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Psychological characteristics in musician’s dystonia: A new diagnostic classification

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A B S T R A C T
Numerous studies suggest that dysfunctional basal ganglia-thalamo-cortical circuits are involved in both movement disorders and psychiatric impairments. The current exploratory investigation explored possible psychological differences, firstly between 24 healthy musicians (HM) and 24 musicians diagnosed with focal dystonia (FDM) (Study I), and secondly among 35 FDM patients only (Study II). Results revealed that FDM patients are six times more likely to exhibit elevated anxiety, perfectionistic and stress characteristics than HM. These psychological conditions might contribute as aggravating risk factors to the development of FDM. However, half of the FDM patients did not demonstrate any signs of anxiety, perfectionism or stress. The findings point to the clear existence of two different psychological profiles among FDM patients. We suggest that this psychological distinction might reflect two different mal-adaptive processes mediated via different circuits of the cortico-basal ganglia-thalamocortical loops. The new classification of FDM patients will contribute to the reinforcement of the diagnostic repertoire, necessary for the selection of more specific treatment methods.

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1. Introduction

Focal dystonia in musicians (FDM) is one of the most severe diseases among instrumental performers. It is a task-specific movement disorder, also known as musician’s cramp, and is commonly sub-classified into two major forms: focal hand dystonia (FHD) and embouchure dystonia (ED) (Altenmüller, 2003). Both types are characterised by muscular incoordination or loss of voluntary motor control during task-specific trained movements, such as playing a musical instrument (Altenmüller, 2003; Furuya & Altenmüller, 2013; Jankovic & Ashoori, 2008). In most cases, pain does not accompany the disorder; occasionally some muscular strain can occur due to efforts to compensate for the dystonic movement by over-activation of the antagonist muscles. It affects approximately 1% of professional musicians, however there is probably a considerable unreported “dark” number, estimated at up to an additional 1%. FDM frequently terminates professional careers and is highly disabling amongst musicians (Altenmüller, 2003; Altenmüller & Jabusch, 2010; Bostantjopoulou, 2009).

The aetiology of FDM is multifactorial and up to now not fully understood (Altenmüller & Jabusch, 2009). Principally the disorder can affect all motor functions relevant for playing the instrument, whether they relate to the hand, the upper arm in string-players, the embouchure and tongue in wind players, or even foot control in drummers (Altenmüller & Jabusch, 2010). Various epidemiological studies have revealed that behavioural and extra-instrumental factors concerning fine motor hand-use influence the location of hand dystonia: higher demands in tempo-spatial precision required to play the instrument seem to contribute to triggering this form of the disorder. For example, guitarists and pianists are more frequently affected in the right hand, while violinists’ left hands tend to suffer. Moreover, classical violinists are more affected than classical double bass players (Altenmüller, 2003; Conti, Pullman, & Frucht, 2008; Frucht, 2009; Jabusch & Altenmüller, 2006; Schuele & Lederman, 2004). With respect to extra-instrumental factors, left-handed musicians develop dystonia more frequently than expected in the left hand, and right-handed musicians in the right hand (Baur, Jabusch, & Altenmüller, 2011). Additionally, social constraints may contribute as extrinsic triggering factors to the development of dystonia in musicians. Classical musicians who mainly perform in a “reproductive” manner (that is, playing music which has already been published and performed) are at far greater risk of developing dystonia than improvising musicians, for example in the jazz genre (Altenmüller & Jabusch, 2010; Chamagne, 2003; Jabusch & Altenmüller, 2006; Schuele & Lederman, 2004). This asymmetry may be related to
stresses induced by error avoidance, since clearly predefined tempor-spatial targets in sheet music provide an unyielding frame for “what is correct, what not” in professional performers.

Furthermore, several intrinsic predisposing factors such as gender and genetics play a role. Males are four times more frequently affected than females (Lederman, 1991; Lim & Altenmüller, 2003) and in up to 35% of cases, other family members suffer from a dystonic movement disorder (Schmidt et al., 2006, 2009). Finally, overuse injuries, nerve-entrapment or related conditions and disorders could be linked to sensory disturbances such as numbness or tingling, and to dysfunctions of sensory motor circuits which can trigger the development of dystonia (Altenmüller, 2003; Chamagne, 2003; Charness, Ross, & Shefner, 1996).

