Focal hand dystonia in musicians: phenomenology, etiology, and psychological trigger factors

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ABSTRACT: Musician's dystonia is a task-specific movement disorder, which manifests itself as a loss of voluntary motor control in extensively trained movements. In many cases, the disorder terminates the careers of affected musicians. Approximately 1% of all professional musicians are affected. In the past, focal dystonia (FD) was classified as a psychological disorder. Over time, the problem was classified as a neurological problem. Although the specific pathophysiology of the disorder is still unclear, it appears the etiology is multifactorial. While there may be a genetic history, neurophysiological, physical, and environmental factors, trauma and stress contribute to the phenotypic development of FD.

Are there potentially psychological factors that also contribute to the manifestation of the disorder? This manuscript analyzes the evidence supporting the potential contribution of the emotional brain systems in the etiology of focal hand dystonia in musicians. In addition, the psychological findings from a large descriptive study comparing healthy musicians, musicians with dystonia, and musicians with chronic pain (CP) are presented. Information about psychogenic characteristics might be used to modify intervention strategies and music instruction to reduce the incidence of musician's dystonia.

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WHAT IS MUSICIAN'S DYSTONIA?

Focal dystonia in musicians, also known as musician's cramp or musician's dystonia, is a task-specific movement disorder, which 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, FD is highly disabling and in many cases terminates musical careers.[1]

Musician's dystonia may be classified according to the task specifically involved. For example, embouchure dystonia may affect coordination of lips, tongue, facial and cervical muscles and breathing in brass and wind players, whereas pianist's cramp and violinist's cramp affect the control of hand movements. According to recent estimates, 1% of all professional musicians are affected.[1] In contrast, in the general population, prevalence of FDs, including writer's cramp, blepharospasm, and cervical dystonia, is estimated as 29.5/100,000 people in the United States and 6.1/100,000 people in Japan.[2,3] In comparison to other activities producing dystonic movements, such as writing, playing golf (the "yips") or dart ("dartism"), classical musicians are at the highest risk of developing FD.

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, possibly due to the degradation of sensory receptive fields in the somatosensory cortex. 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 movements in musicians are shown.

WHAT CAUSES MUSICIAN'S DYSTONIA?

Until today, the etiology of musician's dystonia has not been completely understood, but it is probably multifactorial. It may develop in individuals with a

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

genetic history of dystonia[4] or as a consequence of alterations in the basal ganglia circuitry.[5,6] Most studies of musician's dystonia reveal abnormalities in three main areas: a) reduced inhibition in the motor system at cortical, subcortical, and spinal levels b) reduced sensory perception and integration; and c) impaired sensorimotor integration. The latter changes are mainly believed to originate from dysfunctional brain plasticity. Such a dysfunctional plasticity has been described in the sensory thalamus.[7] Disorganized motor somatotopy could be found in the putamen of patients suffering from writers' cramp.[8] Finally, there is growing evidence for abnormal cortical processing of sensory information as well as degraded representation of motor functions in patients with FD. In monkeys, repetitive movements induced symptoms of focal hand dystonia and a distortion of the cortical somatosensory representation,[9] suggesting that practice-induced alterations in cortical processing may play a role in focal hand dystonia.

Indeed, using somatosensory-evoked potential technology, it was demonstrated that in the somatosensory cortex the topographical location of sensory inputs from individual fingers overlap more in patients with writer's cramp than in healthy controls.[10] Similar observations have been made using magnetoencephalography.[11] Elbert et al.[12] showed that there is an overlap of the representational zones of the digits in the primary somatosensory cortex for the affected hand of musicians with dystonia compared with the representations of the digits in nonmusician control subjects.

In healthy musicians, an increase in sensory finger representations has been described and interpreted as adaptive plastic changes to conform to the current needs and experiences of the individual.[13] It could be speculated that these changes develop too far in musicians suffering from dystonia, shifting brain plasticity from a beneficial to a mal-adaptation level.[14] On the behavioral level, this notion is supported by the fact that healthy musicians have lowered two-point discrimination thresholds compared with nonmusician controls,[15] whereas two point-discrimination thresholds are increased in musicians suffering from dystonia.

Another clinical sign, which emphasizes the important role of sensorimotor 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.[16] 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, for example a rubber gum, between the fingers, thus changing the somatosensory input information.[17]

Interestingly, in musicians with hand dystonia, an association exists between the instrument group and the localization of FD. In instruments with different work loads, different complexity of movements, or different temporospatial precision for both hands, FD appears more often in the more heavily used hand. Keyboard musicians (piano, organ, harpsichord) and

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those with plucked instruments (guitar, e-bass) are primarily affected in the right hand. All these instruments are characterized by a higher workload in the right hand. Additionally, guitar playing requires higher temporospatial precision in the right hand compared with that in the left hand. Bowed string players who have a higher workload and complexity of movements in the left hand are predominantly affected in the left hand.[1]

In summary, in view of neurophysiological and epidemiological findings in musicians with FD, several predisposing factors have been identified, such as male gender,[18,19] as well as a positive family history,[20] which might constitute a particular vulnerability or predisposition to musician's dystonia. Additional extrinsic and intrinsic factors may trigger the manifestation of musician's dystonia on the basis of a given predisposition. Intrinsic triggering factors are, for example, physical disorders resulting in local pain and/or intensified somatosensory input. Traumatic injuries, nerve-entrapment, or overuse injury may also lead to a degradation of sensorimotor representations at several levels of the sensorimotor circuits.[21]

Extrinsic triggering factors, according to epidemiological findings, [22–24] are spatial and temporal sensorimotor constraints as well as musical and social constraints typical of the performance situation in classical music. The question remains open, whether psychological factors contribute to the manifestation of the disorder. In this manuscript, we review the literature on the potential role of psychological factors in musician's dystonia.

