



Personality profiles are different in musician's dystonia and other isolated focal dystonias

Susanne Steinlechner^{a,b}, Hans-Christian Jabusch^c, Eckart Altenmüller^d, Friederike Borngräber^{e,f}, Johann Hagenah^g, Christine Klein^h, Rebekka Lencerⁱ, Alexander Schmidt^{e,f,h,*}

^a Department of Psychiatry and Psychotherapy, University of Lübeck, Lübeck, Germany

^b Helios Fachklinik Schleswig, Schleswig, Germany

^c Institute of Musicians' Medicine, Dresden University of Music "Carl Maria von Weber", Dresden, Germany

^d Institute of Music Physiology and Musicians' Medicine, University of Music, Drama and Media, Hanover, Germany

^e Berlin Center for Musicians' Medicine, Charité – University Medicine Berlin, Berlin, Germany

^f Kurt Singer Institute for Music Physiology and Musicians' Health, Hanns Eisler School of Music Berlin, Berlin, Germany

^g Department of Neurology, Westküstenklinikum Heide, Heide, Germany

^h Institute of Neurogenetics, University of Lübeck, Lübeck, Germany

ⁱ Department of Psychiatry and Psychotherapy and Otto Creutzfeldt Center for Behavioral and Cognitive Neuroscience, University of Münster, Münster, Germany



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ABSTRACT

Psychological abnormalities have been reported in patients with musician's dystonia. To further differentiate these abnormalities, we evaluated personality traits in musician's dystonia and compared them to those in other isolated focal dystonias. Therefore patients with musician's dystonia ($n = 101$) and other isolated focal dystonias ($n = 85$) underwent the Neuroticism Extraversion Openness Five-Factor Inventory (NEO-FFI). Women with musician's dystonia had higher NEO-FFI neuroticism scores, and men significantly higher openness scores compared to women and men with other isolated focal dystonias, respectively. There were negative correlations in men with musician's dystonia between duration of dystonia and the NEO-FFI openness and extraversion scores and between age and extraversion scores. Women with other isolated focal dystonias showed correlations between age and agreeableness and conscientiousness scores. Patients with musician's dystonia are characterized by a specific personality profile with increased neuroticism and openness compared to other isolated focal dystonias. Whether this profile can be traced back to specific underlying disease mechanisms should be further investigated.

1. Introduction

Musician's dystonia, a task-specific form of isolated focal dystonia, presents with loss of voluntary motor control when a musician is playing his instrument (Altenmüller, 2003; Frucht et al., 2001; Jankovic and Ashoori, 2008). In addition to impaired motor function, psychological abnormalities have also been reported in musician's dystonia patients, such as increased anxiety, social and specific phobias, perfectionism and neuroticism compared to both healthy musicians and non-musician controls (Enders et al., 2011; Jabusch et al., 2004; Jabusch and Altenmüller, 2004). Psychological abnormalities have also been identified in patients with other types of isolated focal dystonia, such as increased prevalence of anxiety disorders and major depression in patients with cervical dystonia (Berman et al., 2017), mood disorders

in patients with blepharospasm (Defazio et al., 2017; Lencer et al., 2009), and obsessive compulsive disorder in patients with focal hand dystonia including musician's dystonia cases (Ioannou and Altenmüller, 2014; Voon et al., 2010). Additionally, different personality traits compared to population controls have been found in other isolated focal dystonias, with increased conscientiousness and agreeableness, and reduced openness (Lencer et al., 2009).

Although common pathophysiological mechanisms (i.e. disturbances in basal ganglia-thalamo-cortical motor loops may additionally impact limbic loops) are suggested to underlie motor and psychiatric symptoms in all types of isolated focal dystonia (Enders et al., 2011; Ron, 2009), endophenotype studies identified differences between musician's dystonia and other isolated focal dystonias (Bradley et al., 2012) possibly reflecting different disease mechanisms.

* Corresponding author at: Berlin Center for Musicians' Medicine Department of Audiology and Phoniatrics Charité – University Medicine Berlin, Charitéplatz 1, 10117 Berlin, Germany.

E-mail address: alexander.schmidt@charite.de (A. Schmidt).

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In addition, a recent study identified two different psychological profiles for musician's dystonia (i.e. patients with and without increased anxiety, perfectionistic and stress characteristics) suggesting subtypes of the disease with different disease mechanisms (Ioannou and Altenmüller, 2014). Aims of the present study were therefore to further evaluate personality traits in musician's dystonia and to compare them to other isolated focal dystonias.

2. Methods

2.1. Participants and psychological assessment

We consecutively recruited professional musicians with musician's dystonia and non-musician patients with other isolated focal dystonias from the outpatient clinics of the Hanover Institute of Music Physiology and Musicians' Medicine and the University Hospital in Lübeck, Germany, respectively, and assessed personality traits using the revised German version (Borkenau and Ostendorf, 2008) of the self-administered Neuroticism Extraversion Openness Five-Factor Inventory (NEO-FFI) (Costa and McCrae, 1985). All patients underwent a complete neurological examination and were diagnosed by at least one of the authors. Patients with clinical evidence of secondary dystonias or carrying a mutation in a known dystonia gene (*Tor1A* or *THAP1*) were excluded. The NEO-FFI was distributed and collected by mail accompanied by detailed written instructions and additional questionnaires covering demographic information, medical history and accumulated live practice time on the instrument for the musicians or within a structured personal interview. The study was approved by local ethics committees and written informed consent was obtained from all participants.