FDM is currently categorised as a primary dystonia. Primary dystonia defines situations where dystonia is the only neurological impairment in a patient and there is no symptomatic cause, such as receipt of antipsychotic treatment, or peripheral injury. With respect to the underlying pathophysiological mechanisms, primary focal dystonia (FD), is related to the lack of inhibition in sensory-motor systems and to dysfunctional brain plasticity, such as the reduction of sensory perception and integration (Altenmüller & Jabusch, 2009; Levy & Hallett, 2002; Lin & Hallett, 2009; Torres-Rusotto & Perlmutter, 2008). The distinct enlargement of somato-sensory digit representations has been described as an example of adaptive plasticity among healthy musicians (Elbert, Pantev, Wienbruch, Rockstroh, & Taub, 1995; Ragert, Schmidt, Altenmüller, & Dinse, 2004), whereas an “overlap” of the digits with less precisely defined borders of receptive fields and blurring of the representations in the primary somato-sensory cortex has been observed in musicians suffering from FHD, suggesting mal-adaptive plasticity of the brain (Candia et al., 2002; Elbert et al., 1998). Furthermore, the pathophysiology of FHD, albeit still not fully understood (Lin & Hallett, 2009; Tanabe, Kim, Alagem, & Dauer, 2009) seems to be related to alterations of basal ganglia circuitry (Berardelli, 2006; Lim, Altenmüller, & Bradshaw, 2001; Mink, 2003; Naumann et al., 1998; Preibisch, Berg, Hofmann, Solymosi, & Naumann, 2001), associated with dysfunctional pathways of the sensory thalamus (Lenz & Byl, 1999) and various cortical regions (Byl, Merzenich, & Jenkins, 1996; Delmair et al., 2007; Egger et al., 2007; Garraux et al., 2004; Ibanez, Sadato, Karp, Deiber, & Hallett, 1999; Polujo et al., 2000).

Investigations of the psychological characteristics of various types of FD suggest that many movement disorders may affect similar neurobiological networks to those implicated in common psychiatric diseases (Ron, 2009). For example cervical dystonia and blepharospasm was found to be associated with anxiety, depression (Moraru et al., 2002; Müller et al., 2002), and obsessive-compulsive disorders (Barahona-Corrêa, Bugalho, Guimarães, & Xavier, 2011; Bihari, Pigott, Hill, & Murphy, 1992; Broocks, Thiel, Angerstein, & Dressler, 1998; Mula et al., 2012; Munhoz et al., 2005). In other studies, cervical dystonia was linked to phobia and depression (Gündel, Wolf, Xidara, Busch, & Ceballos-Baumann, 2001), whereas stress and anxiety was observed in writer’s cramp patients (Cottraux, Juenet, & Collet, 1983). It seems that abnormal functions of basal ganglia-thalamo-cortical networks, which also involve limbic structures, could encode motor performance in response to emotions (Jabusch & Altenmüller, 2004; Ron, 2009).

Despite these associations, the contribution of various psychological conditions as aggravating factors in the development of FD remains unclear. Few studies have investigated the psychopathology of FDM. Jabusch, Müller, and Altenmüller (2004) compared healthy musicians (HM) with FDM patients and musicians with chronic pain. They suggested that both patient groups display higher levels of anxiety than HM, with additional higher levels of perfectionism among FDM patients. A second study based on a similar paradigm reported higher levels of social phobia and specific phobias (Jabusch & Altenmüller, 2004), whereas a more recent investigation reported higher levels of anxiety and neuroticism for FDM patients (Enders et al., 2011). However, the above studies were limited in their exploration of various types of psychological condition, while the first two were partially based on non-standardized questionnaires.

Together with clinical observations, the above evidence suggests that many musicians suffering from FDM are psychologically vulnerable. To date, FDM patients have generally been grouped under the umbrella of a task-specific FD. Attempts of further categorise patients have been based largely on instrumental characteristics (e.g. embouchure vs. hand dystonia), or specific movement patterns (flexion vs. extension dystonia, dystonic tremor etc.). Psychological differences have never been systematically considered to explain possible heterogeneity among FDM patients. This is particularly surprising given that the success of different treatment strategies, such as injection therapy with Botulinum-Toxin (BTX) or behavioural therapy, may well depend on different psychological traits (van Vugt, Boulet, Jabusch, & Altenmüller, 2014).

Therefore, the current study aimed for the first time to classify FDM patients according to psychological characteristics. Our paradigm was designed to investigate anxiety, perfectionistic and stress coping features, firstly between HM and FDM patients (Study I), and secondly among FDM patients only (Study II). We predicted that in Study I FDM patients would be characterised by different psychological profiles compared to HM, whereas Study II would reveal the psychological heterogeneity among FDM patients.

2. Material and methods

2.1. Participants

Musicians from the outpatient clinic of the Institute of Music Physiology and Musicians’ Medicine (IMMM), Hanover University of Music, Drama and Media, who had been diagnosed with FHD were invited to participate. FHD is typically characterised by cramps or curling of one or more fingers, involuntary spasms and loss of coordination which leads to deterioration of quality when performing a musical instrument. HM were randomly recruited freelancers or members of orchestras, music schools or music universities, and formed a control group. Participants who reported any additional neurological (e.g. secondary dystonia, tremor) or psychiatric disease (e.g. depression) were excluded. All participants were German speakers.

