However, there is a high variability, with the earliest onset of musician's dystonia around age 18 and the oldest onset after age 60 yrs.[6,7] According to epidemiological data, the probability of developing dystonia depends on the instrument played, with guitar players, pianists, and brass instrument players having the highest risk of developing dystonia.[8]

In the same study, a correspondence was seen between the instrument group and the localization of focal dystonia. This is obvious for brass players, who are predominantly suffering from embouchure dystonia. Additionally, among the musicians with unilateral hand dystonia, the correspondence between the instrument group and the localization of focal dystonia was significant. Keyboard musicians (piano, organ, harpsichord) and those with plucked instruments (guitar, electric bass) were primarily affected in the right hand. All these instruments are characterized by a higher workload in the right hand. Correspondingly, bowed string players, who have a higher workload and complexity of movements in the left hand, were predominantly affected in the left hand.[9] Moreover, focal hand dystonia in both hands was seen almost only in those musicians with instruments requiring similar movement patterns in both hands (although with different workload), such as woodwind and keyboard instruments.

Within the string family, musicians who were playing the high string instruments (violin, viola) were more often affected compared to those with low string instruments (cello, double bass). The observed occurrence in high string players was significantly higher than expected according to the overall distribution of high and low string players in German orchestra musicians. The demands on spatial sensorimotor precision required for violin and viola playing are higher than for the low string instruments. Thus, primarily musicians with those instruments requiring highest spatial sensorimotor precision were affected by dystonia (Figure 2).

Furthermore, an association was seen in musicians with unilateral hand dystonia between handedness and the side affected by dystonia. The majority of right-handed patients were affected in the right hand, and the majority of left-handed patients were affected in the left hand. Although the number of left-handed patients in that sample was small, this difference was significant and indicated that the hand with a higher total workload tended to be affected.[10]

The musical genre of the overwhelming majority of patients suffering from focal dystonia is classical music. In contrast to pop or jazz music with improvised structures and great freedom of interpretation, musical constraints are most severe in classical music. The latter requires a maximum of temporal accuracy in the range of milliseconds, which is scrutinized by the performing musician as well as by the audience at any moment of the performance. This, as a consequence, combines the situation of public performance in classical music with a high level of social pressure; the gap between success and failure is minimal in this genre.

Finally, psychological studies suggest an additional source of pressure in musicians who develop dystonia: a pattern of anxiety and extreme perfectionism was observed in musicians that had already been present before onset of dystonia, according to personal recall.[11] This pattern was not observed in healthy

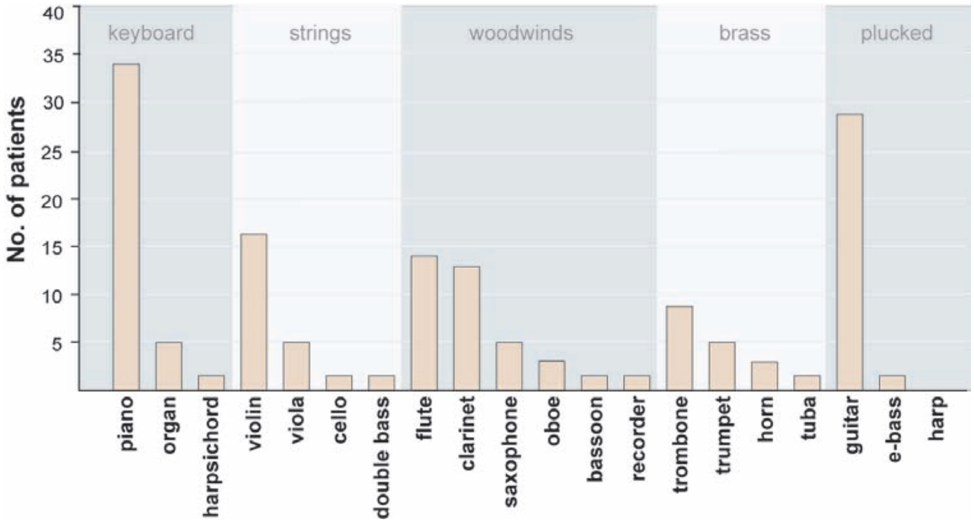

FIGURE 2. Distribution of instruments in 144 patients with musician's dystonia. (Adapted from Jabusch H, Altenmüller E: *Adv Cogn Psychol* 2006; 2:207–220.[9])

musicians. It is possible that this psychological constellation is associated with additional stress and pressure and a more intense working behavior in musicians who develop dystonia.

