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Electrophysiological characteristics of task-specific tremor in 22 instrumentalists

André Lee · Kenta Tominaga · Shinichi Furuya ·
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Abstract Our aim was to address three characteristics of task-specific tremor in musicians (TSTM): First, we quantified muscular activity of flexor and extensor muscles, of coactivation as well as tremor acceleration. Second, we compared muscular activity between task-dependent and position-dependent tremor. Third, we investigated, whether there is an overflow of muscular activity to muscles adjacent to the affected muscles in TSTM. Tremor acceleration and muscular activity were measured in the affected muscles and the muscles adjacent to the affected muscles in 22 patients aged 51.5 ± 11.4 years with a task-specific tremor. We assessed power of muscular oscillatory activity and calculated the coherence between EMG activity of affected muscles and tremor acceleration as well as between adjacent muscles and tremor acceleration. This was done for task-dependent and position-dependent tremor. We found the highest power and coherence of muscular oscillatory activity in the frequency range of 3–8 Hz for affected and adjacent muscles. No difference was found between task-dependent and position-dependent tremor in neither power nor coherence measures. Our results generalize previous results of a relation between coactivation and tremor among a variety of musicians. Furthermore, we found coherence of adjacent muscles and TSTM. This indicates that overflow exists in TSTM and suggests an association of TST with dystonia.

Keywords Dystonia · Coherence · EMG · Coactivation · EMG overflow

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Introduction/background

Task-specific tremors (TST) occur predominantly during a certain task (Deuschl et al. 1998). The most common form is primary writing tremor (PWT), for which two types have been described (Bain et al. 1995): Type A TST, induced by writing (task-induced); and type B TST, induced when holding the arm in the position for writing (position-sensitive). It may occur in sports or in certain professions (e.g., dentists, hair-dressers, musicians) (Rosenbaum and Jankovic 1988; Bain 2011; Roberts et al. 2013). Recent studies have described a TST in high string instrumentalists (violinists and viola) (Lederman 2010; Lee and Altenmüller 2012) that appeared in the right bowing arm while playing the instrument and was thus named primary bowing tremor (PBT). A frequency range of 3–8 Hz with a mean frequency of around 6 Hz was observed.

To understand its pathophysiological mechanism of PWT, muscular activities have been investigated. The main findings were phasic activity in intrinsic hand and forearm muscles as well as a coactivation of antagonist muscles

(Elble et al. 1990; Bain et al. 1995). In a previous study, we found a coactivation of antagonist muscles in PBT in four patients (Lee et al. 2013b) and determined a direct relationship between coactivation and tremor acceleration (Lee et al. 2014b). To assess whether phenomenology of TST occurs independent from the instrument, we wanted to extend these findings to TST in other instrumentalists. The first aim of this paper was to assess muscular activities and its relationship to tremor-related oscillation in 23 musicians with TST. For this, we assessed which body part was affected by tremor during clinical examination. We quantified the amount of coactivation using a sum of agonist–antagonist activities (A–A sum), which was recently developed (Inoue et al. 2011; Ariga et al. 2012).

Since to our knowledge type A and type B tremor have not been quantitatively compared in TSTM, our second aim was to quantify type B tremor and compare it to type A tremor with regard to frequency and amplitude as well as coherence between tremor oscillation and muscular activities. With regard to frequency, we assessed whether it is lower in type A tremor as compared to type B tremor. The rationale behind it are findings that physiological tremor at the hand/finger is lower in a task that involves movement than during an isometric task (Reynolds and Lakie 2010; Lakie et al. 2012), which implicates that in physiological tremor mechanical properties play a major role. Since previous findings suggested that this is not the case for PBT, we expected no difference in tremor frequency between type A and type B tremor.