ARE PSYCHOLOGICAL FACTORS INFLUENCING AND TRIGGERING DYSTONIA?

For many decades, psychological factors were believed to be essential for the development of task-specific FDs and were overemphasized in the assumed pathomechanism. Gowers subsumed writer's cramp and related cramps under the term "occupational neuroses."[25] At that time, "neurosis" was used as a term for a disease when a physical origin was assumed, but a clear cause could not be described. As a consequence of the terminology, a psychogenic origin was assumed (for example: Ref. 26) until in 1982 Sheehy and Marsden postulated a neurological pathomechanism, because psychiatric investigation of 29 patients with writer's cramps and related cramps (four with typist's cramps, one with pianist's cramp) did not reveal a higher incidence of psychiatric disturbance than that in a normal population, as judged by formal Present State Examination.[27] They stated that occupational cramps are symptoms of a physical illness, and they used the term "FD."[28] Subsequently, the neuropathophysiology of FD was investigated systematically and yielded the above briefly reviewed results.

At the same time, a number of studies pointed at psychological findings in patients with writer's cramp and emphasized a psychological and psychosocial part in the etiology of the disorder.[29,30] Windgassen and Ludolph found depressive, anancastic, sensitive and hysteric traits in 22 patients with writer's cramp.[31] The Giessen Test showed an inclination toward compulsive traits in these patients. Anxiety was occasionally observed in patients with writer's cramp.[32–34] It was discussed controversially whether these special psychological conditions are part of the etiology of FD[35] or whether they demonstrate secondary psychoreactive processes.[36] In contrast, in a study with 22 patients suffering from writer's cramp, no significant differences were found compared to a matched normal control group using the Crown-Crisp Experiential Index.[37] This questionnaire assessed traits and symptoms relevant to neurotic illness and contained three anxiety subscales (free-floating anxiety, phobic anxiety, somatic anxiety). Gündel et al.[38] compared psychiatric comorbidity in patients suffering from spasmodic dysphonia, a FD of the voice impairing speaking as well as singing, and compared it to patients with vocal fold paresis. Surprisingly, 41.7% of patients with spasmodic dysphonia, compared with 19.5% of patients with vocal fold paresis, had a current psychiatric diagnosis. Depression, anxiety, and adjustment disorders were the prevailing symptoms in both groups.

Recent studies have revealed additional, though subtle, behavioral dysfunctions in patients with FD. Bugalho and colleagues[39] demonstrated increased perseveration in the Wisconsin Card Sorting Test, indicative of defective set shifting, and higher intensity of obsessive compulsive disorder in patients with FD. These findings may reflect a pattern of complex neurophysiological dysfunction involving dorsolateral, orbitofrontal, and motor frontostriatal circuits. In fact, in an electroencephalograph study in pianists, suffering from dystonia, electrophysiological correlates of such a defective set shifting could be convincingly demonstrated. In comparison with healthy pianists, those with dystonia could not appropriately deactivate the preparatory cortical activation in anticipation of playing a C-major scale, when a No-go condition was introduced requiring them not to play the scale.[40]

THE HANNOVER STUDY: PSYCHOLOGICAL FINDINGS IN MUSICIAN'S DYSTONIA

All the above mentioned psychological studies were undertaken in nonmusician patients suffering

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from various forms of FDs. It is intriguing that new findings point at a different origin of musician's dystonia compared with other FDs.[41] In general, it seems that musician's dystonia is more linked to overuse and overpractice compared with other hand dystonias.

In this context it is worth mentioning that musician's dystonia emerged relatively recently during the early romantic period, when eminent virtuosos such as Paganini or Franz Liszt pushed technical demands to new limits. The first proven records of this condition date back only to 1830, when the ambitious pianist and composer Robert Schumann developed a pianist's cramp, deteriorating the fine motor control of the middle finger of his right hand.[42] As a supposedly precipitating factor, Robert Schumann had dramatically increased his piano practice schedules up to 7 hrs a day to attain the pianistic technique necessary to compete with the eminent virtuosos of the early romantic period. However, besides prolonged practice time and musculoskeletal overuse, psychosocial stressors and personality factors may have contributed to his disorder. According to his diaries and the written testimonials of his friends, Schumann tended to compulsive working behavior, to harsh self-criticism, to anxiety and depression, and to frequent and excessive alcohol consumption. Already at this time, his physicians discussed whether he presented a psychogenic or a neurological condition.[43]

The Hannover study on psychological conditions in musicians

We have investigated personality structures in musicians with FD.[42,43] This study was designed to examine psychological conditions in musicians with FD and to compare them with healthy musicians and those suffering from CP syndromes. The latter group was added to detect unspecific secondary psychological reactions in diseased musicians. Based on clinical observations, the underlying hypothesis was that a) musicians with dystonia more often suffer from social phobias and specific phobias and display more perfectionistic tendencies than healthy musicians and those with CP and that b) these psychological conditions were already present before onset of dystonia.

Two groups of patients were included: One sample comprised 20 professional musicians who had been diagnosed with task-related FDs. Three out of 20 patients were brass players with embouchure dystonias; the other 17 patients suffered from hand dystonias, which presented in the typical manner as painless cramping of one or more fingers while the patients played their instruments. The individual durations of the disorder were between 3 and 12 years (6.9 ± 2.6 years [mean ± SD]) at the time of the study. Twelve of the patients noticed additional dystonic movement patterns in other activities such as writing (five patients), on the computer keyboard (four patients), or everyday activities; the onset of these additional symptoms took place after the onset of musicians cramps. Patients with other neurological disorders or secondary dystonias were excluded from the study.

The second sample of patients comprised 20 professional musicians suffering from CP syndromes related to playing their instruments. Pain was localized in the hand and/or arm region. The durations of CP ranged from 6 months to 30 years (4.9 ± 6.9 years) at the time of the study.

