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Tic-reducing effects of music in patients with Tourette's syndrome: Self-reported and objective analysis

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ABSTRACT

Background: Self-reports by musicians affected with Tourette's syndrome and other sources of anecdotal evidence suggest that tics stop when subjects are involved in musical activity. For the first time, we studied this effect systematically using a questionnaire design to investigate the subjectively assessed impact of musical activity on tic frequency (study 1) and an experimental design to confirm these results (study 2).
Methods: A questionnaire was sent to 29 patients assessing whether listening to music and musical performance would lead to a tic frequency reduction or increase. Then, a within-subject repeated measures design was conducted with eight patients. Five experimental conditions were tested: baseline, musical performance, short time period after musical performance, listening to music and music imagery. Tics were counted based on videotapes.
Results: Analysis of the self-reports (study 1) yielded in a significant tic reduction both by listening to music and musical performance. In study 2, musical performance, listening to music and mental imagery of musical performance reduced tic frequency significantly. We found the largest reduction in the condition of musical performance, when tics almost completely stopped. Furthermore, we could find a short-term tic decreasing effect after musical performance.
Conclusions: Self-report assessment revealed that active and passive participation in musical activity can significantly reduce tic frequency. Experimental testing confirmed patients' perception. Active and passive participation in musical activity reduces tic frequency including a short-term lasting tic decreasing effect. Fine motor control, focused attention and goal directed behavior are believed to be relevant factors for this observation.

1. Introduction

Tourette's syndrome (TS) is a neuropsychiatric disorder characterized by multiple motor tics and at least one vocal tic [1]. Moreover, a large number of patients report to have premonitory urges (sensations) [2–9], also called sensory tics [10], that are perceived directly prior to the execution of a motor or vocal tic. Tics can be triggered not only by emotional stimuli like anxiety [11–13], anger [10], aggression [10] and excitement [11,12,14] but also by boredom [11] or fatigue [11,15,16]. Stress has also been to be a factor worsening tics [11,13,15–17]. The influence of stress, however, is currently a matter of debate. In a systematic study, Conelea et al. (2014) examined tic frequency during periods of heightened and lowered physiological arousal. An increase of tic frequency during periods of heightened physiological arousal was not found in that particular study. Contextual factors have furthermore been reported leading to tic reduction [18]. These include inter alia relaxation [11,15,17,19], distraction [17], sports [12,15,16,19], habitual, automatic actions [20], attention and concentration [11,15–17]. An overview of the different environmental factors having tic-reducing effects is given in a review by Conelea and Woods (2008) [21].

Music seems to have a positive effect on tics, too. There are anecdotal reports of patients reporting about the tic-reducing effect while playing a musical instrument or listening to music [11,22–27]. Furthermore, there are self-reports by professional musicians who had made the experience that tics stopped when they were involved in musical activity, most often when playing an instrument, singing, or composing. Nick van Bloss, for example, noted that he would have no tics when he was playing the piano [27,28]. Similar experiences were reported by James McConnel [29], Michael Wolff [30], David Aldridge [31], Jason Duika [32], Luke Parkin [33] and Jim Couchenour [34]. However, to our knowledge, no previous study has investigated this tic decreasing effect of music systematically.

There is also still a lack of evidence which musical aspects by the inherent nature of music itself like rhythm, timbre, harmony or dynamics or which musical preferences (familiarity) and abilities (musical imagination) do have an influence on tic reduction. There is also a lack

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of knowledge regarding which personal factors (alertness of the recipients/musicians; tension) and which environmental contexts (listening to music/musical performing in groups) and crucial for tic-reducing effects.

The present investigation consists of two studies: study 1 evaluated firstly the subjectively assessed effect of music on tic frequency. Two hypotheses were tested: Tic frequency in patients with TS is reduced during (1) music listening and (2) during musical performance. Study 1 evaluated secondly in an exploratory analysis those musical factors that lead to tics frequency reduction. This was done in order to receive new hypotheses, that will be tested in a future study.

In study 2, effects of musical activity in tic frequency reduction were objectively investigated using an experimental design. Four hypotheses were tested: (1) musical performance reduces tic frequency; (2) musical performance has a short-term effect on reducing tics; (3) mental imagery of musical performance; and (4) listening to music reduces tic frequency.

2. Methods

2.1. Study 1

2.1.1. Subjects

A total of 29 German subjects (13 female, mean age: 35.8 years ± 11.6 $R = 14–55$) were recruited from self-help groups and via Internet. Inclusion criteria were a former diagnosis of TS by a neurologist or a psychiatrist, respectively. Musicians and nonmusicians from 14 years on were included. Both studies were approved by the review board of the Department of neurology, University of Münster. All participants gave their written informed consents together with sending back the questionnaire.

2.1.2. Questionnaire

The two hypotheses were tested using a self-developed questionnaire. The patients had to assess the effect of both listening to music, instrumental play, and singing on tics: tic frequency decreases, remains constant or increases.