For Study I, 24 HM (age: M = 39.7, SD = 8.8) were compared with 24 FDM patients (age: M = 41.1, SD = 9.1), matched as closely as possible for age and gender. The time since the onset of dystonia varied from 0 to 21 years (M = 6.9, SD = 5.8). Three of the patients reported that their symptoms first appeared during activities other than those related to playing their musical instrument. Thirty-five FDM patients (age: M = 45.5, SD = 10.8) participated in Study II. The duration from the onset of dystonia for these patients varied from 0 to 31 years (M = 8.2, SD = 7). Six of these patients reported that their symptoms first appeared during non-musical activities. For further details concerning participants’ characteristics, see Table 1.

2.2. Instruments and procedure

All subjects filled out questionnaires with information about the development of their movement disorder, their medical family history and their musical background. Additionally, three psycho-diagnostic standardised questionnaires were administered to explore participants’ psychological profiles. The Competitive Trait Anxiety Inventory (CTAI), (GER: Wettkampf-Angst-Inventar Trait (WALT-T); Brand, Ehlerspiel, & Graf, 2009), which is widely used in sport science, was used to assess trait anxiety in competition (performance) situations. It consists of 12 items, scored on a 4-point Likert scale (1 = “not at all” to 4 = “very much”) and is divided in three subscales with four items each: (i) the “somatic anxiety” (CTAIsdc) subscale which focuses on physical stress symptoms such as shaky feeling, elevated heart rate etc, (ii) the “self-doubt concern” (CTAIsdc) subscale with statements such as “before competition/performance I am concerned to fail under pressure”, and (iii) the “concentration problems” (CTAIsdc) subscale with statements such as “before competition/performance I am prone to distractions”. The selection of this questionnaire was based on the fact that making music professionally is highly competitive and can occur both individually (e.g. as a soloist) and as part of a team (e.g. as an orchestra member). Music performance requires highly demanding physical and mental training and mostly takes place in stressful environments
Table 1
Participants’ characteristics.

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Study I</th>
<th>Study II</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>HM (n=24)</td>
<td>FDM (n=24)</td>
</tr>
<tr>
<td>Gender: male/female (n)</td>
<td>26/54</td>
<td>26/61</td>
</tr>
<tr>
<td>Age started playing the instrument: (M ± SD)</td>
<td>17/4</td>
<td>28/7</td>
</tr>
<tr>
<td>Affected hand: right/left (n)</td>
<td>–</td>
<td>9.2 ± 2.8</td>
</tr>
<tr>
<td>Cumulative hours of experience: (M ± SD)</td>
<td>38368 ± 16751</td>
<td>30672 ± 18399</td>
</tr>
<tr>
<td>Onset age of dystonic symptoms: (M ± SD)</td>
<td>34.2 ± 8.76</td>
<td>374 ± 10.9</td>
</tr>
<tr>
<td>Occupation: professional player/other (%)</td>
<td>100/0</td>
<td>62.5/37.5</td>
</tr>
<tr>
<td>Musical genre: classical/jazz, rock, pop/other (%)</td>
<td>100/0/0</td>
<td>79.2/20.8/0</td>
</tr>
<tr>
<td>Instrument distribution (per instrument family)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Keyboards (n/%)</td>
<td>11/45.8</td>
<td>16/56.7</td>
</tr>
<tr>
<td>Strings (n/%)</td>
<td>6/25</td>
<td>1/4.2</td>
</tr>
<tr>
<td>Woodwinds (n/%)</td>
<td>4/16.7</td>
<td>1/2.9</td>
</tr>
<tr>
<td>Brass (n/%)</td>
<td>3/12.5</td>
<td></td>
</tr>
<tr>
<td>Plucked (n/%)</td>
<td>–</td>
<td>5/20.8</td>
</tr>
<tr>
<td>Percussion (n/%)</td>
<td>–</td>
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min = minimum; max = maximum; M = mean; SD = standard deviation; n = number of subjects; \% = percentage.

(Engendörfer, Hodapp, Kreutz, & Bongard, 2006). These characteristics are closely aligned to the performance of athletes (Lacalle, Whipple, & Koestner, 2005; Powell, 2004). Two minor adaptations to the content of the CTI were required. First, any occurrence of the word “competition” was supplemented with the word “performance” (as “competition/performance”) in order to be more relevant to musical activities. Second, item 12 was modified to “… I have concerns to be distracted by heckling or noises from the audience.”

The German version of the Frost Multidimensional Perfectionism Scale (FMPS) (Frost, Marten, Lahart, & Rosenblate, 1990) (GER: Mehrdimensionalen Perfektionismus Skala von Frost (MPS-F); Altstötter-Gleich & Bergemann, 2006) was used to examine different features of perfectionism. It consisted of six subscales: “concern over mistakes” (CM), “personal standards” (PS), “parental expectations” (PE), “parental criticism” (PC), “doubts about actions” (DA) and “organisation” (O). These covered a total of 35 statements, each of which was evaluated on a 6-point Likert scale ranging from 1 = “does not apply at all” to 6 = “applies very well”.

