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Musician’s cramp as manifestation of maladaptive brain plasticity: arguments from instrumental differences

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Musician’s cramp 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. It is characterized by task specificity and gender bias, affecting significantly more males than females. The etiology is multifaceted, however, electrophysiological, genetic, and epidemiologic findings point at combination of a genetic predisposition, termed endophenotype, and behavioral triggering factors leading to the manifestation of the disorder.

We present epidemiological data from 591 musician patients from our outpatient clinic demonstrating an influence of fine motor requirements on the manifestation of dystonia. Brass, guitar, and woodwind players were at greater risk than other instrumentalists. With respect to the body part affected, high temporospatial precision of movement patterns and synchronous demands on tonic and precise phasic muscular activation constitute triggering factors, especially in combination with daily-life fine motor burdens of the dominant hand.

Key words: musician’s cramp; dystonia; maladaptive plasticity; endophenotype; instrument; gender

Introduction

Musician’s dystonia (MD), also known as musician’s cramp or focal dystonia in musicians, 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

MD 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,² whereas pianist’s cramp and violinist’s cramp may affect the control of finger, hand, or isolated arm movements.³ Typically, MD occurs without pain, although muscle aching can present after prolonged spasms. The loss of muscular coordination is frequently accompanied by a cocontraction of antagonist muscle groups. For example in pianist’s cramp, the coactivation of wrist flexor and wrist extensor muscles is frequently observed. Other diagnostic criteria are the consistency of dystonic symptoms when playing, although the degree of severity may vary according to general tension and mechanical properties of the instrument. However, as a diagnostic criterion, dystonic symptoms should be apparent shortly after beginning to play the instruments. We therefore do not consider abnormal fatigue or degradation of motor executive functions after prolonged playing as MD. According to recent estimates, one percent of all professional musicians are affected.¹ In contrast, in the general population, prevalence of focal dystonias, including...
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writer’s cramp, blepharospasm, and cervical dystonia is estimated as 29.5 per 100,000 in the United States and 6.1 per 100,000 in Japan.\textsuperscript{4,5}

With respect to the etiology, it is generally acknowledged that MD may represent a syndrome of central nervous maladaptive plasticity.\textsuperscript{6} In healthy musicians, adaptive plastic reorganization of brain circuits has been demonstrated. Sensory–motor cortex receptive fields of single digits are enlarged,\textsuperscript{7} and both, motor excitability and long-term potentiation-/long-term depression-like plasticity are enhanced.\textsuperscript{8} It has been speculated that this has behavioral advantages because it may facilitate motor control of adjacent fingers, for example, for the playing of fast passages.

However, in MD patients, the reorganization of sensory–motor networks goes one step further: spatial differentiation of single digits disappears and the topography of receptive fields becomes disorganized.\textsuperscript{9} Furthermore, the excitability of projections to all hand muscles is enhanced in an undifferentiated manner.\textsuperscript{10} Although this excess reorganization is associated with loss of task-specific motor control, it is not possible to say whether it causes the breakdown in motor control, or whether it is a consequence of persistent abnormal movement patterns. However, the restitution of the topography of receptive fields along with clinical recovery in MD strongly suggests a causal relationship.\textsuperscript{11}

Based on neurophysiological, genetic, and epidemiologic findings, we have recently proposed a heuristic model of the manifestation of MD.\textsuperscript{12} We assume a genetic predisposition, the endophenotype, which is characterized by a deficit in inhibitory mechanisms in sensory–motor networks on several levels of the central nervous system. This translates into temporal and spatial overshoot in motor activation, leading to less-focused movement patterns and to degradation of sensory discrimination abilities.\textsuperscript{13–15}

Whereas in nonmusicians, in most instances the demands on temporal and spatial precision of movement patterns are not high enough to trigger clinical manifestations of these maladaptive plastic changes, in professional musicians, several conditions seem to contribute to the manifestation of MD: prolonged practice; chronic pain; predefined temporospatial constraints—for example, need to play “correct notes” in sheet-music\textsuperscript{16}; psychological conditions, such as anxiety and perfectionism\textsuperscript{17}; gender\textsuperscript{18}; and requirements of the musical instrument. All these factors may contribute to trigger the development of a dystonia on the basis of an endophenotype.

