Anxiety as an Aggravating Factor During Onset of Focal Dystonia in Musicians

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Abstract-Focal dystonia in musicians is a task-specific movement disorder that in many cases leads to the end of musical careers. In a recent study, perfectionism and anxious traits were found to be elevated in musicians with dystonia. Dynamics of different anxiety disorders and their possible role during the development of musician's dystonia still are unclear. Dystonic musicians (n = 20) were investigated by means of self-estimation and compared with healthy musicians (n = 30) and musicians with chronic pain syndromes (n = 30)20). Participants completed a questionnaire focusing on different anxiety disorders, particularly with regard to their dynamics. Musicians with focal dystonia more often reported social phobia and specific phobias than healthy musicians. In the retrospective analysis, these differences already were present before onset of dystonia. Musicians who later developed focal dystonia more often suffered from specific phobias than musicians who later developed chronic pain. Musicians with chronic pain more often reported free-floating anxiety compared with healthy musicians. In the subscale stage fright, no significant differences were observed between the groups. The pattern of specific phobias and social phobia were reported to have been preexisting before the onset of dystonia. Anxiety and perfectionism may be aggravating factors during the development of musician's dystonia. Med Probl Perform Art 2004; 19:75-81.

 $\Gamma_{cramp,\ is\ a\ task-specific\ movement\ disorder\ that\ manifests}$ as a painless muscular incoordination or loss of voluntary motor control of extensively trained movements while a musician is playing the instrument.¹⁻⁵ For musicians who are affected, FD is highly disabling and in many cases ends musical careers. The pathophysiology is unclear. For many decades, psychological factors were believed to be essential for the development of task-specific FDs and were overemphasized in the assumed pathomechanism. In 1888, Gowers⁶ subsumed writer's cramp and related cramps under the term occupational neuroses. 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. Later, the meaning of the term shifted in the direction of a neurotic illness. As a consequence, for almost a century, occupational cramps were regarded as psychological diseases by most authors.⁷ In 1982, Sheehy and Marsden⁸ reported the absence of psychiatric disorders in patients with typist's and writer's cramp. They

stated that occupational cramps are symptoms of a physical illness, and they used the term *focal dystonia*.^{8,9} Subsequently the neuropathophysiology of FD was investigated in numerous studies.¹⁰⁻¹² The findings included alterations in the basal ganglia circuitry^{13,14} and dysfunctional plasticity in the sensory thalamus^{15,16} and somatosensory cortex¹⁷⁻¹⁹ and changes in the activation patterns of the sensorimotor cortex²⁰⁻²² and alterations in sensorimotor integration.^{23,24}

At the same time, many studies focused on psychiatric comorbidity in patients with various forms of dystonia.²⁵⁻³⁰ Depressive, anancastic, sensitive, and hysteric traits were observed in patients with writer's cramp.²⁷ Anxiety occasionally was seen in patients with writer's cramp.³¹⁻³³ It was discussed controversially whether psychological findings were present before onset of dystonia^{26,34} or whether patients showed secondary psychoreactive processes.³⁵ In contrast, in a study with 22 patients with writer's cramp no significant differences were found compared with a matched normal control group using the Crown-Crisp Experiential Index.³⁰ This questionnaire assessed traits and symptoms relevant to neurotic illness and contained three anxiety subscales (freefloating anxiety, phobic anxiety, somatic anxiety). Scheidt et al²⁹ investigated patients with blepharospasm and patients with hemifacial spasm using the SCL-90R and found scores of psychopathology ranging close to normal in both patient groups. Phobic anxiety was more pronounced, however, in patients with blepharospasm than in controls and in patients with hemifacial spasm. Social phobia was found in patients with spasmodic torticollis.³⁶

Until more recently, psychological conditions of musicians with FD had never been investigated. In one study, psychological conditions of dystonic musicians were compared with those of healthy musicians and of musicians with chronic pain (CP) syndromes.³⁷ Perfectionism and anxious traits were reported more often in musicians with FD compared with healthy musicians. In both patient groups, anxiety was found more often than in healthy musicians. Anxious traits and perfectionism were reported to have been present before onset of FD, and they could not be interpreted as secondary psychoreactive phenomena.