Finally, the Stress Coping Questionnaire (SCQ) (GER: Stressverarbeitungsfragebogen (SVF-78); Erdmänn & Janke, 2008) was used to assess different ways of coping with stress. The SCQ consists of 78 items, each evaluated on a Likert scale ranging from 1 = “not at all” to 4 = “most likely”. It is divided into 13 subscales: “play down” (PD), “guilt denial” (GD), “distraction” (D), “substitutional satisfaction” (SS), “situation control” (SC), “reaction control” (RC), “positive self-insistence” (PSI), “need for social support” (NSS), “active avoidance” (AA), “flight tendency” (FT), “mental perseveration” (MP), “resignation” (R) and “self-incrimination” (SI).

The first seven subscales (PD, GD, SS, SC, RC and PSI) explore positive stress coping behaviour (stress reduction; SCQ-POS) and the last four subscales (FT, MP, R, SI) cope negative stress coping behaviour (stress increase; SCQ-NEG). For the present study, we use the English questionnaire and subscale abbreviations mentioned above.

Due to the time-demanding nature of filling in the three questionnaires, healthy musicians, who were not part of the IMMM clinic database, were funded with an Amazon voucher as an additional motivation for completion. Questionnaires were available online as well as in paper form. In both versions participants had the possibility of returning to previous questions. Additionally, in order to avoid any fatigue effects the questionnaires could be completed in segments, allowing participants breaks as required, before final submission. Finally, participants were instructed to return or submit the questionnaires within two weeks of first receiving them.

The current study is part of a larger project conducted at the Institute of Music Physiology and Musicians’ Medicine at the Hannover University of Music, Dance and Media, and the Institute of Psychology, German Sport University Cologne. It focuses on the comparison of psychological trigger factors in PD in musicians and athletes (golfers “yips”). The protocol was conducted in accordance with the declaration of Helsinki and with permission of the ethics committee of the board of the German Association of Psychology.

2.3. Data analysis

Multiple comparisons (Mann–Whitney U tests) were used to explore mean differences in all three questionnaires (sum-scales) and their respective subscales between HM and FDM patients. An exploratory cluster analysis was conducted in order to classify participants according to common psychological characteristics (hierarchical clustering, K-means). Resulting subgroups (clusters) were further compared for possible differences, for example in age, years of experience, etc. (Mann–Whitney U tests and Kruskal–Wallis). The proportion of HM and FDM patients within clusters was compared using a Chi-square test. Correlations between psychological features or other variables of interest were also performed (Spearman’s rho, r, two-tailed). The significant level of acceptance was p = .05 and Bonferroni corrections were applied as required in order to prevent inflated type I error. Effect size was estimated by the use of Pearson’s correlation coefficient, r. Data analyses were performed in IBM SPSS Statistics software package (version 21) and R (version 2.15.1).

3. Results

3.1. Study I

Comparisons between HM and FDM patients revealed no significant differences for any of the three psycho-diagnostic questionnaires, neither for sum-scales (p accepted at .05/4 sum-scales = .0125, Bonferroni-corrected) nor for subscales (p accepted at .05/22 subscales = .0022, Bonferroni-corrected) (Table 2).

For the cluster analysis all musicians were grouped together and all 22 subscales were used as single variables. First, all variables were correlated with each other in order to ascertain whether any substantial collinearities existed. All bivariate correlations indicated an r < .741, consequently there was no need for any variable to be excluded. Afterwards, all scores were standardised to z-values due to the different Likert point-scales used between the questionnaires. A hierarchical clustering analysis (Ward’s method – Squared Euclidean distance) was used to estimate the number of possible clusters (Punjil & Stewart, 1983; see Supplementary appendix, left dendrogram). Results indicated that the division of all participants in two possible clusters. Different clustering procedures, algorithms and distance measures revealed similar clustering patterns, indicating the stability of the results. Consequently, a K-means classification analysis based on two clusters was followed in order to identify cluster centroids and classify participants accordingly. Reliability was tested by a cross-tabulation percentage agreement between two randomly divided subsamples (split-half) of the original data. It indicated a Rand index (RI) of .66 (Rand, 1971), and an Adjusted Rand Index (ARI) of .21 (Hubert & Arabie, 1985).

Follow-up Mann–Whitney U tests (p accepted at < .05/22 subscales = .002, Bonferroni-corrected) between the two clusters indicated eight specific variables (subscales) out of the 22 as the primary contributors for the classification of all participants into those who revealed high
scores (Cluster 1) and those with low scores (Cluster 2). These subscales are: “self-doubt concern” (CTAIsdc), “concern over mistakes” (CM), “somatic anxiety” (CTAIsa), “resignation” (R), “flight tendency” (FT), “personal standards” (PS), “mental perseveration” (MP) and “self-incrimination” (SI) (Fig. 1). Correlational analyses among all the above primary characteristics found that out of 28 combinations, only one was not significant (CTAIsa vs MP, $r_{s} = .263, p = .071$). The remainder of the combinations were positively correlated ($r_{s} = .295$ to $r_{s} = .741, p < .05$).