A third group consisting of 30 professional musicians was recruited as healthy controls. They were working in German orchestras, Colleges of Music, and as freelance professional musicians. By means of questionnaires, they were asked if they were suffering from any kind of ill health, especially from movement disorders or CP. Musicians with any somatic complaints or neurological or psychiatric diseases were excluded from the normal control sample. The samples of patients and the control group were similar in age. However, both patient groups differed markedly in gender distributions. There was a preponderance of males (16 males, four females) in the sample of musicians with FDs, whereas females were predominant in the group with CP (five males, 15 females). The respective gender distributions are in keeping with reports of other authors who found more male than female musicians suffering from FD[46] and a preponderance of females in patients with CP.[47] Gender distribution was almost balanced in the sample of normal controls with 16 males and 14 females. It should be mentioned that the process of finding subjects for the healthy control group was protracted due to the high number of symptomatic musicians among the professionals. This observation coincides with the results of Fishbein et al., who found that 76% of orchestra musicians reported at least one medical problem that was severe in terms of its effect on performance.[49]

Methods of the study

The assessment of psychological conditions was based on self-estimation using the following German inventories: the revised version of the Freiburg Personality Inventory (FPI-R)[49] is a personality questionnaire, which consists of 12 bipolar subscales being life satisfaction, social orientation, achievement orientation, inhibitedness, irritability, aggressiveness, strain, somatic complaints, health concern, frankness, extraversion, and emotionality. Additionally, the Questionnaire for Competence and Control Orientations (QCC)[50] was used to investigate features such as self-concept of abilities, internal control

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orientation, others control orientation, and chance control orientation (primary subscales). In order to assess perfectionistic tendencies, a questionnaire was developed with five items on a six-point scale. Finally, six questions were designed focusing on anxiety disorders subgrouped as considerable stage fright, panic attacks, free-floating anxiety, agoraphobia, social phobia, and specific phobias such as acrophobia, claustrophobia, and so on. The respective symptoms were explained to the subjects, and they were asked whether they felt that these anxieties were present or absent. To investigate whether certain psychological conditions were already present before the onset of FD or CP in the patient groups, a second step was added: subjects were asked to decide whether the particular anxieties had been present or absent before onset of their disorders, and whether their attitudes concerning perfectionism were the same or different before and after onset.

Informed consent was obtained from all subjects. They were instructed to fill out the questionnaires without the presence of other persons. Distribution and collection of the questionnaires were done by mailing, and precise written explanations were attached. Questionnaires that were not filled out properly were excluded from the study. FPI-R and QCC questionnaires with incomplete data were treated following the rules for missing data treatment recommended by the authors of the questionnaires. Data of subjects who showed dissimulative tendencies in the frankness subscale of the FPI-R were not included in the study.

Statistical analyses were performed (STATISTICA Version 5.5 A, StatSoft Inc.): Differences between groups and gender concerning FPI-R, QCC (primary subscales), and the perfectionism scale were calculated using multivariate analyses of variances (MANOVA). Statistical analyses of the differences in anxiety disorders between the groups were performed using chi-square test (χ^2). Two-tailed Fisher's exact tests were applied when χ^2-tests were not possible due to methodological requirements. χ^2-tests/Fisher's exact tests were performed separately for evaluation of the differences in the anxiety subscales before and after onset of the disorders.

Results of the study

Anxieties were present more often in both patient groups (FD and CP) than in normal controls (FD: $\chi^2 = 5.33$, $p < 0.05$; CP: $\chi^2 = 9.07$, $p < 0.01$) as shown in Figure 2.

Patients were asked to specify whether their tendencies concerning anxieties had been the same or different before and after the onset of their diseases. In this respect, conclusive statements could be obtained in all patients with FD and in 19 out of 20 patients with CP. χ^2-tests/Fisher's exact tests were performed separately for evaluation of the differences in the anxiety subscales before and after onset of the disorders. Significantly, in both patient groups anxieties had been more often present already before onset of the disorders compared with healthy musicians (FD: $\chi^2 = 5.33$, $p < 0.05$; CP: $\chi^2 = 6.38$, $p < 0.05$). A closer look at the different anxiety subscales showed that musicians with FD suffered