It is an ongoing discussion, whether TST is a form of dystonia, of essential tremor or an entity of its own. In dystonia, an overflow to muscles not involved in the task has been described (Cohen and Hallett 1988; van der Kamp et al. 1989), whereas no such overflow was found in PWT (Bain et al. 1995). However, in the latter study (Bain et al. 1995), the conclusion was drawn based on the finding of an equal proportion between patients and controls who had rhythmic activity in adjacent muscles, but no direct relationship between tremor acceleration and (electromyography) EMG signals was investigated. Our third aim therefore was to investigate, whether an overflow of muscular activity to the adjacent muscles can be detected in TSTM. We investigated, whether a coherence between EMG activity of adjacent muscles and tremor acceleration exists for type A as well as for type B tremor, where EMG activity was defined as the muscular oscillatory activity as measured by the EMG.

We thus tested the following hypotheses:

1. (a) Mean power of EMG activity (flexor, extensor and A–A sum) and (b) mean coherence between EMG activity and tremor acceleration is highest in the frequency range of 3–8 Hz.

2. There is no difference in mean tremor power and mean coherence between type A and type B tremor in the 3–8 Hz frequency range.
3. An overflow of EMG activity to adjacent muscles can be detected in TSTM measured as a coherence between EMG activity and tremor acceleration for type A and type B tremor.

Methods

The study was approved by the local ethics committee, conducted according to the declaration of Helsinki and written informed consent was obtained from all participants.

Participants

We included 23 patients (4 female, 19 male; mean age 51.5 ± 11.4 years) who presented to our outpatient clinic because of a TST at their respective instrument. The accelerometer and EMG data of one guitarist had to be excluded because of technical problems during data acquisition. Therefore, data of only 22 patients were analyzed and are reported here. Of those, twelve patients played the violin, one the viola, three the cello, one the guitar, two the oboe, two the saxophone, one the piano and one the percussion. All patients reported that tremor occurred (at least initially) only at the instrument. In 15 patients, we observed a wrist flexion–extension tremor, in three a pronation–supination tremor, in four an elbow flexion–extension tremor, in one a shoulder tremor (Table 1).

Epidemiological data were acquired, as well; however, details will be reported elsewhere.

In all patients a task-specific tremor occurred at the instrument. Exclusion criteria were dystonic symptoms as assessed during clinical examination and other neurological or psychiatric disorders. Furthermore, potentially tremorgenic medication or disorders (e.g., hyperthyreosis) were exclusion criteria.

Measurement

Surface EMG was recorded with AG/AG^{2+} surface electrodes (biovision, Wehrheim, Germany), which were placed above the muscle belly of the respective muscles identified as being responsible for the oscillatory activity. For a wrist flexion–extension tremor, we recorded wrist flexor and extensor EMG activity and for elbow-tremor biceps and triceps EMG activity. For supination–pronation tremor, we recorded supinator–pronator EMG activity, where for analysis the pronator muscle was considered as

Table 1 Patient characteristics of 22 patients

	Gender	Age (yrs)	Instrument	Tremor location	Affected muscles	Adjacent muscles	Type A tremor	Type B tremor	Tremor frequency (Hz)
Pat. 1	Male	40	Sax	Wrist	Wrist flex/ext	–	l	n	7.3
Pat. 2	Male	62	Violin	Wrist	Wrist flex/ext	–	r	n	6.4
Pat. 3	Male	54	Violin	Wrist	Wrist flex/ext	–	r	n	4.9
Pat. 4	Male	48	Violin	Wrist	Wrist flex/ext	–	r	n	6.8
Pat. 5	Female	50	Violin	Wrist	Wrist flex/ext	–	r	y	6.4
Pat. 6	Female	24	Violin	Wrist	Wrist flex/ext	–	r	n	6.8
Pat. 7	Male	59	Violin	Wrist	Wrist flex/ext	Biceps/ triceps	r	n	8.3
Pat. 8	Male	62	Violin	Wrist	Wrist flex/ext	Biceps/ triceps	r	n	7.1
Pat. 9	Male	54	Violin	Wrist	Wrist flex/ext	Biceps/ triceps	r	y	4.6
Pat. 10	Male	54	Guitar	Pron/sup	Pron and sup	–	r	y	6.8
Pat. 11	Female	58	Cello	Pron/sup	Pron and sup	–	l	n	6.1
Pat. 12	Male	63	Cello	Pron/sup	Pron and sup	–	l	n	5.1
Pat. 13	Male	56	Sax	Wrist	Wrist flex/ext	–	l	n	6.4
Pat. 14	Female	55	Oboe	Elbow	Biceps/ triceps	Wrist flex/ext	r	n	6.4
Pat. 15	Male	57	Violin	Shoulder	Delt/pect	Biceps/ triceps	r	y	6.6
Pat. 16	Male	63	Viola	Elbow	Biceps/ triceps	Wrist flex/ext	l	y	6.4
Pat. 17	Male	23	Piano	Wrist	Wrist flex/ext	Biceps/ triceps	l	n	6.4
Pat. 18	Male	38	Cello	Wrist	Wrist flex/ext	Biceps/ triceps	r	y	6.8
Pat. 19	Male	58	Violin	Wrist	Wrist flex/ext	Biceps/ triceps	r	y	7.6
Pat. 20	Male	55	Violin	Wrist	Wrist flex/ext	Biceps/ triceps	r	y	6.1
Pat. 21	Male	59	Violin	Elbow	Biceps/ triceps	Wrist flex/ext	r	y	7.8
Pat. 22	Male	55	Oboe	Elbow	Biceps/ triceps	Wrist flex/ext	l	y	6.1