In summary, the association between highly skilled movement patterns and the development of focal task-specific dystonia is highly suggestive of an environmental contribution from prolonged repetitive use under conditions of high demand on temporal-spatial precision in the context of reward and punishment. However, the observation that the majority of performing artists do not develop musician's dystonia is further suggestive of the above-mentioned assumption of an underlying genetic predisposition.

PATHOPHYSIOLOGICAL FINDINGS IN MUSICIAN'S DYSTONIA

In the preceding section, we identified two important risk factors, i.e., repetitive use and a positive family history. With respect to the pathophysiology of musician's dystonia, these risk factors seem to be related to characteristic adaptations in the central nervous system. Here, most studies reveal abnormalities in three main areas: 1) reduced inhibition in the sensory-motor system, 2) altered sensory perception, and 3) impaired sensorimotor integration.

Lack of Inhibition

A lack of inhibition is a common finding in studies of patients with all forms of dystonia (for a recent review, see Lin and Hallett[12]). Generally, nervous system function requires a constant, subtle balance between excitation and inhibition in neural circuits. This is particularly important in precise and smooth movements of the hand or the embouchure. For example, rapid individualized finger movements in piano playing require selective and specific activation of muscles moving the intended finger in the intended way and inhibiting movements of uninvolved fingers. In patients suffering from hand dystonia, electromyographic recordings revealed abnormally prolonged muscle firing with co-contraction of antagonistic muscles and overflow of activation of inappropriate muscles.[13]

Lack of inhibition is found at multiple levels of the nervous system: At the spinal level, lack of inhibition leads to a reduced reciprocal inhibition of antagonistic muscle groups producing co-contraction, for example, of wrist flexors and extensor muscles, which in turn leads to the feeling of stiffness and immobility and frequently to abnormal postures with predominant flexion of the wrist due to the predominance of the stronger flexor muscles.

Abnormal inhibition also has been demonstrated at the cortical level by measuring intracortical inhibition using non-invasive transcranial magnetic stimulation.[14] Interestingly, at the cortical level, abnormal inhibition frequently is seen in both hemispheres, even in patients with unilateral symptoms, hinting at a more generalized nature of the inhibition deficit.

Finally, lack of inhibition was also seen in complex tasks, such as in affected pianists' scale playing that required movement preparation prior to playing and subsequent sudden movement inhibition when a no-go signal appeared.[15] The ubiquitous demonstration of deficient inhibition is suggestive of a common underlying genetic cause.

Altered Sensory Perception

Several studies have demonstrated that the ability to perceive two stimuli as temporally or spatially separate is impaired in patients with musician's dystonia, be it the fingertips in hand dystonia or the lips in embouchure dystonia. This behavioral deficit is mirrored in findings of the cortical somatosensory representation of fingers or lips. Using evoked potential technology, it was demonstrated that in the somatosensory cortex, the topographical location of sensory inputs from