In this article, we report the dynamics of different anxiety disorders in musicians with FD. Findings were compared with those in healthy musicians and musicians with CP syndromes. Based on clinical observations, the underlying hypothesis was that dystonic musicians more often have social phobias and specific phobias than healthy musicians and that these anxiety disorders were present before onset of

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TABLE 1.	Distribution of Instruments in Patient Groups
	and Normal Controls

Group	FD	СР	Control
Keyboard instruments (no.)	4	10	8
String instruments (no.)	2	7	9
Woodwinds (no.)	5	1	4
Brass (no.)	4	0	8
Plucking instruments (no.)	5	2	1

CP, chronic pain; FD, focal dystonia.

dystonia. The possible role of anxiety during development of FD is discussed.

PATIENTS AND METHODS

Patients and Controls

All participants included in the study were able to speak, read, and write German fluently. Patients were recruited randomly from the outpatient clinic of the Institute of Music Physiology and Musicians' Medicine of the University of Music and Drama in Hannover, Germany. They underwent complete neurologic and psychiatric examination and were diagnosed by at least one of the authors (E.A.). Two groups of patients were included. One sample comprised 20 professional musicians (16 men and 4 women; age 36.7 ± 6.4 years) who had been diagnosed with task-related FDs. Three out of 20 patients were brass players with perioral dystonias; the other 17 patients had hand dystonias, which manifested in the typical manner as painless cramping of one or more fingers while the patients were playing their instruments. The individual duration of the disorder was 3 to 12 years (6.9 \pm 2.6 years [mean \pm SD]) at the time of the study. Twelve patients noticed additional dystonic movement patterns in other activities, such as writing (five patients), on the computer keyboard (four patients), or in everyday activities; the onset of these additional symptoms occurred after the onset of musician's cramps. Patients with other neurologic disorders or secondary dystonias were excluded from the study.

The second sample of patients comprised 20 professional musicians (5 men and 15 women; age 32.6 ± 8.7 years) with CP syndromes related to playing their instruments. Pain was localized in the hand or arm region or both. The individual duration 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 (16 men and 14 women; age 32.9 ± 5.4 years). They were working in German orchestras, in music schools, and as freelance professional musicians. By means of questionnaires, they were asked if they had any kind of ill health, especially movement disorders or CP. Musicians with any somatic complaints or neurologic or psychiatric diseases were excluded from the normal control sample. The distribution of instruments in all groups is given in Table 1.

Methods

The assessment of anxiety disorders was based on self-estimation using a questionnaire comprising six questions focusing on anxiety disorders. These were subgrouped as considerable stage fright; panic attacks; free-floating anxiety; agoraphobia; social phobia; and specific phobias, such as acrophobia and claustrophobia. 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 anxieties were preexistent before the onset of FD or CP in the patient groups, a second step was added: Subjects were asked to decide whether these particular anxieties had been present or absent before onset of their disorders and since when the respective anxieties were experienced. 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 was done by mailing; precise written explanations were attached.

Statistical analyses of the differences of anxiety disorders between the groups was performed using χ^2 tests. Two-tailed Fisher's exact tests were applied when χ^2 tests were not possible due to methodologic requirements. χ^2 tests and Fisher's exact tests were performed separately for evaluation of the differences in the anxiety subscales before and after onset of the disorders. Differences between groups with a *p* value <0.05 were considered statistically significant.

RESULTS

All questionnaires were filled out properly and could be included in the analysis. 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 of 20 patients with CP.

Analysis of the different anxiety subscales yielded the following results (Figure 1): Musicians with FD experienced social phobias remarkably more often than healthy musicians (twotailed Fisher's exact test [Fisher's], p < 0.01), which already had been the case before onset of FD (Fisher's, p < 0.01). Patients with CP did not report more social phobias having been preexisting before onset of CP compared with healthy musicians. This comparison revealed a significant difference (Fisher's, p <0.05), however, after development of CP due to an increase of social phobias in these patients after onset. Only patients with FD significantly more often reported specific phobias, such as acrophobia or claustrophobia, compared with healthy musicians (χ^2 = 4.69, p < 0.05). These specific phobias had already been present before they developed FD. Musicians who later developed FD more often suffered 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 freefloating anxiety than normal controls. This was the case before $(\chi^2 = 5.21, p < 0.05)$ and $(\chi^2 = 8.0, p < 0.01)$ after onset of CP. In the subscales considerable stage fright, panic attacks and



FIGURE 1. Anxiety disorders in controls and musicians with chronic pain and with focal dystonia. Percentage of subjects (of respective groups) who reported social phobia, specific phobias, and free-floating anxiety. Hatched bars: percentage with anxiety before onset of the respective playing-related problem. Filled bars: percentage with anxiety after onset of the respective playing-related problem. *p < 0.05. **p < 0.01. n.s., not significant.

agoraphobia, no significant differences were observed between any patient group and healthy musicians (Figure 2). No significant differences were found in any of the anxiety subscales between male and female subjects.