According to the above results the two clusters were labelled as “high psychological effects” (HPE) subgroup (Cluster 1), and “low psychological effects” (LPE) subgroup (Cluster 2). The subgroups did not differ significantly in age ($Mdn$ HPE: 43, LPE: 40, $U = 210, z = -1.144, p > .05$; age of having started playing their musical instruments ($Mdn$ HPE: 7, LPE: 9.5, $U = 211, z = -1.136, p > .05$; or cumulative hours of practice ($Mdn$ HPE: 29834, LPE: 33032, $U = 249, z = -3.313, p < .05$).

There was a significant association between the type of musician (HM or FDM) and their psychological classification (HPE or LPE subgroup), $\chi^{2} (1) = 7.38, p = .007$ (2-sided) (cross tabulation: $2 \times 2$). The odds of FDM patients being part of the HPE subgroup were 5.9 times higher than those of HM. FDM patients were equally distributed in both subgroups ($\chi^{2} (1) = 1.19, p = .23$). The majority of HM belonged to the LPE subgroup, $\chi^{2} (1) = 10.667, p = .001$ (Table 3).

### 3.2. Study II

Results of the first study (i.e. the equal distribution of FDM patients into HPE and LPE) led to a more detailed exploratory cluster analysis among a larger group of FDM patients only ($n = 35$). Again all 22 subscales were used as single variables. Correlations revealed no substantial collinearity between them, all $r_{s} < .717$. Hence all scores were standardised to z-values. Hierarchical clustering analysis (Ward’s method – Squared Euclidean distance) yielded the existence of two distinct clusters (see Supplementary appendix, right dendrogram). Different clustering procedures, algorithms and distance measures revealed similar clustering patterns, indicating the stability of our results. Therefore, a $K$-means classification analysis based on two clusters was conducted in order to classify participants accordingly. Reliability, which was tested by a cross-tabulation percentage agreement between two randomly divided subsamples (split-half) of the original data, indicated a RI of .62 and an ARI of .015.

Follow-up Mann–Whitney $U$ tests ($p$ accepted at $< .05/22$ subscales = .002, Bonferroni-corrected) between the two resulting clusters indicated seven specific variables (subcales) as the primary contributors for the classification of all FDM patients to those with high scores (Cluster 1) and those with low scores (Cluster 2). These subscales are: “mental perseveration” (MP), “concern over mistakes” (CM), “self-incrimination” (SI), “self-doubt concern” (CTAIsdc) “somatic anxiety” (CTAIsa), “resignation” (R), “flight tendency” (FT), “personal standards” (PS).
were averaged together into a single variable. The area (grey zone, [GZ]) between the lower 25th percentile of the HPE-FDM and the upper 25th percentile of the LPE-FDM of this new variable could indicate patients who fell in between the two distinct subgroups (Fig. 3). The resulting GZ between the two subgroups could be utilised in order to evaluate and classify single FDM patients as HPE-FDM, LPE-FDM or GZ. Based on the above distinction, FDM patients were distributed as follows. HPE-FDM: n = 14, GZ-FDM: n = 8 and LPE-FDM: n = 13. Further analysis revealed that the three subgroups did not differ significantly in, age, \( H(2) = 1.455, p > .05 \); cumulative hours of practice, \( H(2) = .250, p > .05 \); age at onset of dystonia, \( H(2) = 1.821, p > .05 \); or age of having started playing their musical instruments, \( H(2) = 2.892, p > .05 \).

Apart from the above primary psychological characteristics (variables), additional characteristics (i.e. PS, R, AA, CTAIcp and RC), which contributed less (\( p_{\text{uncorrected}} < .05, r = -.5 \), see Fig. 2) to the classification of FDM patients into two clusters could also be of major importance; especially those characteristics, which were also found to be significantly correlated with the average score (across all FDM patients, \( n = 35 \)) of the above six primary psychological characteristics. These are “personal standards” (PS), \( r_p = .560, p < .001 \) and “concentration problems” (CTAIcp), \( r_p = .492, p = .003; (p \text{ accepted at } < .05/6_{\text{subscals}} = .008, \text{Bonferroni-corrected}). \) The above approach (averaged primary and secondary characteristics) was graphically implemented in a heuristic model (Fig. 4). The importance of this model was firstly to map the psychological profiles of FDM patients and secondly to be used as an experimental diagnostic tool for the psychological classification of individual FDM patients.