It has been found in previous studies that MD is not equally distributed among different instrumentalist’s groups.\textsuperscript{19,20} For example, among string players, focal dystonia seems to be more frequent in high strings than in low strings.\textsuperscript{21} Interestingly, in musicians with hand dystonia, an association exists between the instrument group and the localization of focal dystonia. In instruments with different workload, different complexity of movements or different temporospatial precision for both hands, focal dystonia appears more often in the more heavily used hand. Keyboard musicians (piano, organ, and harpsichord) and those with plucked instruments (guitar and e-bass) are primarily affected in the right hand. All these instruments are characterized by a higher workload in the right hand. In addition, guitar playing requires higher temporospatial precision in the right hand compared to the left hand. Bowed string players who have a higher workload and complexity of movements in the left hand are predominantly affected in that hand.\textsuperscript{22}

In a recent study, we were able to demonstrate that not only the nature of movements required by the respective instruments were related to dystonic movements’ patterns, but also extra instrumental burdens of daily-life activities: in left-handed musicians, MD occurs more frequently in the left hand compared to right-handed musicians and vice versa, suggesting that fine motor activities of the dominant writing hand should be added as an additional triggering factor.\textsuperscript{23}

In summary, we suggest that phenotypic occurrence of MD is more heavily influenced by environmental and behavioral factors than previously assumed. To further substantiate this notion, this paper focuses on three aspects: Taking advantage of a very large patient group suffering from MD, we first sought to confirm that the differential workload related to playing different instruments is an important trigger factor. Second, the effect of gender on the prevalence of musicians’ cramp was also examined by comparing the patients’ records with those of eight music conservatories in Germany. In these respects, the present work is an extension of a previous paper on 183 patients.\textsuperscript{18} Third, we included self-declared handedness as a factor in our statistical analysis. The rationale behind this was the idea
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that handedness might also constitute an additional risk factor, given that sensory–motor workload does not only occur during instrumental playing but also during other skilled movements executed primarily by the dominant hand, for example writing or operating a cell phone. This latter analysis was partly published previously.23

Methods

Patients

Patient files of 591 consecutive patients diagnosed with MD between 1994 and 2007 in the outpatient clinic of the Institute of Music Physiology and Musicians’ Medicine (IMMM) of the Hanover University of Music, Drama and Media (Hannover, Germany) were evaluated. Patient files comprised diagnosis, description of symptoms, and additional information, such as self-declared handedness, which was used as a variable for statistical analysis. All patient files were examined, and only those with a clear diagnosis of focal dystonia were included in this study. Exclusion criterion was any other or secondary neurological disorder. Diagnoses were confirmed by a trained movement disorders specialist and neurologist (author E.A.).

Controls

The control group was taken from the previous study of Lim and Altenmüller18 and consisted of 2,651 students enrolled in studies of all instrumental groups from eight different music conservatories. Mean age differed between the patient and control group, because the students were slightly younger than the patients, given that the age of onset of MD is generally in the fourth decade of life.16

Data analysis

For statistical analysis, the software package SPSS 15.0 was used. Chi-square ($\chi^2$) tests were employed for comparisons of ratios between groups. In cases of small sample sizes, Fisher’s exact test was applied. The level of significance was set at $\alpha = 0.05$ and all $P$ values are reported uncorrected for multiple comparisons because we applied a strongly hypothesis-driven analysis. Instruments were categorized in the following manner: keyboard (piano, organ, harpsichord, accordion, and keyboard), string (violin, cello, viola, and double bass), plucked instruments (guitar, e-bass, and mandoline), woodwind (flute, clarinet, saxophone, oboe, bassoon, recorder, and bagpipes), brass (trombone, horn, trumpet, tuba, and bariton), and percussion (drums and kettledrum).

Results

The majority of the patients were professional musicians (93.9%). From 478 patients, self-declared handedness was noted in the files and was distributed in the following manner: 89.3% right-handed, 9.2% left-handed, and 1.5% ambidextrous. Thirty-two (5.4%) of the patients were simultaneously affected by the writer’s cramp.

Localization of focal dystonia

Focal dystonia was predominantly located in the upper limb (hand and arm; 83.1%); whereas focal lower limb dystonia could be seen in four patients, cervical dystonia (e.g., torticollis) could be seen in seven patients. One hundred and twenty (20.1%) patients suffered from embouchure dystonia (cramping or tremor of the lips). There was a higher ratio of patients with embouchure dystonia than upper limb dystonia when comparing brass with woodwind players ($\chi^2 = 121.76$, $df = 1$, $P < 0.001$). Not surprisingly, percussionists were more likely to have lower limb dystonia than all other musicians (Fisher’s exact test, $P < 0.001$). Among all cases with upper limb dystonia ($n = 465$), arm dystonia was much less common than hand dystonia (3.64% vs. 96.36%). String players had a higher rate of arm dystonia than all other musicians (16.05% vs. 1.03%; Fisher’s exact test, $P < 0.001$). Within patients playing strings, arm dystonia was more often in the right side than left (seven right vs. three left; right is the bowing hand) when compared with hand dystonia (Fisher’s exact test, $P = 0.049$).