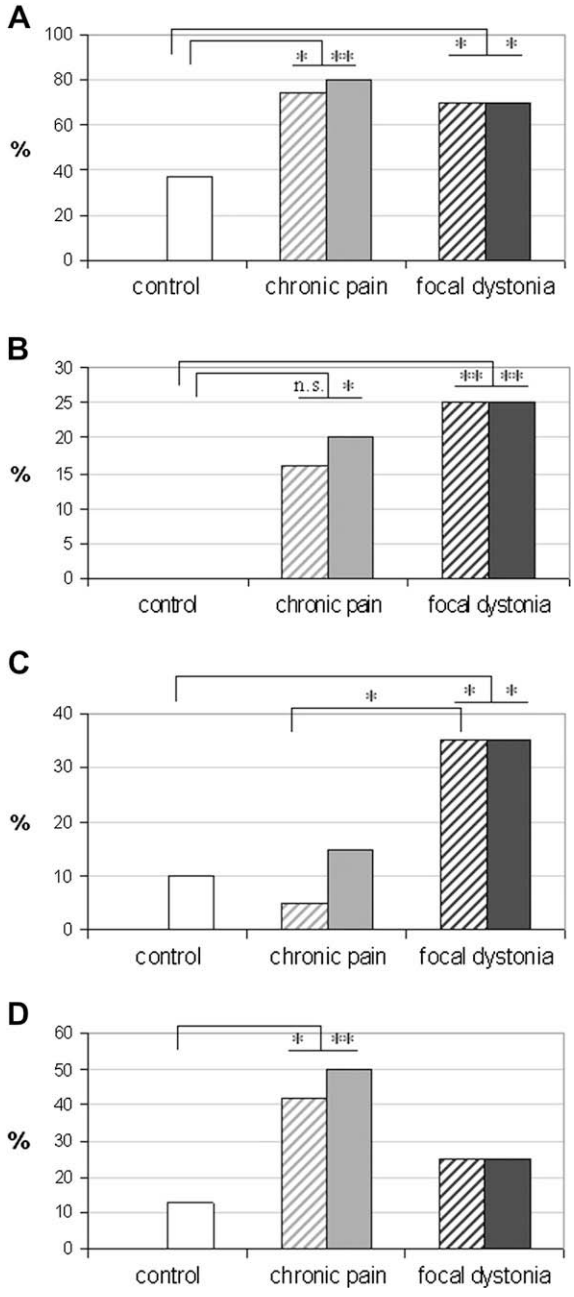

FIGURE 2. *Findings of the anxiety questionnaire of controls and musicians with chronic pain and those with focal dystonia: percentage of subjects (of respective groups) who reported any anxiety disorder (A), social phobia (B), specific phobias (C), free-floating anxiety (D). Hatched bars: percentage with anxiety before onset of disorder. Filled bars: percentage with anxiety after onset of disorder (*$p < 0.05$, **$p < 0.01$, n.s.: not significant).*

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from social phobias remarkably more often than healthy musicians (two-tailed Fisher's exact test (Fisher's), $p < 0.01$) which had already been the case before onset of FD (Fisher's, $p < 0.01$). Patients with CP did not report more social phobias present already before onset of CP compared with healthy musicians. However, this comparison revealed a significant difference (Fisher's, $p < 0.05$) after development of CP due to an increase in social phobias in these patients after onset. Only patients with FD significantly more often reported specific phobias such as acrophobia, claustrophobia, and so on, compared with healthy musicians ($\chi^2 = 4.69$, $p < 0.05$). These specific phobias had already been present before they developed FD. Musicians who later developed FD suffered markedly more often from specific phobias than musicians who later developed CP (Fisher's, $p < 0.05$). The opposite result was found in the subscale free-floating anxiety: only patients with CP significantly more often reported free-floating anxiety than normal controls. This was the case before ($\chi^2 = 5.21$, $p < 0.05$) and after onset of CP ($\chi^2 = 8.0$, $p < 0.01$). In the subscales agoraphobia, panic attacks, and remarkable stage fright, no differences were observed between any patient group and healthy musicians. No significant differences were found in any of the anxiety subscales between male and female subjects.

Using 3 (group affiliation) × 2 (gender) MANOVA for the perfectionism questionnaire and all subscales of the QCC and FPI-R, significant differences were seen between the groups FD, CP, and control (Wilks' Lambda = 0.291; $p < 0.01$) and between male and female subjects (Wilks' Lambda = 0.552; $p < 0.05$). No interaction was found between group affiliation and gender. Follow-up ANOVA revealed significant differences between the groups for the variable perfectionism ($F_{(2,58)} = 3.28$; $p < 0.05$). Patients with FD attained the highest scores (24.3 ± 4.8) in perfectionism compared with controls who reached 21.9 ± 3.3 ($p < 0.05$; contrast analysis) on a scale ranging from minimum =5 to maximum =30. Figure 3 shows the findings of the perfectionism scale in the different groups.

Patients suffering from CP syndromes achieved [Q2] scores (23.4 ± 4.0) tendentially in between the healthy musicians and those with FD. Differences in both directions (CP vs. FD; CP vs. control) were nonsignificant.

Patients were asked to specify whether their attitudes concerning perfectionism were the same or different before and after the onset of their disorders. All patients with FD and 19 out of 20 with CP were able to give information about their attitudes toward perfectionism before onset of the disorders. All 20 patients with FD (100%) estimated their attitudes concerning perfectionism as the same before and after onset. In one patient with CP, perfectionist tendencies had increased after onset. Eighteen CP patients (94.7%) had experienced no change in this parameter. No difference was seen between perfectionistic tendencies of male and female subjects. Cronbach's alpha was 0.72, displaying an adequate internal consistency of the scale.

The QCC did not reveal any statistically significant differences between the groups (FD, CP, control) nor between male and female subjects. In the FPI (Figure 4), follow-up ANOVA revealed significant differences between the groups for the subscales somatic complaints ($F_{(2,58)} = 10.16$; $p < 0.001$) and emotionality ($F_{(2,58)} = 3.55$; $p < 0.05$). Both patient groups showed more somatic complaints than healthy musicians (FD: $p = 0.01$; CP: $p < 0.001$). Musicians suffering from CP reached higher scores in the subscale of emotionality compared with healthy musicians ($p < 0.05$).

Gender differences were observed in two subscales of the FPI-R: in the social orientation subscale, male musicians had higher scores (6.3 ± 1.4) than female musicians, who reached 5.1 ± 1.5 ($p < 0.01$, $F_{(1,58)} = 11.07$). Similarly, male musicians had more somatic complaints (5.0 ± 1.8) than female musicians who had 4.0 ± 1.8 ($p = 0.01$, $F_{(1,58)} = 6.9$).

PERFECTIONISM AND ANXIETY IN MUSICIAN'S DYSTONIA: PREEXISTENT OR PSYCHOREACTIVE?

The results demonstrated that psychological conditions in musicians with FD differed from those of healthy musicians. Anxiety disorders, and above all, social phobias, and specific phobias occurred more often in musicians with dystonia. Additionally, musicians with FD were found to have highly perfectionist tendencies. Musicians with CP also showed different psychological patterns from those of healthy musicians. They also more often reported anxiety, above all, free-floating anxiety. In the perfectionism scale, they recorded scores tendentially in between those of the control group and the musicians with dystonia. In the FPI-R, both patient groups showed more somatic complaints than healthy musicians. Furthermore, musicians with CP turned out to be more emotional than controls. Taken together, a certain overlap was observed between the psychological conditions of musicians with FD and those suffering from CP. However, musicians with dystonia showed a pattern of exaggerated perfectionism, social phobia, and specific phobias that was not seen in healthy musicians or those with CP.

