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Analysis of dystonic tremor in musicians using empirical mode decomposition

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HIGHLIGHTS

- This is the first study, objectively assessing finger tremors in musicians during movements.
- We applied a novel method that allows for the separation between physiological and pathological tremor.
- We did not find dystonic tremor in musicians dystonia (a disorder with the loss of motor control), indicating that tremor is not associated more frequently here than in healthy musicians.

ABSTRACT

Objective: Test the hypotheses that tremor amplitude in musicians with task-specific dystonia is higher at the affected finger (dystonic tremor, DT) or the adjacent finger (tremor associated with dystonia, TAD) than (1) in matched fingers of healthy musicians and non-musicians and (2) within patients in the unaffected and non-adjacent fingers of the affected side within patients.
Methods: We measured 21 patients, 21 healthy musicians and 24 non-musicians. Participants exerted a flexion–extension movement. Instantaneous frequency and amplitude values were obtained with empirical mode decomposition and a Hilbert-transform, allowing to compare tremor amplitudes throughout the movement at various frequency ranges.
Results: We did not find a significant difference in tremor amplitude between patients and controls for either DT or TAD. Neither differed tremor amplitude in the within-patient comparisons.
Conclusion: Both hypotheses were rejected and apparently neither DT nor TAD occur in musician's dystonia of the fingers.
Significance: This is the first study assessing DT and TAD in musician's dystonia. Our finding suggests that even though MD is an excellent model for malplasticity due to excessive practice, it does not seem to provide a good model for DT. Rather it seems that musician's dystonia may manifest itself either as dystonic cramping without tremor or as task-specific tremor without overt dystonic cramping.

1. Introduction

Tremor is one of the most common movement disorders, characterized by an involuntary, oscillatory and rhythmic movement with a heterogeneous etiology. It may occur in dystonic syndromes where it has been defined as dystonic tremor (DT) with three characteristics (Deuschl et al., 1998): (1) Mostly postural- and action tremor in one body part affected by dystonia, (2) focal tremor with an irregular amplitude, and (3) a frequency mostly below 7 Hz. Three forms of DT syndromes have been described: (1) DT as a tremor occurring in a body part affected by dystonia, (2) tremor associated with dystonia (TAD), which occurs in body parts not affected by dystonia, and (3) dystonia-gene associated tremor, referring to tremor in patients with a positive family history of dystonia (Deuschl et al., 1998).

In musician's dystonia (MD) we could observe a movement-induced tremor of the affected fingers or the finger(s) adjacent to

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the affected fingers (adjacent finger(s)) during clinical examination when the patients exerted a slow flexion–extension movement of the respective finger. No tremor was observed at the other finger(s) of the affected side. During the movement, tremor amplitude appeared to change depending on the position of the finger during the movement. This observation is in accordance with observations by Gironell and Kulisevsky (2009), who reported position dependence of DT, with tremor occurring predominantly in a position that is opposite to the pulling direction of dystonia and disappearing when the finger is placed at a position "where dystonia wants to pull it".

We considered movement-induced tremor at the fingers affected by dystonia as DT and tremor at the adjacent fingers TAD. Adjacent fingers were those fingers next to the affected fingers, i.e. for an affected index finger, the adjacent finger was the middle finger and for an affected ring finger, the adjacent fingers were the middle and little finger. In 2004 criteria for DT were established that allow a distinction between definite DT, probable DT and possible DT (McAuley and Rothwell, 2004). We therefore considered the observed tremor as probable DT/TAD, since it was an irregular unilateral tremor of the finger that appeared movement specific (rather than position specific). For embouchure dystonia, another form of MD, tremor has been described before (Frucht et al., 2001; Torres-Russotto and Perlmutter, 2008). However, phenomenological characteristics of DT or TAD in MD of the upper limb have not been described in detail before. The aim of this study was thus to assess, whether the tremor observed during a flexion–extension movement of the affected or adjacent fingers as defined above in MD are a specific characteristic for patients with MD as compared to healthy musicians or healthy non-musicians and whether it is position specific. For this we tested the first hypothesis (between groups) whether there is a significant difference in tremor amplitude between the affected fingers ($\triangleq$DT) or the adjacent fingers ($\triangleq$TAD) of patients displaying tremor during clinical examination as compared to matched fingers of healthy musicians and healthy non-musicians. Secondly, we were interested in assessing whether tremor amplitude is higher in the affected and adjacent fingers as compared to the unaffected and non-adjacent fingers of the affected side (remaining fingers). For this we tested the second hypothesis that there is a significant within-patient difference in tremor amplitude between the affected and adjacent fingers, respectively, as compared to the remaining fingers of the affected side. If such tremor existed, an interesting question would be, whether this phenomenon may have a prognostic relevance. This would have to be investigated in further studies comparing the course of the disease between those who display tremor and those who don't.