Finally, no correlations were found between the duration of dystonia (years after onset) and any of the 22 subscales. This was the case for all FDM patients (\( n = 35 \)), as well as for the HPE-FDM (\( n = 18 \)), and the LPE-FDM (\( n = 17 \)) subgroups separately. Moreover, no mean differences (Mann–Whitney U) in any subscale were found between those patients who did not use BTX (\( n = 17 \)) and those who had been injected with it (\( n = 18 \), usage period in years: \( Mdn = 1, \text{min} = 1, \text{max} = 8 \)). No correlations (Spearman’s rho) were also found between the duration of dystonia and any of the subscales for patients who had been injected with BTX, and for those who did not apart from only one subscale (i.e. R) of the second subgroup which found to be significant, \( r_p = .746, p = .001; (p \text{ accepted at } < .05/22_{\text{subscals}} = .002, \text{Bonferroni-corrected}). \)

**Fig. 1.** Final cluster centres of both subgroups (clusters) (HPE and LPE) derived by clustering all musicians together (HM and FDM patients). Psychological characteristics are ordered (left to right) from those which contributed the most to the classification of all musicians; i.e. primary contributors: \( p < .002, r = -.7 \), secondary contributors: \( p < .05, r = -.4 \), non-contributors: \( p > .05 \).

**Table 3** Distribution of participants.

<table>
<thead>
<tr>
<th>Clusters (subgroups)</th>
<th>Groups</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>HM</td>
</tr>
<tr>
<td>1 (HPE)</td>
<td></td>
</tr>
<tr>
<td>Count (n)</td>
<td>4</td>
</tr>
<tr>
<td>Within subgroups (%)</td>
<td>23.5</td>
</tr>
<tr>
<td>Within groups (%)</td>
<td>16.7</td>
</tr>
<tr>
<td>2 (LPE)</td>
<td></td>
</tr>
<tr>
<td>Count (n)</td>
<td>20</td>
</tr>
<tr>
<td>Within subgroups (%)</td>
<td>64.5</td>
</tr>
<tr>
<td>Within groups (%)</td>
<td>83.3</td>
</tr>
</tbody>
</table>
4. Discussion

The current study attempted to investigate and describe the psychological profiles of FDM patients. We examined possible psychological differences, firstly between HM and FDM patients (Study I) and secondly, among FDM patients only (Study II). An open design exploratory analysis based on clustering procedures was performed in order to detect different psychological types among participants.

4.1. Study I (HM vs. FDM)

Direct comparisons between HM and FDM patients revealed no mean differences for any of the sum- or subscales of the three psycho-diagnostic questionnaires. However, cluster analysis indicated that all musicians could be divided into two different subgroups (clusters) (HPE and LPE), based on contrasting levels of the following eight characteristics; “self-doubt concern” (CTAIsc), “concern over mistakes” (CM), “somatic anxiety” (CTAIa), “resignation” (R), “flight tendency” (FT), “personal standards” (PS), “mental perseveration” (MP) and “self-incrimination” (SI). CTAIsc and CTAIa are competitive trait anxiety characteristics, CM and PS are perfectionistic characteristics, and R, FT, MP and SI are all negative stress coping characteristics.

The first subgroup (HPE) describes musicians who exhibit high levels of the above psychological features. They are characterised by elevated trait anxiety (i.e. psychological state before concerts or various music performances) combined with high personal standards and excessive concerns about mistakes. The last two of these features are well known among professional classical musicians and could also contribute to the reinforcement of anxiety and stress (Wesner, Noyes Jr., and Davis, 1990; Wolfe, 1989). The final characteristic of HPE musicians is their inability to cope with stress.
effectively. A lack of stress coping strategies could lead to a vicious cycle of permanent and uncontrolled elevated anxiety. The second subgroup (LPE) describes personalities, which show mirrored psychological effects to those of HPE musicians. They have fewer indications of any anxiety and perfectionism together with strong stress coping strategies. A large majority (96%) of the above primary characteristics, which describe both psychological profiles of all musicians (HM and FDM), were found to be positively correlated. This indicates the inter-dependency and common behaviour of those features from which the two profiles are constructed.

Concerning the proportion of HM and FDM patients within the two subgroups, we initially observed that both healthy and dystonic musicians could be characterised by both profiles, HPE and LPE. However, the chances of FDM patients falling into the HPE subgroup were found to be 5.9 times greater than those of HM. On the other hand, and as expected, between-subgroups distributions revealed that the majority of HM are characterised by LPE (ratio 5:1), whereas FDM patients are characterised by both HPE and LPE profiles (ratio 1:1). These findings could lead to two conclusions. Firstly, that FDM patients exhibit higher levels of the above anxiety, stress and perfectionistic characteristics than HM and secondly, that FDM patients who are equally distributed across both profiles are characterised by psychological heterogeneity. These findings emphasise the necessity of further and deeper classification of musicians suffering from FD.