DISCUSSION

Inventories

The present study aimed at the assessment of different anxiety disorders in musicians before and after the onset of



FIGURE 2. Anxiety disorders in controls and musicians with chronic pain and with focal dystonia. Percentage of subjects (of respective groups) who reported considerable stage fright, panic attacks, and agoraphobia. Hatched bars: percentage with anxiety before onset of the respective playing-related problem. Filled bars: percentage with anxiety after onset of the respective playing-related problem.

FD and the comparison with healthy musicians and musicians with CP syndromes. Because of a prevalence of FD between 1:200 and 1:500 in professional musicians⁵ with the consequence of a low incidence, a prospective study with the particular aim of assessing anxiety disorders in musicians before the development of FD was not realizable. For retrospective comparison of anxiety disorders before and after the onset of FD or CP syndromes, state-trait anxiety inventories were not applicable. Because the duration of FD was 6.9 ± 2.6 years and duration of CP was 4.9 ± 6.9 years at the time of the study, it could not be excluded that anxiety traits detectable by state-trait anxiety inventories might have developed after onset of the respective playing-related problem. The temporal relationship between the onset of anxiety traits and the onset of playing-related problems could not be detected using state-trait anxiety inventories. A retrospective inquiry was undertaken that was based on personal recollection of the patients. Under the given circumstances, this procedure was the only practical approach to obtain the desired information. Although the exact date of onset of anxieties in the past might not be determinable, a retrospective inquiry does provide an image of individual developments of these psychological conditions in the past.

Findings

Social phobias and specific phobias occurred more often in dystonic musicians than in healthy musicians. This already was the case before onset of FD. The comparison between both patient groups revealed a clear difference: Musicians with FD more often suffered from specific phobias before onset of FD compared with CP patients before onset of CP. In the dystonic group, all patients described these anxieties as long-term phenomena already present before FD. As a limitation of the study, these retrospective reports are subject to bias and not always reliable. In view of the unambiguity and consistency of the reports of all patients with FD, however, we postulate that anxiety may have been preexisting and was not a psychoreactive phenomenon. Musicians with FD were reported to display more perfectionism than healthy musicians.³⁷ In the retrospective analysis, this difference already was present before onset of FD. In patients with CP, no higher level of perfectionism was found compared with healthy musicians. Taken together, dystonic musicians showed a pattern of social phobia and specific phobias and of exaggerated perfectionism before onset of dystonia, which was not seen in healthy musicians or musicians with CP. In view of the findings for musicians with FD, the question arises whether the described psychological patterns with anxiety and exaggerated perfectionism might have an impact on the development of FD in musicians.

Anxiety as an Aggravating Factor During Onset of Musician's Dystonia

There is evidence that FD is a complex disorder accompanied by neuropathologic findings, such as alterations in the basal ganglia circuitry^{13,14} and dysfunctional plasticity in the sensory thalamus^{15,16} and somatosensory cortex.^{17–19} Preexisting anxiety and perfectionism may serve as additional, aggravating factors in the development of musician's FD in the following way. During onset of musician's FD, dystonic movements are realized as disturbing and threatening, especially by musicians with an inclination toward anxiety and extreme perfectionism. Dystonic movements might become a focus in terms of a specific phobia in musicians with these tendencies. This focus might induce the cascade of emotionally induced memory consolidation that previously has been described and applied to different forms of memory and that mainly relies on noradrenergic activation of the basolateral amygdala.³⁸⁻⁴⁰ The primary motor cortex as an essential locus of representation of digital motor sequences⁴¹ receives a basolateral amygdala projection.⁴² We suggest that during the development of musician's FD in predisposed individuals, basolateral amygdala-mediated consolidation of dystonic movements as dysfunctional motor programs in the primary motor cortex may be an aggravating epiphenomenon (Figure 3).