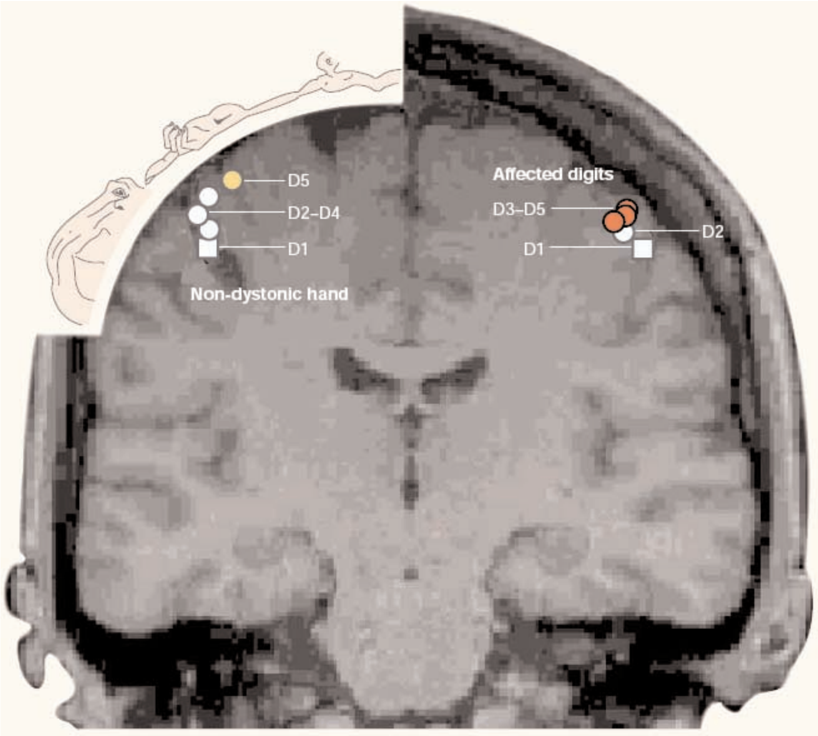

FIGURE 3. Neuronal correlates of dysfunctional plasticity. Fusion of the somatosensory representation of single digits of the hand in a musician suffering from focal dystonia. The best fitting dipoles, explaining the evoked magnetic fields following sensory stimulation of single fingers, are shown projected on the patient's MRI. Whereas for the nonaffected hand, the typical homuncular organization (*see inset*) reveals a distance of about 2.5 cm between the sources for the thumb and the little finger (*white square D1* and *circle D5* on the left), the somatosensory representations of the fingers on the dystonic side are blurred, resulting from a fusion of the neural networks that process incoming sensory stimuli from different fingers (*circles D3-D5* on right). (Modified from Elbert et al.[16] and Münte et al: *Nat Rev Neurosci* 2002; 3:473-478; with permission.[19])

individual fingers overlap more in patients with musician's cramp than in healthy controls[16] (Figure 3). Similarly, lip representation was altered in patients suffering from embouchure dystonia.[17]

Since in healthy musicians an increase of sensory finger representations has been described and interpreted as adaptive plastic changes to conform to the current needs and experiences of the individual,[18] it could be speculated that these changes develop too far in musicians suffering from dystonia, shifting brain plasticity from a benefit to a maladaptation.[19] In this context, it is worth mentioning that local pain and intensified sensory input due to various origins, such as nerve entrapment, trauma, or overuse, have been described as potential triggers of dystonia.[20] In a large group of musicians suffering from focal dystonia, local pain preceded focal dystonia in 9% of the patients.[9]

Interestingly, there are obvious parallels of abnormal cortical processing of sensory information and cortical reorganization in patients with chronic pain and those with focal dystonia.[21] This suggests parallels in the pathophysiological mechanisms in these selected groups of patients. Furthermore, in an animal model of focal dystonia, which was established in overtrained monkeys, repetitive movements induced both types of symptoms, pain syndromes as well as dystonic movements. Brain mapping of the receptive fields demonstrated a distortion of the cortical somatosensory representation,[22] suggesting that overtraining and practice-induced alterations in cortical processing may play a role in focal hand dystonia.

Impaired Sensory-Motor Integration

A clinical sign that emphasizes the important role of sensory-motor integration in the pathophysiology of musician's dystonia is the "sensory trick" phenomenon. This phenomenon is known from patients with cervical dystonia: touching the face contralateral but also ipsilateral to the direction of head rotation can reduce or abolish involuntary muscle activity.[23] In a similar way, musicians suffering from dystonia frequently experience marked improvement of fine motor control when playing with a latex glove or when holding an object, such as a rubber eraser, between the fingers, thus changing the somatosensory input information.[1]

In experimental settings, tonic vibration leads to a worsening of symptoms of musician's dystonia: When transcranial magnetic stimulation was used in conjunction with muscle vibration, the motor-evoked potentials were decreased in agonist muscle and increased in antagonist muscle.[24] These data again suggest an altered central integration of sensory input in musician's cramp, which might be due to the failure of focusing of the proprioceptive input onto the appropriate motor cortical area. Reversal of these effects of sensory-motor disintegration is the rationale of several retraining therapies. Sensory retraining in the form of tactile discrimination practice can alleviate motor symptoms, suggesting that the above-mentioned sensory abnormalities may drive the motor disorder.