Pat patients, *yrs* years, *pron* pronation, *sup* supination, *flex* flexor, *ext* extensor, *delt* deltoid muscle, *pect* pectoralis muscle, *l* left, *r* right, *Sax* Saxophone

flexion and supinator as extension. For shoulder tremor deltoideus and pectoralis-major EMG activity were recorded, where for analysis deltoid activity was considered extension and pectoralis activity as flexion. The adjacent muscles were defined as muscles adjacent to those muscles, which were diagnosed to contribute to tremor. For wrist and pronation–supination tremor, the adjacent muscles were the biceps and triceps muscle; for elbow tremor, adjacent muscles were wrist flexor and extensor and for shoulder tremor, adjacent muscles were biceps and triceps muscles. To detect an overflow of EMG activity, we measured EMG at adjacent muscle groups. This was done in 12 patients only due to technical difficulties at the beginning of measurements. Type A tremor was recorded while patients were playing the instrument and type B tremor was measured while patients were holding the

respective body part in a position for playing. During clinical examination at the instrument, the playing condition evoking the severest symptoms was identified and taken for tremor measurement. EMG data were bandpass filtered (cutoff 1–450 Hz) and rectified before applying a low-pass Butterworth filter of 13 Hz. Data were normalized with regard to the maximum amplitude of the respective signal (maximum voluntary contraction). We then divided the dataset into every 2,048 points, which, at a sample rate of 500/s, equals 4.096 s. Using this sub-dataset, FFT was performed, which yielded a vector consisting of complex number in 2,048 dimensions. Next, we computed the squared absolute value, through multiplying this vector by its complex conjugate number. The power was derived as the ensemble average of the squared value of this absolute value. Furthermore, we obtained the peak frequency of the EMG and accelerometer data. We subdivided the frequency into three bands from 1 to 3 Hz, 3 to 8 Hz and 8 to 12 Hz as described before (Lee et al. 2013b) and calculated the maximum power for each frequency band.

We then applied a novel method for which the A–A sum is calculated (Inoue et al. 2011; Ariga et al. 2012) with the following formula:

$$m1 + m2,$$

where $m1$ and $m2$ are the normalized EMG data for the flexor and extensor muscle, respectively. A–A sum is linearly correlated to the joint stiffness (Ariga et al. 2012) i.e., a higher A–A sum is equivalent to a higher joint stiffness caused by coactivation. In the following section, we define the A–A sum as "coactivation index" for simplicity.

Statistics

To assess hypothesis (1a), we performed a one-way ANOVA for each of the flexor and extensor muscle and coactivation index with frequency range (1–3 Hz, 3–8 Hz, 8–12 Hz) as an independent variable. To assess hypothesis (1b), we performed a one-way ANOVA for coherence between tremor acceleration and EMG signals with frequency range (1–3 Hz, 3–8 Hz, 8–12 Hz) as an independent variable.