Laterality of focal upper limb dystonia

Dystonic symptoms among patients with unilateral focal upper limb dystonia (localized in hand or arm, $n = 452$) occurred on different sides ($\chi^2 = 66.59$, $df = 3$, $P < 0.001$). Because only 2 brass players and 11 percussionists were affected in the hand or arm, these two instrument groups were excluded from this test. Follow-up $\chi^2$-tests yielded significant differences for keyboard versus string players ($\chi^2 = 49.31$, $df = 1$, $P < 0.001$), keyboard versus woodwind players ($\chi^2 = 31.68$, $df = 1$, $P < 0.001$), string players versus players with plucked instruments ($\chi^2 = 29.87$, $df = 1$, $P < 0.001$), and woodwind players versus players with plucked instruments ($\chi^2 = 17.35$, $df = 1$, $P < 0.001$). This implies that

keyboard musicians and those with plucked instruments were predominantly affected in the right hand and string players in the left hand. No general lateralization could be seen in woodwind players (46.2% right, 49.5% left, and 4.4% on both hands). However, within woodwind players, patients playing the flute were mainly affected on the left and those playing oboe or clarinet on the right side ($\chi^2 = 5.87$, df = 1, $P = 0.015$). Keyboard musicians suffered more often from bilateral focal upper limb dystonia than musicians of all other instrumental groups (7.3% in keyboards vs. 3% in other instruments, $\chi^2 = 5.30$, df = 1, $P = 0.021$).

As already mentioned, an association between handedness and laterality of dystonia could be observed in patients with unilateral hand dystonia ($n = 362$, ambidextrous patients were excluded from this test). That is, the percentage of right-handers was higher among patients with right-sided dystonia compared to left-sided dystonia, and accordingly, the percentage of left-handers was higher among patients with left-sided dystonia compared to right-handed dystonia ($\chi^2 = 4.07$, df = 1, $P = 0.044$).

**Rare symptoms of focal dystonia**

Focal task-specific tremor affected only 3.4% ($n = 20$) of the patients, whereas the majority of the patients showed the typical symptom of cramping. String players had focal tremor more often than all other instrumentalists (Fisher’s exact test, $P < 0.001$). This tremor was exclusively (100%) located in the right side, whereas cramping occurred to a lower percentage (30%) on the right side (Fisher’s exact test, $P < 0.001$).

Among the patients with cervical dystonia ($n = 7$), three suffered from dystonic tremor in this body region, whereas torticollis, respectively anterocollis, could be seen in the other four patients. Compared to other areas of the body, the ratio of tremor was higher in cervical dystonia than in all other dystonias (42.9% vs. 2.9%, Fisher’s exact test, $P < 0.001$; the small sample size of patients with focal tremor should be noted).

**Interaction between instrument and population**

This comparison involved instrument versus population (dystonia vs. healthy; $\chi^2 = 146.61$, df = 5, $P < 0.001$). Gender was not a factor in this analysis. Follow-up two-by-two tests were employed (e.g., keyboard players and nonkeyboard players separated in patients and controls; Fig. 1).

The observed number of patients was higher than the expected number in the following three instrumental groups: plucked instruments ($\chi^2 = 60.48$, df = 1, $P < 0.001$), woodwind ($\chi^2 = 5.00$, df = 1, $P = 0.025$), and brass ($\chi^2 = 20.79$, df = 1, $P < 0.001$). No significant differences between observed and expected numbers could be seen in keyboard players ($\chi^2 = 0.15$, df = 1, n.s.) and percussionists ($\chi^2 = 0.39$, df = 1, n.s.). Only in the group of string

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**Figure 1.** The distribution of instruments in 591 patients with MD as compared to 2,651 healthy musicians displayed as relative ratios. Highly significant differences in both populations ($P < 0.001$) are marked with three asterisks and significant findings ($P < 0.025$) with one asterisk. MD is more frequent than expected in plucking instruments, brass instruments, and woodwinds and less frequent in string players.
Figure 2. Gender distribution in 591 patients suffering from MD as compared to 2,651 healthy musicians displayed as relative ratios. Significantly more male musicians than females suffer from MD ($P < 0.001$).

players, the observed number of patients was lower than the expected number ($\chi^2 = 99.23$, df = 1, $P < 0.001$).