The above results fit with previously published research. Jabusch et al. (2004) and Jabusch and Altenmüller (2004) also reported elevated perfectionistic tendencies, anxious traits and somatic complaints for FDM patients compared to HM. However, perfectionism and anxiety were tested based on self-designed questionnaires, therefore lack of consistency and reliability do not allow direct comparisons. On the other hand, Enders et al. (2011) assessed various psychological conditions by the use of standardised questionnaires and also reported higher levels of state and trait anxiety for FDM musicians as compared to HM and non-musicians. However, the above studies explored possible psychological differences only via direct comparisons between HM and FDM patients. In the current study, direct comparisons between these two populations revealed no significant differences. This could be explained by the lack of psychological homogeneity among FDM patients, already described above. Furthermore in the current study, differences between HM vs. FDM emerged after a more detailed exploration based on the classification of musicians according to common psychological traits. Additionally, we believe that awareness of dystonia as a disease has expanded significantly during recent years. The faster and more precise diagnosis of FDM patients is in contrast to earlier prevalence of repeated misdiagnoses, which led to delays, deterioration of symptoms, uncertainty and therefore avoidable anxiety and stress. Summarising the results of the first study, we underline the existence of two different psychological types of FDM patients. The second study of the present paper was conducted in order to explore the suggested psychological taxonomy in a larger group of FDM patients.

4.2. Study II (FDM patients)

The second exploratory analysis indicated that FDM patients could indeed be divided into two different psychological subgroups (HPE-FDM and LPE-FDM), based on contrasting levels of the specific psychological characteristics: “mental perseveration” (MP), “concern over mistakes” (CM), “self-incrimination” (SI), “self-doubt concern” (CTAIstdc) “somatic anxiety” (CTAIns) “doubts about actions” (DA), and “flight tendency” (FT). CM, and DA express perfectionistic characteristics, MP, SI and FT explain negative stress coping characteristics whereas CTAStdsc and CTAIns describe competitive trait anxiety characteristics; an almost identical psychological profile to that of the HPE and LPE subgroups of the first study. The analysis of a larger group of FDM patients supported the main finding from the first study: the existence of two different psychological types of FDM. Moreover the above classification remains highly psycho(bio)logical as both subgroups (HPE-FDM vs. LPE-FDM) are not affected by other factors such as age, hours of experience, age at the onset of dystonia, or age of having started playing their musical instrument.

Research during the last two decades has revealed that basal ganglia circuits are not only involved in motor control functions but are also associated with the learning of complex new behaviours (Graybiel, 2005; Pasupathy & Miller, 2005). These procedures include emotions, motivation and cognition (Everitt, & Robbins, 2005; Shattar, Havazelet-Heimer, Raz, & Bergman, 2003), all involved in the design, expression and execution of goal-directed behaviours (Haber & Calzavara, 2009). It has been suggested that parallel and segregated pathways organised in different regions of the cortico-basal ganglia-thalamic structures are linked to motor, cognitive and limbic networks (Alexander & Crutcher, 1990; Haber & Calzavara, 2009; Haber, Fudge, & McFarland, 2000; Nauta, Smith, Faull, & Domesick, 1978; Pasupathy & Miller, 2005). Consequently, basal ganglia mechanism related to movement executions should be explored from a neuro-psychomotor-cognitive perspective. This holistic approach plays a crucial role firstly in the development of learning new behaviours and secondly in the restoration of old ones. The classification of FDM patients into HPE-FDM and LPE-FDM may explain two different pathophysiological paths to the development of dystonia. The first path (HPE-FDM) may describe a form of dystonia which affects basal ganglia circuits in a holistic, “psychomotoric” manner, meaning that motor circuit dysfunctions of the basal ganglia could also be accompanied by mal-adaptation of emotional-memory pathways via limbic channels. In contrast, in the second approach (LPE-FDM) limbic channels could be less involved in the encoding of dystonia, supporting the idea that dystonia can result from a dysfunction of the pure motor circuits of the basal ganglia. However the exact involvement of each sub-circuit (i.e. motor, emotional or “cognitive”) to the encoding of dystonia remains difficult to specify and these factors could have differential impacts on individual patients. This could also explain why uniform (e.g. only BTX) or multiformal (e.g. psychotherapy, retraining etc.) methods applied to FDM patients have been only partially effective and highly individualistic to date (Schuele & Lederman, 2004; van Vugt et al., 2014).

The above classification concerning the development of dystonia in musicians should contribute not only to a more precise diagnosis, but will be particularly relevant to treatment. The heuristic model (Fig. 4) derived from the current analysis could be used for the taxonomy of individual FDM patients into HPE-FDM or LPE-FDM simply by averaging the specific primary correlated characteristics. Two supplementary psychological characteristics, which could accompany the main profiles of both subgroups, have also been provided. Finally, a grey zone suitable for classifying patients who may fall between the two distinct subgroups, has been established. However, the above model which could comprise an additional diagnostic tool for the classification of individual FDM patients, could be further reinforced and developed by including additional psychological characteristics. For clinical applications, a new questionnaire based on the current primary characteristics should be designed, standardised and validated. The precise psycho-diagnostic classification of FDM patients into HPE-FDM or LPE-FDM could also specify the treatment methods which should be applied. For example HPE-FDM patients may benefit more from psychological treatment methods such as psychotherapy and retraining sessions combined with...
injections of BTX etc. whereas treatments for LPE–FD patients could focus on neurological methods such as BTX and anticholinergic drugs.