For the exploratory analysis in study 1, three test batteries 'music listening' (18 items), 'playing a musical instrument' (20 items) and 'singing' (20 items), were presented (see Table 1). Scoring was identically to the preceding part in study 1. For the statistical analysis, the batteries 'playing a musical instrument' and 'singing' were put together into one battery 'musical performance'. When a patient both played an instrument and sang, the ratings were scored twice.

2.1.3. Statistical analysis

For testing the two hypotheses (one-tailed testing) and for the data analysis of the exploratory study (two-tailed testing), a chi-square test was applied. An alpha level of .05 was used for all statistical tests of both studies.

2.2. Study 2

2.2.1. Subjects

Eight German subjects (4 female, mean age = 37.8 years ± 13.0, $R = 17–50$) were recruited from a German self-help group via Internet and local print media. Inclusion criteria were former diagnosing of TS by a neurologist or a psychiatrist. Additionally, only patients from 14 years on with musical experience of 2 years minimum were included. Informed consent was obtained from all individual participants included in the study. For patients under the age of 18 years parents provided additionally written informed consent,

Table 1
Explorative analysis of the test batteries 'listening to music' and 'musical performance' of the study 1 sample, $N = 29$.

Item no.	Items test battery 'listening to music'	Items test battery 'musical performance'
01	Listening to loud music ns	Performing loud music $\chi^2(2, N = 27) = 13.56, p = .001$
02	Listening to soft music $\chi^2(2, N = 28) = 7.36, p = .025$	Performing soft music $\chi^2(2, N = 27) = 14.89, p = .001$
03	Listening to fast music ns	Performing fast music $\chi^2(2, N = 27) = 8.22, p = .016$
04	Listening to slow music ns	Performing slow music $\chi^2(2, N = 27) = 18.67, p < .001$
05	Listening to boring music ns	Performing boring music ns
06	Listening to exciting music ns	Performing exciting music ns
07	Listening to calming music $\chi^2(1, N = 28) = 24.14, p < .001$	Performing calming music $\chi^2(2, N = 27) = 32.89, p < .001$
08	Listening to pleasant music $\chi^2(2, N = 29) = 31.93, p < .001$	Performing pleasant music $\chi^2(2, N = 27) = 24.22, p < .001$
09	Listening to unpleasant music ns	Performing unpleasant music ns
10	Listening to my favorite music $\chi^2(2, N = 29) = 28.41, p < .001$	Performing my favorite music, $\chi^2(2, N = 27) = 28.22, p < .001$
11	Listening to new music ns	Performing new music $\chi^2(2, N = 28) = 7.36, p = .025$
12	Listening to familiar music $\chi^2(2, N = 27) = 14.22, p = .001$	Performing familiar music $\chi^2(2, N = 27) = 21.56, p < .001$
13	Listening when resting $\chi^2(1, N = 29) = 5.83, p = .016$	Performing when resting $\chi^2(2, N = 28) = 26.64, p < .001$
14	Listening when being tired ns	Performing when being tired $\chi^2(2, N = 27) = 6.22, p = .045$
15	Listening when being alone $\chi^2(2, N = 29) = 13.52, p = .001$	Performing when being alone $\chi^2(2, N = 28) = 23.21, p < .001$
16	Listening together with others	Performing together with others $\chi^2(2, N = 26) = 12.08, p = .002$
17	Listening when being stressed	Performing when being stressed
18	Listening when being relaxed $\chi^2(1, N = 29) = 9.97, p = .002$	Performing when being relaxed $\chi^2(2, N = 27) = 24.89, p < .001$
19	–	Performing at public gigs, ns
20	–	Imagine yourself performing $\chi^2(2, N = 28) = 14.00, p = .001$

ns = not significant.

2.2.2. Equipment

The equipment for the procedure included a camcorder installed atop a tripod to record the patients in all experimental conditions, an MP3-Player and earphones for the condition 'music listening'.

2.2.3. Test material

A new questionnaire was constructed to evaluate personal, clinical and musical data of the patients. To measure the subjectively perceived general effect of both, playing an instrument/singing and listening to music, patients had to consent to the items 'tic frequency decreases', 'tic frequency remains constant' and 'tic frequency increases', respectively. Finally, participants were asked to list further emotional, personal or environmental factors that could have a decreasing effect on tics. Severity of tics was rated using the Yale Global Severity Scale (YGTSS) [35] and tic frequency per minute was assessed applying the modified Rush Video-Based Tic Rating Scale (mRVRS) [36].

2.2.4. Design

We used a repeated measures within-subject design, including a baseline and four experimental conditions: (a) baseline: subjects sat on a chair in a quiet room facing the camera, feet on the floor and hands in lap; (b) musical performance: subjects played their musical instrument or sang a piece of their choice; (c) subjects sat in silence for 15 min after musical performance (short-term effect) in the same position as in the first condition, (d) mental imagery of musical performance: subjects were instructed to imagine the same musical piece as