2. Methods

The study was approved by the local ethics committee and conducted according to the Declaration of Helsinki.

2.1. Participants

Twenty-three patients who presented to our outpatient clinic with primary MD in at least one finger and who displayed movement induced tremor in the affected or adjacent finger during clinical examination were included. Due to technical problems during the data acquisition two patients were excluded, reducing the patient group to 21 (17 male, 4 female; mean age 44.3 ± 9.2 years; mean disease duration 7.8 years ± 7.2). The dystonic finger was identified at the instrument during clinical examination. The patients had not received Botulinumtoxin-A for at least three months prior to tremor measurement. 17 of the patients were classical musicians, 2 were jazz musicians, and 2 played both styles. The daily amount of practice was 2.8 h (±1.4). Only those patients with a negative history for other neurological diseases were included. In 10 patients the right hand and in 11 patients the left hand was affected by dystonia. In 4 patients two fingers were affected. Patient data are summarized in Table 1.

Two control groups were included, the first consisting of 21 healthy musicians (13 male, 8 female; mean age 28.4 ± 9.3 years) and the second of 24 healthy non-musicians (age 28.0 ± 11.3). From the musicians control group two participants had to be excluded due to technical problems during the data acquisition, reducing its size to 19.

2.2. Measurement

Tremor was measured with a calibrated 3D accelerometer (biovision, Wehrheim, Germany, 8 × 8 × 11 mm; 4 g; DC – 500 Hz; max 50 g) and Ag–AgCl-surface-EMG (biovision, Wehrheim, Germany). The EMG surface electrodes were attached above the finger flexors and extensors of the forearm; the accelerometer was placed on the fingernail of the finger to be measured, with the *z*-axis in the direction of the flexion–extension movement. The focus of the current study is on the accelerometer signals; the EMG measurements will be considered elsewhere.

For examination patients were seated in a comfortable chair. Their forearm was placed on a comfortable armrest and they held their hand with the palm facing upwards. Digits II to V of both hands were measured separately in randomized order. Participants were asked to exert a 180° flexion–extension movement of the finger, starting from an extended position of the finger and instructed to move 4 s in each direction paced by a metronome set to 60 bpm. Thus, the duration of a complete flexion–extension cycle was 8 s. During the movement patients were asked to allow for any passive movement of other fingers, since the attempt to stabilize them in a certain position would have induced artificial cocontraction and therefore confounded the measurement. The recording duration for each finger was 3.5 min, yielding to a total of about 26 flexion–extension cycles per finger.

2.3. Data processing

It is known that pathological tremor is a nonlinear process (Gantert et al., 1992; Deuschl et al., 1998). Furthermore, based on clinical observation we expected that dystonic tremor would be non-stationary and position dependent. Given these properties we applied empirical mode decomposition (EMD) (Huang et al., 1998), a data-driven signal-processing technique suitable for nonlinear and non-stationary signals. It has also been shown that with EMD voluntary movement can be distinguished from tremor (de Lima et al., 2006; Gallego et al., 2011). In a sifting process, acquired signals are decomposed into basic components, called intrinsic mode functions (IMF), which are used to identify distinct frequency bands (de Lima et al., 2006; Silchenko et al., 2010; Li et al., 2012) and may be related to biological phenomena like tremor. It has thus been applied in recent tremor research (Rocon et al., 2006; Silchenko et al., 2010; Li et al., 2012; Lee et al., 2013).

The accelerometer signal was processed as follows (see Fig. 1 for an overview). (1) The inclination angle of the finger (flexion–extension) was calculated via a four-quadrant inverse tangent (atan2 function in Matlab) of the z- and the y-components of the accelerometer (low-pass filtered at a cut-off frequency of 2 Hz in order to isolate the voluntary part of the finger movement). (2) The tremor signal was extracted by applying a band-pass filter to the z-component of the accelerometer (1–50 Hz, 4th order butterworth, applied back-and-forth to obtain zero phase shift). (3) EMD was performed in Matlab using the EMD package by Rilling et al.