The question arises whether anxiety was present before the playing-related disorders in both patient groups and whether perfectionism was present in musicians before they developed FD. Alternatively, both psychological features might reflect psychoreactive symptoms as a consequence of playing-related

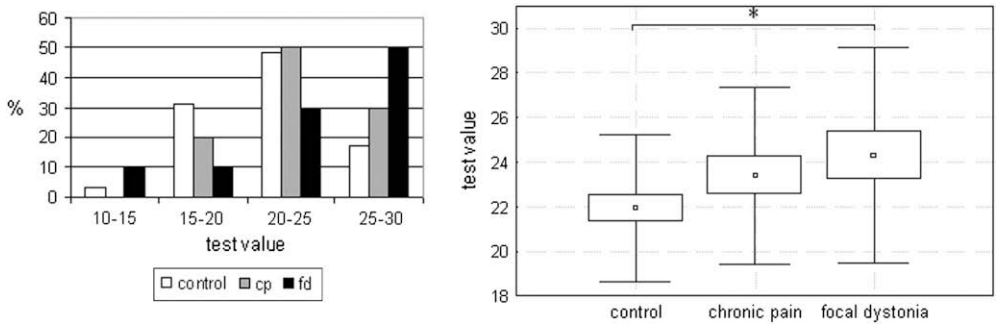

FIGURE 3. *Findings of the perfectionism scale of controls and musicians with chronic pain and those with focal dystonia. Left side: frequency distribution: percentage of subjects (of respective groups) who reached different perfectionism levels. Right side: results of all groups in the perfectionism scale: small boxes: mean values/large boxes: standard errors/whiskers: standard deviations (*$p < 0.05$).*

disorders. We hypothesized that these psychological conditions were already present in musicians before the onset of playing-related disorders. A prospective study with the particular aim of assessing psychological conditions in musicians before the development of FD was not realizable due to a prevalence of FD between 1:200 and 1:500 in professional musicians with the consequence of a low incidence.[1] A retrospective analysis was the only realizable approach. A retrospective inquiry was undertaken based on personal recollection. Under the given circumstances, this procedure was the only practical approach to obtain the desired information. As a limitation of the study, such retrospective reports are subject to bias and not always reliable. However, in view of the unambiguity and consistency of the reports of all patients with dystonia, we postulate that perfectionism and anxiety have been preexistent and were not psychoreactive phenomena.

In view of the findings for musicians with FD, the question arises whether and in which way the described psychological conditions with anxiety and exaggerated perfectionism might be involved in the etiology of FD in musicians.

THE POSSIBLE ROLE OF PERFECTIONISM AND ANXIETY DURING ONSET OF MUSICIAN'S DYSTONIA

Phenomenologically, musician's dystonias are dysfunctional movement patterns almost inextinguishably fixed in the procedural motor memory. How can the observed psychological characteristics in musicians with dystonia be interpreted in context with the neurobiological basis of memory systems?

We suggest that specific affective conditions of making music underlie the interaction between psychological phenomena and motor memory. Music performance is strongly linked to the emotions in a way that is not comparable to any other activity in human life. On one hand, music is the ''language of emotions'' and is able to communicate positive or negative feelings. On the other hand, instrumental music is based on extreme spatiotemporal sensorimotor precision, which can be scrutinized by both the musician and the audience. Therefore, many musicians experience strong and contrary feelings, with the joy of performing alongside the fear of playing

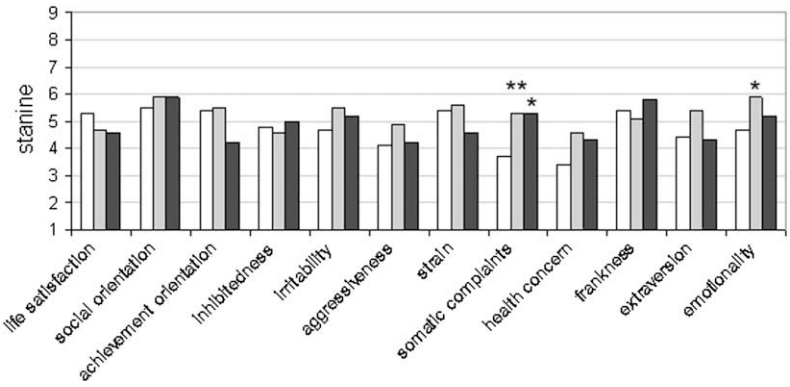

FIGURE 4. *Results of the FPI-R subscales: means of controls and musicians with chronic pain and those with focal dystonia. Stanine test values scaled from one (minimally pronounced) to nine (maximally pronounced). (*$p < 0.05$ compared to control; **$p < 0.01$ compared to control).*

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wrong notes or the fear of failure, which reflects the extremely strict system of reward and punishment in professional musicianship. This double link to the emotions is a unique characteristic of making music.[51]

We suggest that during onset of musician's dystonia, this link between music performance and strong emotions may play a role in the establishment of dystonic movement patterns in patients with perfectionism and anxiety. It is possible that emotionally induced motor memory consolidation may facilitate the onset of dystonia in the subgroup of patients with these psychological conditions. The process might start with the occurrence of a dystonic movement for which the cause is unknown. Musicians with an inclination toward anxiety and extreme perfectionism may emphasize the disturbing and threatening element in the occurred wrong movement. This psychological stress might induce a cascade of emotionally induced memory consolidation, which has previously been described and applied to different forms of memory and which mainly relies on noradrenergic activation of the basolateral amygdala (BLA).[52] The primary motor cortex, which is an essential locus of representation of digital motor sequences, receives a BLA projection.[53] Thus, it may be assumed that consolidation of early dystonic movements as dysfunctional motor programs may be facilitated by a BLA-mediated process in the primary motor cortex.