Interaction between gender and population
This interaction involved gender and population, whereas instrumental groups were not split up in this analysis. There were more males than females in the dystonia population (77.8% vs. 22.2%, i.e., a ratio of 3.5:1), whereas the proportion of females was higher than that of males in the control group (55.5% vs. 44.5%). This interaction was significant ($\chi^2 = 214.68$, df = 1, $P < 0.001$; Fig. 2).

Discussion
The results again confirm the “trigger hypothesis,” which poses that the clinical manifestation of mal-adaptive brain plasticity in MD is strongly influenced by specific triggering factors, for example, high requirements on manual skills, temporospatial precision of movements, and extra instrumental burdens linked to handedness and skilled use of the dominant hand.21,23 The cause of the gender prevalence of MD in male musicians still remains unclear. Previous studies report gender ratios between 2:1 and 5:1 (male: female) in focal dystonia patients.24–26 This study with its large sample size reveals a ratio of 3.5:1. Taking into account that there are more females in the control group by a factor of 1.2, it can be concluded that males are at greater risk for developing MD than females by a factor of 4.3. It is an open question why males’ susceptibility for MD is higher. There might be hormonal reasons or possibly different psychological attitudes concerning work behavior and coping with stressors in men and women.

Localization of MD
With respect to the higher prevalence of embouchure dystonia in brass and woodwind players, it is obvious that in brass playing, the control of lip movements is more critical for sound production when compared to woodwind players. Brass players are required to fine tune the tension of embouchure muscles to obtain a highly precise control of frequency and amplitude of lip vibrations. In woodwinds, embouchure adjustments do not require lip vibration, and therefore are less time-constrained. Conversely, finger movement patterns are more complex in woodwind than brass players, explaining why hand dystonia is a rare exception in brass players. A similar influence of workload, related to the body part affected can be seen in arm dystonia: in string players, the right arm is involved in precise bowing movements. Therefore, a relatively high incidence of left arm dystonia can be observed only in this group.
The results for the laterality of focal upper limb dystonia indicate a similar influence of workload as a trigger. In keyboard and plucked instruments, movement patterns are more complex in the right upper limb, whereas in string players, there is higher spatial and temporal precision needed in the left hand.27

Within woodwind players, flutists have MD mainly on the left side, whereas oboists and clarinetists show their symptoms predominantly in the right hand. This might be due to the combination comprising prolonged support of the instrument with the thumb (left in the flute and right in clarinet and oboe) and fine motor control of the other four fingers. Obviously, this double task in one hand represents a challenge for the motor system since tonic activation and, at the same time, fine motor phasic control of adjacent fingers in the millisecond range is required.

Although there are different workloads in the hands for keyboard players (76% of them are affected in the right upper limb), this instrumental group has the highest percentage of bilateral dystonia (12%). One explanation at hand is that there are similar movements in the right and left hand.

Interaction between instrument and population
As already shown,16 the distribution of musicians’ cramp across instrumental groups differs and certain instrumental groups are at higher risk for developing MD. In contrast to the previous study, here we show that musicians’ cramp occurs more often than expected in brass players as well, whereas percussionists are not at special risk for focal dystonia. The results for the other instrumental groups are in line with the previous study: plucked instruments and woodwind players have higher numbers of observed patients and string players have lower numbers. For keyboard players, observed and expected numbers do not differ significantly. This suggests that workload and complexity of movements may depend on the instrument group, with brass instruments, plucked instruments, and woodwind players showing the highest risk for focal dystonia.

Within the patients who play stringed instruments, high strings (violin and viola) are overrepresented. Compared to the ratio of high and low string players in German orchestras, it can be concluded that high string players are at higher risks for MD than low strings.28 Again, this suggests that movement patterns requiring higher spatial and temporal precision are a risk factor for the manifestation of MD.

Conclusions
This study confirms in a large sample of musicians suffering from MD that high demands on temporospatial precision and a complex nature of movement patterns are the risk factors for developing MD. Because MD is a disorder based on maladaptive changes in the central nervous sensory motor networks triggered by a variety of behavioral factors, it seems appropriate to pay specific attention to all young musicians learning to play “instruments at risk” to teach them prevention strategies such as avoidance of overuse and chronic pain.

Conflicts of interest
The authors declare no conflicts of interest.

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Queries

Q1  Author: Please check the affiliations as typeset for correctness.

Q2  Author: Please check the edit to the sentence “Focal dystonia was...in seven patients.” for correctness.