Taking the data from epidemiological, genetic and electrophysiological investigations together, a heuristic model of this multifaceted disorder can be proposed: On the basis of a genetic predisposition, prolonged practice, spatiotemporal constraints, social stressors, pain, anxiety, and altered sensory input may trigger the development of a dystonia in a minority of musicians. On the neurophysiological level, the genetic predisposition leads to reduced inhibition, sensory deficits, and deficient sensory-motor integration. In Figure 4, the possible interplay between predisposition and intrinsic and extrinsic triggering factors is displayed.

GENERAL TREATMENT STRATEGIES

Treatment for musician's dystonia is mostly symptomatic and depends on the type of dystonia. Optimally, a multidiscipli-

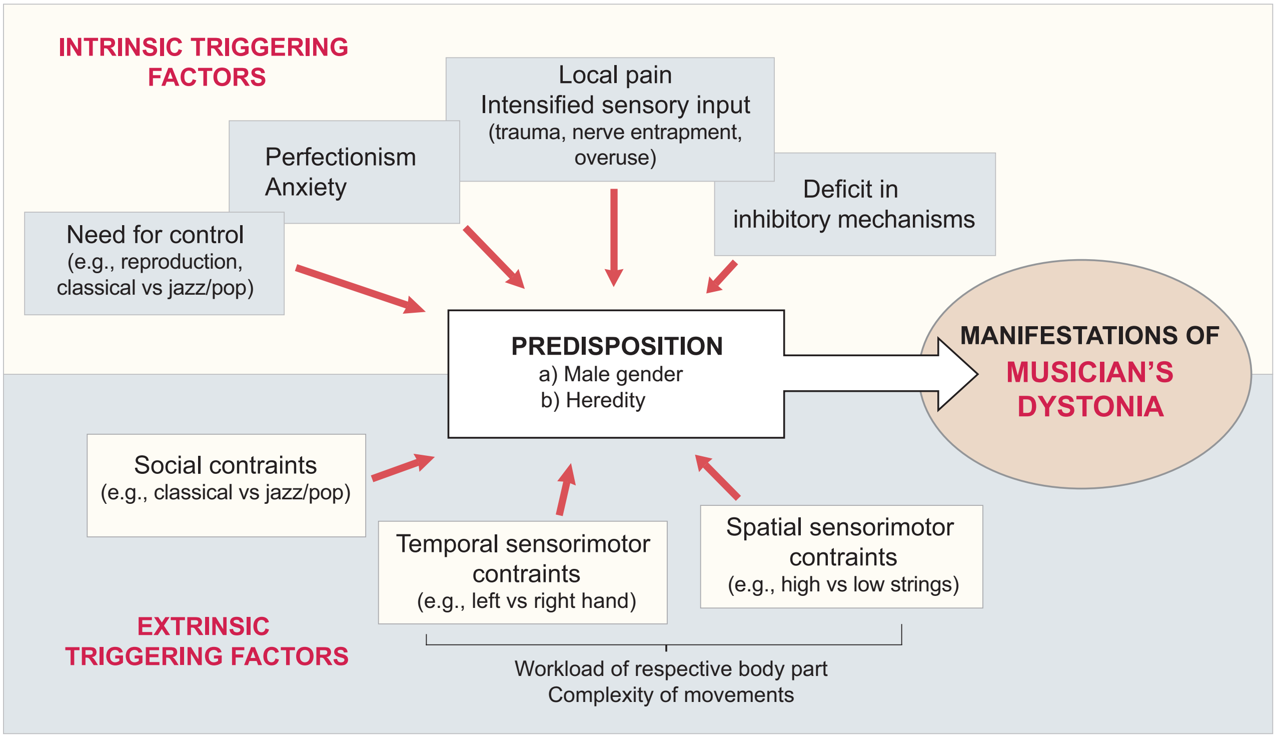

FIGURE 4. The possible interplay between predisposition and intrinsic and extrinsic triggering factors in the manifestation of musician's dystonia. (Adapted from Jabusch H, Altenmüller E: *Adv Cogn Psychol* 2006; 2:207-220.[9])

nary team involving neurologists, psychologists, and physical and occupational therapists should interact. The medical treatments available may be grouped into oral medications, injections with botulinum toxin, and surgical modalities. Furthermore, several retraining techniques are established for rehabilitation of focal hand dystonia.