Undoubtedly, the above outlined scenario constitutes only an epiphenomenon in a subgroup of patients with the described psychological conditions. Further support for a participation of limbic circuits in the development of FD comes from the phenomenologies of related FDs, for example, in calligraphists, telegraphers, money counters, golfers etc. Two common features can be observed: 1) the necessity for high precision and 2) a strong emotional component. The first is related to the nature of the respective activities, the latter originates in the fact that a strict system of reward and punishment underlies these activities. In sports, this is provided by victory and defeat with the resulting advantages and disadvantages; the other activities are performed in professional contexts, which means that people's income depends on the quality of their work. One can speculate that in all of these activities, an inclination toward anxiety and extreme perfectionism may also foster consolidation of dystonic movement patterns.

PARALLELS BETWEEN MUSICIANS WITH FOCAL DYSTONIA AND THOSE WITH CHRONIC PAIN

The findings in musicians with CP harmonize with other reports on CP patients. The association between anxiety and CP has often been described for CP syndromes in different sites (for example Ref. 54). In particular, social phobia has been found to be related to physically unexplained CP in a study with 130 patients. Additionally, in keeping with the findings of the Hannover study, agoraphobia was found minimally pronounced in CP patients.[55] Free-floating anxiety, which was more often present in musicians with CP of this study, turned out to be a predictor for early retirement in patients with CP.[56]

The emotionality subscale (FPI-R), in which CP patients had higher scores than the other groups, recorded substantial components of neuroticism, which was found to be more pronounced in patients with CP in other studies.[57]

Finally, the FPI-R showed more somatic complaints and higher levels of health concern in both patient groups than in normal controls. It should be mentioned that the questions for the somatic complaints subscale did not focus on the diseases of the patients specifically, neither on FD, nor on CP syndromes. The similarity in the findings of elevated somatic complaint scores and health concern scores of the FPI-R in both patient groups might be based on a generally more intense perception of somatic sensations in diseased musicians and a more health-focused orientation as an unspecific reaction to the disorders.

A certain overlap was seen between the psychological conditions in musicians with FD and those suffering from CP. In some of the musicians with dystonia, intensified sensory input, as in CP syndromes, trauma, or nerve entrapment, preceded FD.[21] This CP−FD sequence means that some of the musicians with dystonia suffered from CP before they developed FD. That implies a certain overlap of both patient groups. However, among the sample of musicians with dystonia of the Hannover study, CP had preceded FD in only one patient. Thus, the CP−FD−sequence cannot explain the overlap in psychological findings of both groups.

The amount of perfectionism in musicians with CP was ranked in between that in the control group and the group with FD. The combination of a tendency toward perfectionism and anxiety—either social phobia or free-floating anxiety—might promote a certain working behavior, which might be a triggering factor for both disorders, FD and CP syndromes.

CONCLUSION AND CONSEQUENCES

In summary, special psychological conditions including anxiety and extreme perfectionism were seen in musicians with dystonia. We suggest that these conditions may facilitate the onset of the disorder. It should be emphasized that the emotional aspects of music performance, specifically the enormous professional pressure, substantially contribute to stress-

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induced processes that may foster consolidation of dystonic movements. In part, the unyielding reward and punishment frame in the reproductive classical music scene provides a fertile ground for these stresses in musicians. This in turn could explain why, for example, improvising 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.

In Figure 5 we propose a model, specifying the role of anxiety and perfectionism in triggering musician's dystonia. Furthermore, the possible co-action between predisposition and intrinsic and extrinsic triggering factors is displayed.

With this model, we now have means at hand contributing to the prevention of musician's dystonia. 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 (for a review see Ref. 58).

Concerning prevention, exaggerated perfectionism and anxiety as triggering factors should be addressed in the education of musicians. This has to be started at early infancy. 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 movement patterns, maintenance of motivation and avoidance of mechanical repetitions and frustration, healthy living habits, warm-ups and cool-down exercises, regular physical exercise, sufficient breaks, and sleep are the cornerstones of healthy musical practice.

Finally, the role of societal constraints should not be neglected. In the last decades, the classical music sector was inundated by CD recordings of peers in the fields. Frequently, 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 as the "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 we all are subjected to. In music, this frequently creates an attitude of great artistic accomplishment, which, however, frequently is not nurtured by a sufficient personal expression of emotional experience. The