2.2. Statistical analyses

To account for differences in age and gender distribution between the study groups, NEO-FFI z-scores were calculated for each participant. The individual NEO-FFI raw scores were subtracted from age- and sex-specific mean scores of a standardized norm sample consisting of 11,724 German-speaking healthy individuals provided in the German NEO-FFI handbook (Borkenau and Ostendorf, 2008) and the difference was divided by the standard deviation of the corresponding sample. These sex- and age-normalized z-scores and other continuous variables were compared between the two study groups using *t*-tests, categorical variables using chi-square tests. Correlations between clinical parameters, the accumulated live practice time on the instruments and the NEO-FFI raw scores were determined using Pearson's *r*. All statistical tests were performed with a significance level of 0.05 using PSpP statistics (version 0.9.0-g3a3d58), an open source statistical software by GNU. The Holm sequential Bonferroni procedure was applied as a correction for multiple comparisons.

3. Results

The musician's dystonia group ($n = 101$) consisted of musicians with hand and embouchure dystonia, the other isolated focal dystonias group ($n = 85$) of patients with cervical dystonia, blepharospasm, writer's cramp and other task-specific dystonias such as dystonia when knitting. Patients with musician's dystonia included significantly fewer women ($\chi^2 = 26.10$; $df = 2$; $p = 0.000$), were younger ($T = -4.74$; $df = 184$; $p = 0.000$) and had an earlier age of onset ($T = 4.33$; $df = 179$; $p = 0.000$) compared to the other isolated focal dystonia patients (Table 1). However, these differences were negligible in further analyses due to computing sex- and age-normalized z-scores. Psychiatric disorders were reported by 28 (27.8%) and 47 (55.3%) patients with musician's dystonia or other isolated focal dystonias, respectively, however heterogeneous methods (questionnaires or personal interview) were used for the assessment (Table 1).

Stratified analyses by gender revealed that women with musician's dystonia had higher NEO-FFI neuroticism subscale scores ($T = 2.22$; $df = 87$; $p = 0.029$) compared to women with other isolated focal dystonias, but this result did not meet significance after correction for multiple testing. Musician's dystonia men showed significantly higher openness scores ($T = 4.5$; $df = 95$; $p = 0.000$) than men with other isolated focal dystonias (Table 2). In musician's dystonia men, there were negative correlations between the duration of dystonia and the NEO-FFI openness (Pearson's $r = -0.27$, $p = 0.027$) and extraversion (Pearson's $r = -0.35$, $p = 0.004$) subscale scores and also between age and the extraversion subscale scores (Pearson's $r = -0.29$, $p = 0.016$). In women with other isolated focal dystonias, there were correlations between age and the NEO-FFI agreeableness (Pearson's $r = 0.39$, $p = 0.003$) and conscientiousness (Pearson's $r = 0.27$, $p = 0.043$) subscale scores. However, these correlations did not meet significance after correction for multiple testing. No further differences or correlations were detected.

4. Discussion

The present study demonstrates that patients with musician's dystonia have different personality profiles compared to patients with other isolated focal dystonias, i.e. women showed higher NEO-FFI neuroticism subscale scores and men significantly higher openness scores. Increased neuroticism has already been identified in musician's dystonia patients compared to healthy musicians and non-musician controls using the NEO-FFI (Enders et al., 2011). Similarly, increased conscientiousness and agreeableness, and reduced openness have been previously reported in other isolated focal dystonia patients compared to population controls (Lencer et al., 2009). Neuroticism has also been linked to healthy musicians (Kemp, 1996). Furthermore, the dimension of openness has previously been connected to a specific cognitive style characterized by independence which is strongly linked to healthy musicians (Kemp, 1996). Therefore, higher openness scores in male musicians with musician's dystonia and higher neuroticism scores in female musicians with musician's dystonia compared to their counterparts with other isolated focal dystonias may in part be a consequence of their occupation as a professional musician. Though, these personality traits might also have contributed to the overall decision to choose music as a profession.

In men with musician's dystonia, there were negative correlations between the NEO-FFI openness and extraversion subscale scores and the duration of dystonia and between the extraversion scores and age and, in women with other isolated focal dystonias, correlations between the agreeableness and conscientiousness scores and age. A negative correlation between NEO-FFI openness scores and the duration of dystonia has previously also been reported in musician's dystonia patients (Enders et al., 2011). These results could be interpreted as psychoreactive phenomena as a consequence of dystonia and may indicate different disease coping strategies in musician's dystonia compared to other isolated focal dystonias. However, since none of the correlations yielded significant effects after correction for multiple testing, these considerations are only made tentatively.

Shared pathophysiological mechanisms are hypothesized to underlie motor and psychiatric symptoms in all types of isolated focal dystonia (Altenmüller et al., 2015; Enders et al., 2011; Lencer et al., 2009; Ron, 2009) leading to shared motor features (e.g. involuntary finger flexion/extension). However, endophenotypic differences between musician's dystonia and other isolated focal dystonias (Bradley et al., 2012) and two different psychological profiles for musician's dystonia have recently been identified (Ioannou and Altenmüller, 2014) suggesting different disease mechanisms. The identified differences in personality profiles in this study may also underline the assumption of different disease mechanisms underlying the focal dystonia phenotypes. However, specific pathophysiological studies on personality profiles in different types of focal dystonia are

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