To assess the hypothesis (2), we performed a two-way ANOVA with tremor type (A, B) and frequency range (1–3 Hz, 3–8 Hz, 8–12 Hz) as independent variables. This test was performed with the ten musicians displaying type B tremor.

To assess hypothesis (3), we performed a one-way ANOVA for flexor and extensor muscle and coactivation index, respectively, with frequency range (1–3 Hz, 3–8 Hz, 8–12 Hz) as an independent variable with the 12 musicians in whom EMG of adjacent muscles was recorded. We furthermore applied a two-way ANOVA with tremor type

(A, B) and frequency range as independent variables, with the 8 musicians displaying type B tremor.

For all ANOVAs, Tukey post hoc tests were applied.

To assess the gender distribution, we applied two χ^2 tests: The first assuming no gender predominance and a second one assuming a gender predominance as in musician's dystonia (female:male = 1:4), if the first should reveal a significant difference.

Results

Power and coherence of tremor-related signals

Hypothesis (1a) The result revealed a main effect of frequency range [flexor: $F_{(2,42)} = 10.2$, $p < 0.01$; extensor: $F_{(2,42)} = 19.1$, $p < 0.01$; coactivation index: $F_{(2,42)} = 13.2$, $p < 0.01$]. Tukey post hoc analysis revealed a significantly higher power at the 3–8 Hz range compared to the other two ranges (1–3 Hz and 8–12 Hz, respectively. Flexor and extensor $p < 0.01$). No significant difference was found between the 1–3 and 8–12 Hz ranges.

Hypothesis (1b) The results revealed a main effect for frequency range [between tremor acceleration and flexor: $F_{(2,42)} = 22.2$, $p < 0.01$; between tremor acceleration and extensor: $F_{(2,42)} = 27.9$, $p < 0.01$; between tremor acceleration and coactivation index: $F_{(2,42)} = 31.3$, $p < 0.01$]. Tukey post hoc analysis revealed a significantly higher power at the 3–8 Hz range compared to the other two ranges (1–3 Hz and 8–12 Hz, respectively. Flexor and extensor $p < 0.05$). No significant difference was found between the 1–3 and 8–12 Hz range for the coherence of the extensor, while coherence was significantly higher at the high frequency range as compared to the low frequency range for the flexor and for coactivation index (Fig. 1).
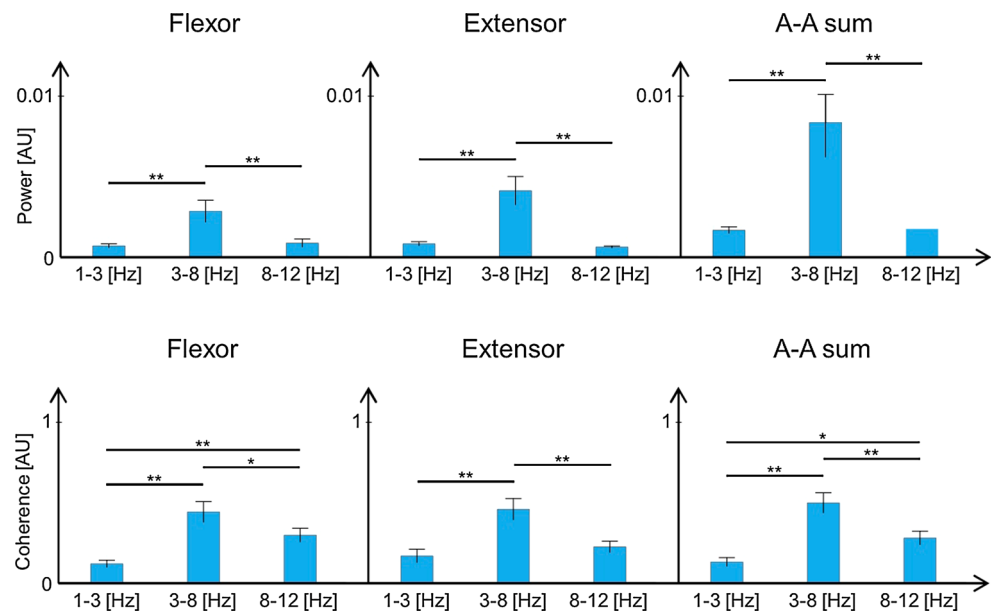
Comparison between type A and B tremors

Type B tremor was detected in ten patients.