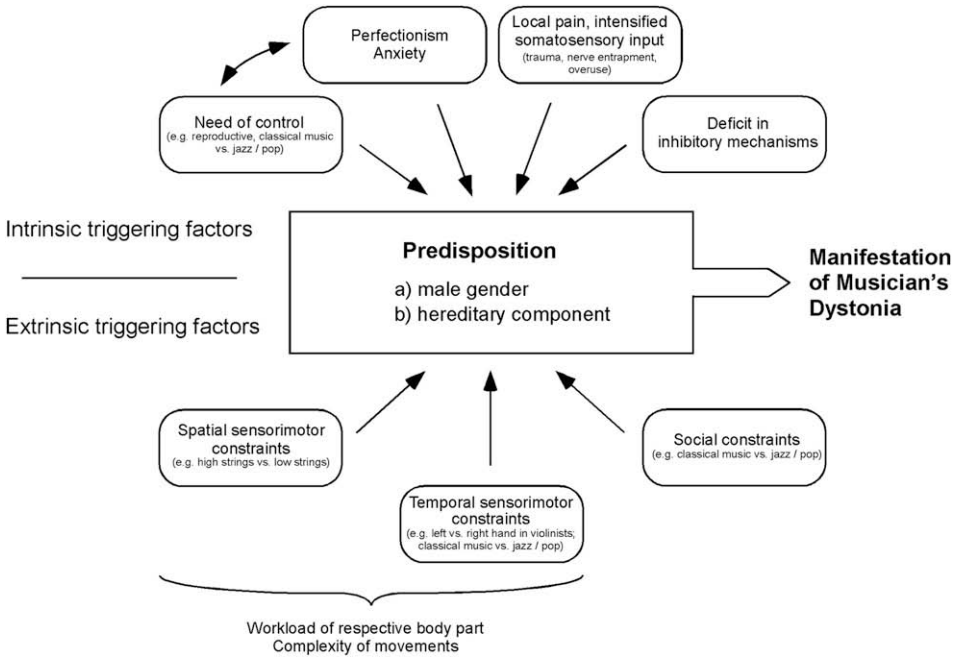

FIGURE 5. *The possible co-action between predisposition and intrinsic and extrinsic triggering factors in the manifestation of musician's dystonia. Further explanations are given in the text.*

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latter, of course, has to be collected somewhere outside the practicing room. As a consequence, we, therefore, should correct our expectations and listening habits, replacing the fascination of mere perfection and virtuosity by the joy of emotional communication shared with the audience and the musicians.

REFERENCE

1. Altenmüller E. Focal dystonia: advances in brain imaging and understanding of fine motor control in musicians. Hand Clin. 2003;19:523–38.
2. Nutt JG, Muenter MD, Melton LJ, Aronson A, Kurland LT. Epidemiology of dystonia in Rochester, Minnesota. Adv Neurol. 1988;50:361–5.
3. Nakashima K, Kusumi M, Inoue Y, Takahashi K. Prevalence of focal dystonias in the western area of Tottori Prefecture in Japan. Mov Disord. 1995;10:440–3.
4. Schmidt A, Jabusch HC, Altenmüller E, et al. Dominantly transmitted focal dystonia in families of patients with musicians cramp. Neurology. 2006;67:691–3.
5. Naumann M, Pirker W, Reiners K, Lange KW, Becker G, Brucke T. 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;13:319–23.
6. Preibisch C, Berg D, Hofmann E, Solymosi L, Naumann M. Cerebral activation patterns in patients with writer's cramp: a functional magnetic resonance imaging study. J Neurol. 2001; 248:10–7.
7. Lenz FA, Byl NN. Reorganization in the cutaneous core of the human thalamic principal somatic sensory nucleus (Ventral caudal) in patients with dystonia. J Neurophysiol. 1999;82: 3204–12.
8. Delmaire C, Krainik A, Tézenas du Montcel S, et al. Disorganized somatotopy in the putamen of patients with focal hand dystonia. Neurology. 2005;64:1391–6.
9. Byl NN, Merzenich MM, Jenkins WM. A primate genesis model of focal dystonia and repetitive strain injury: I. Learning-induced dedifferentiation of the representation of the hand in the primary somatosensory cortex in adult monkeys. Neurology. 1996;47:508–20.
10. Bara-Jiminez W, Catalan MJ, Hallett M, Gerloff C. Abnormal sensory homunculus in dystonia of the hand. Ann Neurol. 1998;44:828–31.
11. McKenzie AL, Nagarajan SS, Roberts TP, Merzenich MM, Byl NN. Somatosensory representation of the digits and clinical performance in patients with focal hand dystonia. Am J Phys Med Rehabil. 2003;82:737–49.
12. Elbert T, Candia V, Altenmüller E, et al. Alteration of digital representations in somatosensory cortex in focal hand dystonia. NeuroReport. 1998;16:3571–5.
13. Elbert T, Pantev C, Wienbruch C, Rockstroh B, Taub E. Increased cortical representation of the fingers of the left hand in string players. Science. 1995;270:305–7.
14. Altenmüller E. From the Neanderthal to the concert hall: development of sensory motor skills and brain plasticity in music performance. In: Williamon A, Coimbra D (eds). Proceedings of the international symposium on performance science. Utrecht: European Association of Conservatoires (AEC), 2007:1–14.
15. Ragert P, Schmidt A, Altenmüller E, Dinse R. Meta-plasticity in musicians. Eur J Neurosci. 2004;19:473–8.
16. Naumann M, Magyar-Lehmann S, Reiners K, Erbguth F, Leenders KL. Sensory tricks in cervical dystonia: perceptual dysbalance of parietal cortex modulates frontal motor programming. Ann Neurol. 2000;47:322–8.
17. Jabusch HC, Altenmüller E. Epidemiology, phenomenology and therapy of musician's cramp. In: Altenmüller E, Kesselring J, Wiesendanger M (eds). Music, motor control and the brain. Oxford: Oxford University Press, 2006:265–82.
18. Lederman RJ. Focal dystonia in instrumentalists: clinical features. Med Probl Perform Art. 1991;6:132–6.
19. Lim V, Altenmüller E. Musicians cramp: instrumental and gender differences. Med Probl Perform Art. 2003;18:21–7.
20. Schmidt A, Jabusch HC, Altenmüller E, et al. Clinical genetics of musician's dystonia: familial aggregation of dystonia and other movement disorders. Neurology, in press. [Q3]
21. Charness ME, Ross MH, Shefner JM. Ulnar neuropathy and dystonic flexion of the fourth and fifth digits: clinical correlation in musicians. Muscle Nerve. 1996;19:431–7.
22. Chamagne P. Functional dystonia in musicians: rehabilitation. Hand Clin. 2003;19:309–16.
23. Brandfonbrener AG, Robson C. A review of 111 musicians with focal dystonia seen at a performing artist's clinic 1985—2002. Mov Disord. 2002;17:1135–8.
24. Schuele S, Lederman RJ. Focal dystonia in woodwind instrumentalists: long-term outcome. Med Probl Perform Art. 2003; 18:15–20.
25. Gowers WR. A manual of diseases of the nervous system. London: Churchill, 1888.
26. Brain WR. Diseases of the nervous system. London: Oxford University Press, 1933.
27. Sheehy MP, Marsden CD. Writer's cramp — a focal dystonia. Brain. 1982;105:461–80.
28. Marsden CD, Sheehy MP. Writer's cramp [Review]Trends Neurosci. 1990;13:148–53.
29. Frommer J. Idiopathic writer's cramp as a psychosomatic disorder. Z Psychosom Med. 1992;38:49–62.
30. Schulze A, Jacob HW, Guckler A. Psychosomatic determinants of writer's cramp. Psychother Psychosom Med Psychol. 1992; 42:201–5.
31. Windgassen K, Ludolph A. Psychiatric aspects of writer's cramp. Eur Arch Psychiatry Clin Neurosci. 1991;241:170–6.
32. Pai MN. The nature and treatment of writer's cramp. J Ment Sci. 1947;93:68–81.
33. Gibson HB. Writer's cramp: a behavioural approach. Behav Res Ther. 1972;10:371–80.
34. Cottraux JA, Juenet C, Collet L. The treatment of writer's cramp with multimodal behaviour therapy and biofeedback: a study of 15 cases. Br J Psychiatry. 1983;142:180–3.
35. Zacher A. Writer's cramp — focal dystonia or psychogenic movement disorder? A critical literature study [Review]-Fortschr Neurol Psychat. 1989;57:328–36.
36. Ludolph AC, Windgassen K. Writer's cramp: a study of 30 cases. Nervenarzt. 1992;63:462–6.
37. Harrington RC, Wieck A, Marks IM, Marsden CD. Writer's cramp: not associated with anxiety. Mov Disord. 1988;3: 195–200.
38. Gündel H, Busch R, Ceballos-Baumann AA, Seifert E. Psychiatric comorbidity in patients with spasmodic dysphonia: a controlled study. J Neurol Neurosurg Psychiatry. 2008;78: 1398–400.
39. Bugalho P, Correa B, Guimaraes J, Xavier M. Set-shifting and behavioral dysfunction in primary focal dystonia. Mov Disord. 2008;23:200–6.
40. Herrojo-Ruiz M, Senghaas P, Grossbach M, et al. Defective inhibition and inter-regional phase synchronization in pianists with musician's dystonia (MD): an EEG study. Hum Brain Mapp, in press.
41. Rosenkranz K, Williamon A, Butler K, Cordivari C, Lees AJ, Rothwell JC. Pathophysiological differences between musicians dystonia and writers cramp. Brain. 2005;128: 918–31.
42. Altenmüller E. Robert Schumann's focal dystonia. Bogousslavsky E, , Boller E (eds). Neurological disorders on famous artists. Front Neurol Neurosci. 2005:179–88, Karger, Basel.
43. Altenmüller E. The end of the song: Robert Schumann's focal dystonia. In: Altenmüller E, Kesselring J, Wiesendanger M (eds). Music, motor control and the brain. Oxford: Oxford University Press, 2006:251–64.