Previous studies with psychological factors in these patients have suggested that higher levels of stress, anxiety, and perfectionism in musicians could be additional risk factors in the development of FD (Enders et al., 2011; Jabusch et al., 2004; Jabusch & Altenmüller, 2004). However, the current study also indicated that FD patients are more likely to exhibit HPE than HM. Nevertheless the current findings suggest that the above assumption should not be applied for all FD patients. Results derived from the first study indicated that half of FDM patients share the same psychological profile as HM. Therefore, we suggest that HPE among musicians could indeed be an aggravating risk factor to the development of FDM, but at the same time the manifestation of dystonia could develop in musicians with no signs of any psychological abnormalities. Heterogeneity among patients was also reported by Fabbriini et al. (2010) who examined psychiatric disturbances in patients with other types of primary FDs. They reported that only 57.3% of those patients were diagnosed with additional psychiatric disorders. Similarly, Munhoz et al. (2005), who examined obsessive–compulsive disorders (OCD), reported no group differences between patients with blepharospasm and hemifacial spasm. However OCD were found in 66.6% and 70% of the patients with blepharospasm and hemifacial spasm respectively. These findings emphasise the necessity for further classification and more precise diagnoses in all forms of primary dystonias.

An important suggestion by all previous studies dealing with psychopathology of FDM (Enders et al., 2011; Jabusch et al., 2004; Jabusch & Altenmüller, 2004) is that anxiety and/or other psychological features may not be a psychoreactive phenomenon; instead anxiety characteristics are likely to pre-exist in musicians before the onset of dystonia. Support for this view is also found in our study; firstly in the fact that no direct psychological differences were found between HM and FDM patients and secondly in the lack of correlations between the duration of dystonia and any of the psychological characteristics. Only a few marginal correlations were detected which did not provide any evidence of a psychoreaction due to the onset of dystonia. Similar results have been found in investigations of other types of primary dystonia, such as that of Fabbriini et al. (2010) who reported that psychiatric disorders pre-dated the onset of dystonia.

With regard to psychological measurements after the onset of dystonia, the effect of BTX treatment on patients’ mood should be considered. Previous studies reported an alleviation of anxiety (Grafman, Cohen, & Hallett, 1991; Liu et al., 1998) and depression (Barahona-Corrêa, Bugalho, Guimarães, & Xavier, 2011; Jahanshahi & Marsden, 1992; Müller et al., 2002) in FD patients after treatments with BTX. Only 42% of our FDM sample used BTX, most of these patients having been treated within a timeframe of 1–2 years. These psychological improvements cannot be attributed to BTX-treatment in the current study; firstly, no differences were found in any psychological characteristic between those FDM patients who used and those who did not use BTX and secondly, no important correlations for either of these two groups were found between any psychological characteristic and the duration of dystonia. However further investigations with larger sample sizes are needed to clarify the possible effects of BTX.

Two limitations of the present study should be also noted. Firstly, the number of participants per group might be considered relatively small. It is, however, the case that FDM is a rare disease among professional musicians, and even with access to the large database provided by the IMMML clinic it is very difficult to achieve higher numbers of patients per group. Nevertheless, the effect sizes presented above are relatively large and unlikely to be the result of unrepresentative sampling. Secondly, the psychological classification of all patients in the current study is based on subjective measurements. Following the classification guidelines suggested above will provide a more objective assessment of the phenotype and the symptomatology of FHD in the future.

5. Conclusion

The current study was an open design exploratory investigation of the psychological profile of FDM. Psycho-diagnostic questionnaires explored anxiety, perfectionism and stress coping strategies. Musicians (HM and FDM patients) were found to be sub-divided into two subgroups (HPE and LPE) with contrasting levels of several psychological characteristics. FDM patients were found to have higher chances of being classified as HPE musicians, compared to HM. However, FDM patients were equally distributed across subgroups (HPE and LPE), which was not the case for HM. Therefore, the current study suggests the existence of two different psychological types of FDM patients and emphasises the necessity of further diagnostic classification. The next steps in this project will be to evaluate the phenotype of FHD in the two FDM subgroups (HPE-FDM and LPE-FDM) and to assess the alleviation of their dystonic symptoms after the application of specifically designed interventions.

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Appendix A. Supplementary information

Supplementary data associated with this article can be found in the online version at http://dx.doi.org/10.1016/j.neuropsychologia.2014.05.014.

References