Various oral medications have been used for the symptomatic treatment of musician's dystonia. These medications affect different neurochemical systems thought to be involved in the pathophysiology of dystonia. The anticholinergic drug trihexyphenidyl has proven to be the most effective agent in musician's dystonia.[25] Occasionally, patients may benefit from baclofen or antiepileptic drugs such as phenytoin or primidone.

Chemical denervation using botulinum toxin has been used for many forms of focal dystonia with considerable success. Botulinum toxin blocks the transmission of nerve impulses to the muscle and weakens the overactive muscles involved. Results in focal hand dystonia and musician's cramp depend on the dystonic pattern, on the injection technique, and on the precise localization of the dystonic muscles. Especially in musician's cramp, focal dystonia usually involves dystonic movements (for example, the involuntary flexion of the middle finger) and compensating movements (for example, the extension of the adjacent index and ring fingers). Erroneous injection into the compensating extensor muscles consequently will lead to a deterioration of symptoms. With correct identification of dystonic movements and application of an electromyographically guided injection technique for proper localization of the respective muscular fascicles, results of botulinum toxin injections are frequently satisfying. However, it should be emphasized that this treatment is symptomatic, requiring the patients to return at regular intervals of about 3 to 5 mos to receive the injections.

In our series, injections of botulinum toxin were applied in 71 musicians suffering from hand dystonia. Fifty-seven percent of patients reported long-term improvement.[9] A meta-analysis of the patients' data revealed that botulinum toxin treatment was successful in those patients in whom primary dystonic movements could be clearly distinguished from secondary compensatory movements. This was difficult when compensatory movements were more pronounced than primary dystonic movements. The best outcome was reported after injections in forearm muscles. Musicians with embouchure dystonia and with dystonia affecting the upper arm and shoulder muscles did not benefit from botulinum toxin treatment.[26]

REHABILITATION OF FOCAL HAND DYSTONIA

In the last decade, based on the hypothesis of aberrant learning and maladaptive plasticity, several treatment protocols were designed to address a reversal of the central nervous system degradation that accounts for one origin and expression of musician's dystonia. These therapies aim at re-differentiation of the disturbed somatotopic hand representations.

The principles of neuroplasticity are most closely aligned to constraint-induced training, immobilization, learning-based sensorimotor training, and retraining at the instrument. In a study of 11 musicians with focal hand dystonia, the unaffected fingers of the dystonic hands were constrained with splints.[27] For 2 wks, 2 to 3 hrs each day, the patients were trained to use the dystonic fingers on fine motor tasks emphasizing instrumental play. When control of the most involved digits improved, then gradually the splints on the less involved digits were removed and practice at the instrument was increased. Eight out of 11 musicians showed an improvement of their motor skills. These results were supported by a brain imaging study, which demonstrated that patients with focal hand dystonia showed some reorganization of receptive fields in the somatosensory cortex after treatment.[28]

Based on the evidence that topographical representation of the hand is abnormal in patients with focal hand dystonia, immobilization of the hand has been proposed as an appropriate treatment at the first signs of dystonia. In individuals without dystonia, limb immobilization leads to a shrinkage of the cortical representation of the immobilized limb. Thus, the objective of immobilization in patients with limb dystonia is to place the affected finger, hand, wrist, and forearm into a static splint to stop all movement in order to decrease the size of the representational area of the involved hand: In another study, 19 patients wore a splint 24 hrs a day for 4 to 6 wks. Subsequently, careful retraining and physiotherapy were applied. The outcome of this study was heterogeneous: in 6 out of 19 patients, there was marked improvement, and in the others, no or only minor improvements could be observed.[29]

Learning-based sensorimotor training (LBST) is another approach to rehabilitation for patients with focal hand dystonia.[30] The objective of LBST is to redefine spatial and temporal processing capacities in the sensory and motor cortices in order to restore task-specific skills, e.g., writing. Different exercise strategies address subtasks of the target tasks, emphasizing different aspects of sensory feedback (e.g., somatosensation, proprioception, kinesthesia, haptics). The patients initially make relatively simple distinctions about relatively large objects, with task difficulty increasing as each patient masters progressively more difficult distinctions. One set of tasks includes reading Braille or embossed letters, making roughness estimations, and performing grating orientation discriminations. A series of haptic tasks requires the subjects to manipulate and identify surface features and shapes of three-dimensional objects held in their hands.