Findings in Musicians with Chronic Pain

The findings in musicians with CP harmonize with other reports on CP patients. The association between anxiety and CP often has been described for CP syndromes in different sites.⁴³⁻⁴⁵ In particular, social phobia was found to be related to physically unexplained CP in a study with 130 patients. Additionally, in keeping with the findings of the present study, agoraphobia was found minimally pronounced in CP patients.⁴⁶ Free-floating anxiety, which was present more often in CP musicians than in healthy musicians of this study, turned out to be a predictor for early retirement in patients with CP.⁴⁷

There was a preponderance of male musicians in the sample of patients with FD. This finding is in agreement with consistent reports about higher prevalence of musician's dystonia in men.^{2-5,48,49} A preponderance of female musicians in our sample of CP patients is in keeping with other reports of a higher prevalence of CP syndromes in women.⁵⁰ Gender distribution was almost balanced in the sample of normal controls. Beside of the group affiliation, gender also was a factor in the analysis: no differences were found between male and female musicians in any of the anxiety subscales. This result suggests that the differences between the groups as described for the subscales specific phobia, social phobia, and free-floating anxiety were not distorted by different gender distributions in the groups.

No significant difference was found in the subscale of considerable stage fright between all groups. The percentage rate for stage fright (30% in healthy musicians) in this study was higher than in other reports (in Fishbein et al's⁵¹ study, 24% of 2,212 orchestra musicians had stage fright). This might be explained with the fact that our samples comprised orchestra musicians and freelance professional musicians who also were playing as soloists. Our findings suggest that stage fright does not increase the susceptibility for FD in musicians.

CONCLUSION

The occurrence of anxieties, such as specific phobias and social phobias, was higher in musicians with FD than in healthy musicians. These anxieties were present before onset of FD. In a subgroup of dystonic musicians with an inclination toward perfectionism and anxiety, in particular specific anxieties, this psychological constellation might be an aggravating factor in the development of FD. In such patients, early intervention during onset of FD including management of these particular psychological patterns, might be benefi-



FIGURE 3. Anxiety and perfectionism as aggravating factors in the development of musician's dystonia. Perfectionistic and anxious musicians experience dystonic movements as emotional stress, which may cause the release of epinephrine and glucocorticoids from the adrenal glands. Epinephrine via stimulation of the nucleus of the solitary tract causes an increased norepinephrine release of the basolateral amygdala (BLA).^{52,53} This β -adrenergic activation within the amygdala is a prerequisite for memory-enhancing effects of glucocorticoids.⁵⁴ The primary motor cortex (M1) is an essential locus of representation of digital motor sequences⁴¹ and receives a BLA projection.⁴² Emotion-dependent consolidation of dystonic movements in M1 might be an aggravating factor during the development of musician's dystonia. It is unclear whether the modulary effects of the BLA influence the basal ganglia circuitry. GPe, lateral globus pallidus; PM, premotor cortex; S1, somatosensory cortex; SMA, supplementary motor area; Gpi, medial globus pallidus; STN, subthalamic nucleus. (Parts adapted from McGaugh JL: Memory– a century of consolidation. *Science* 2000;287:248–251.)

cial. Further studies are required to illuminate the possible involvement of the limbic system in the pathogenesis of musician's dystonia.

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APPENDIX

Questionnaire

Please answer the following questions by checking the appropriate box. If you answer yes to any of the questions, please try to specify since when you have experienced the respective fears or anxieties by using the following terms:

- always
- long before the onset of the movement disorder*
- after the onset of the movement disorder*
- not any more

	No	Yes	Since When?
Are you suffering from acute (considerable) stage fright?			
Have you ever have experienced anxiety attacks (i.e., were you suddenly and unexpectedly overcome by strong fear or apprehension in a situation normally considered nonthreatening)?			
Have you ever felt anxious, tense, or worried for a prolonged period (≥ 6 months)? Did or do you often worry about events that are unlikely to occur?			
Some people are afraid of going out alone; encountering crowds of people; or traveling by car, bus, or train. Have you ever experienced such a fear?			
Some people are afraid to be, speak, or work in the presence of others, so they avoid those situations or endure them only with great discomfort. Have you ever had anxious feelings of that kind?			
In certain situations, some people experience intense anxiety (e.g., fear of heights, fear of flying, fear of enclosed spaces), so they try to avoid those circumstances. Have you ever had anxious feelings of that kind?			

*In the questionnaires for patients with chronic pain syndromes, movement disorder was replaced by pain.

This is an English translation of the questionnaire that was designed to assess anxiety disorders. The original questions were written in German.