44. Jabusch HC, Müller SV, Altenmüller E. Anxiety in musicians with focal dystonia and those with chronic pain. Mov Disord. 2004;19:1169–75.
45. Jabusch HC, Altenmüller E. Anxiety as an aggravating factor during onset of focal dystonia in musicians. Med Probl Perform Art. 2004;192:75–81.
46. Newmark J, Hochberg FH. Isolated painless manual incoordination in 57 musicians. J Neurol Neurosurg Psychiatry. 1987; 503:291–5.
47. Smith BH, Elliott AM, Chambers WA, Smith WC, Hannaford PC, Penny K. The impact of chronic pain in the community. Fam Pract. 2001;18:292–9.
48. Fishbein M, Middlestadt E, Ottati V, Straus S, Ellis A. Medical problems among ICSOM musicians: overview of a national survey. Med Probl Perform Art. 1988;3:1–8.
49. Fahrenberg J, Hampel R, Selg H. Das Freiburger Persoenlichkeitsinventar FPI. Revidierte Fassung FPI-R und teilweise geaenderte Fassung FPI-A1. Goettingen: Hogrefe-Verlag fuer Psychologie, 1994.
50. Krampen G. Fragebogen zu Kompetenz- und Kontrollueberzeugungen (FKK). Goettingen: Hogrefe-Verlag fuer Psychologie, 1991.
51. Altenmüller E. Causes et traitements de la dystonie de fonction chez les musiciens. Medicine des arts. 2001;36:19–27.
52. McGaugh JL. Memory – a century of consolidation [Review]Science. 2000;287:248–51.
53. Sripanidkulchai K, Sripanidkulchai B, Wyss JM. The cortical projection of the basolateral amygdaloid nucleus in the rat: a retrograde fluorescent dye study. J Comp Neurol. 1984;229: 419–31.
54. Craig KD. Emotional aspects of pain. In: Wall PD, Melzack R (eds). Textbook of pain. Edinburgh: Churchill Livingstone, 1984:153–61.
55. Linton SJ. A review of psychological risk factors in back and neck pain [Review]Spine. 2000;25:1148–56.
56. Haerkaepaeae K. Psychosocial factors as predictors for early retirement in patients with chronic low back pain. J Psychosom Res. 1992;36:553–9.
57. Harkins SW, Price DD, Braith J. Effects of extraversion and neuroticism on experimental pain, clinical pain, and illness behavior. Pain. 1989;36:209–18.
58. Jabusch HC, Altenmüller E. Focal Dystonia in musicians: from phenomenology to therapy. Adv Cogn Psychol. 2006; 2:207–20.