This training protocol has been tested in a series of patients with focal hand dystonia, including patients suffering from musician's dystonia (for a review, see Byl and McKenzie[31]). In the first study that focused on this training protocol, all 12 trained patients showed improvements in performance ability on a variety of tests of sensory discrimination, fine motor accuracy, and speed, strength, flexibility, and functional independence. Motor control improved to an average of 70 to 94% of normal. All but 3 patients returned to their usual work.

Pedagogical retraining has been applied in patients with all forms of musician's dystonia. Studies utilizing these techniques have comprised a variety of behavioral approaches under the supervision of instructors and included elements based on the following principles (e.g., Boullet[32]):

1) Movements of affected body parts were limited to a level of tempo and force at which the dystonic movement would not occur;
2) Compensatory movements (e.g., of adjacent fingers) were avoided, partially under the application of splints;
3) Instant visual feedback with mirrors or monitors helped patients to recognize dystonic and nondystonic movements;
4) Body awareness techniques (e.g., Feldenkrais®) were applied to increase the patient's perception of nondystonic movements.

In common to all these therapies is the "long-term" approach. For example, in a large series of 145 musicians, an average rehabilitation time of 24 mos, after which 35 musicians returned to playing in public, was reported.[33]

In our series of musicians suffering from focal dystonia, pedagogical retraining was applied in 24 patients according to the aforementioned guidelines. Twelve patients (50%) experienced improvement. Patients reporting an improvement had undergone this treatment for an average of 28 mos (range, 3 to 72). Two patients with embouchure dystonia took part in pedagogical retraining and reported no improvement.[25]

CONCLUSION AND CONSEQUENCES

In summary, overuse and intense working behavior, spatiotemporal constraints, and special psychological conditions, including anxiety and extreme perfectionism, may trigger the onset of musician's focal dystonia on the basis of a given predisposition that may be genetically determined. It should be emphasized that the emotional aspects of music performance, especifically the enormous professional pressure, substantially contribute to stress-induced processes that may foster consolidation of dystonic movements. In part, the unyielding reward and punishment frame in the classical music performance scene provides a fertile ground for these stresses in musicians. This in turn could explain why, for example, improvisational jazz musicians are much less likely to develop musician's dystonia. Here, as in many other music cultures, reproduction of the precise musical notation plays only a minor role. Learning is frequently based on imitation, and movements frequently can be selected deliberately, obeying the individual's anatomical prerequisites.

Preventing dystonia is important, since successful treatment is still a challenge. Many of the available medical approaches are only moderately effective, and other options have yet to be developed. 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.

Concerning prevention, exaggerated perfectionism and anxiety as triggering factors should be addressed in the education of musicians. This has to be started early. From the first lesson on, music educators should strive to create a friendly, supportive atmosphere focusing on creativity, curiosity, and playful experiences in the world of sounds. It is not by chance that we commonly speak of "playing an instrument" and not of "working an instrument." Of course, structured, goal-directed learning is a prerequisite of musical mastery. Here, reasonable practice schedules, economic technique, prevention of overuse and pain, mental practice, variations of movements patterns, maintenance of motivation and avoidance of mechanical repetitions and frustration, healthy living habits, warm-ups and cool-down exercises, regular physical exercise, and sufficient breaks and sleep are the cornerstones of healthy musical practice.

Finally, the role of the societal constraints should not be neglected. In the last few decades, the classical music sector was inundated by CD-recordings of peers in the fields. Many of these recordings can be regarded as "laboratory music," composed in the tranquility of the studios with the help of tone engineers and electronic "re-mastering." These recordings are considered a "gold standard." They frequently create unrealistic expectations in listeners and interpreters, adding stress to the performers.

In addition, our classical music culture reflects the general societal pressures of the developed countries. Highest precision and efficiency are the demands to which we are all subjected. In music, this often creates an attitude of great artistic accomplishment, which, however, frequently is not nurtured by a sufficient personal expression of emotional experience. The latter, of course, has to be collected somewhere outside the practice room. As a consequence, we therefore should correct our expectations and listening habits, replacing the fascination of mere perfection and virtuosity with the joy of emotional communication shared with the audience and musicians.

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