Hypothesis (2) the result revealed a main effect for frequency range [flexor: $F_{(2,18)} = 6.6$, $p < 0.01$; extensor: $F_{(2,18)} = 3.6$, $p < 0.05$] but not for task. Post hoc analysis did not reveal a difference in tremor power between type A and type B tremor in any frequency range for either muscle. Two-way ANOVA for power of type A and type B tremor for coactivation index did not reveal a main effect for frequency range or task.

For the coherence data, a main effect was found for frequency range [flexor: $F_{(2,18)} = 23.0$, $p < 0.01$; extensor: $F_{(2,18)} = 20.4$, $p < 0.01$; coactivation index: $F_{(2,18)} = 22.0$, $p < 0.01$] but not for task (Fig. 2). Post hoc analysis did not reveal a difference in coherence between type A and type B tremor in any frequency range for either muscle.

Fig. 1 *Top panel* mean and standard error of the power of the peak frequency for flexor, extensor and A–A sum for each frequency band. *Bottom panel* mean and standard error of the coherence between the respective EMG signal and tremor acceleration for each frequency band. *AU* arbitrary units. $*p < 0.05$, $**p < 0.01$

Fig. 2 *Top panel* mean and standard error of the power of the peak frequency for flexor, extensor and A–A sum for each frequency band and type A and B tremor. *Bottom panel* mean and standard error of the coherence between the respective EMG signal and tremor acceleration for each frequency band for type A and B tremor. *AU* arbitrary units, *Inst* task-induced tremor (i.e., at the instrument), *POS* position-dependent tremor. $*p < 0.05$, $**p < 0.01$, $*** p < 0.001$
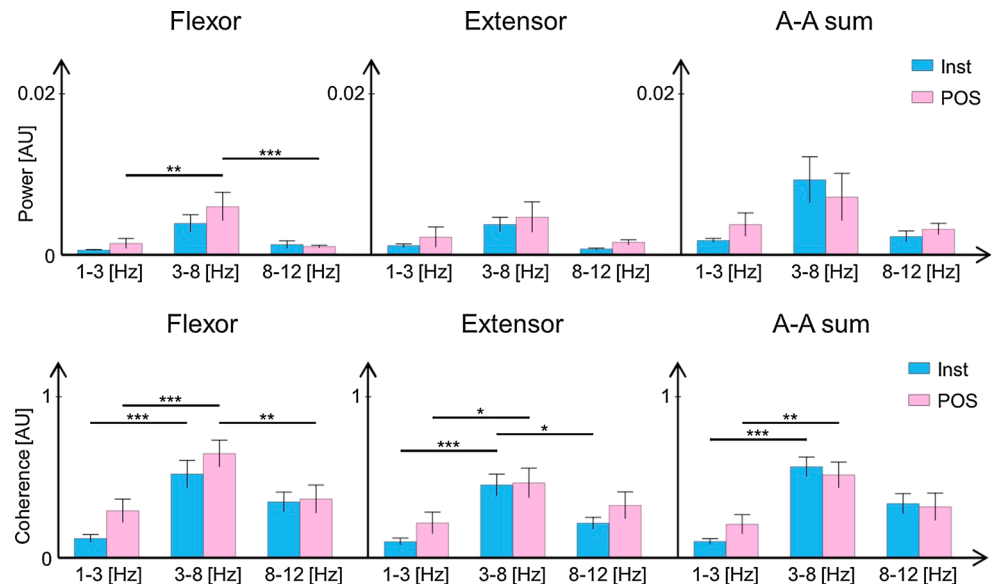
Adjacent muscular activity

EMG activity of muscles adjacent to a symptomatic muscle was successfully measured in twelve patients. The assumption for hypothesis (3) was that the mean coherence between EMG activity of adjacent antagonist muscles and limb acceleration is not identical between the frequency ranges but higher for the middle range, where TSTM is expected. One-way ANOVA for coherence between EMG activity of adjacent antagonist muscles (flexor, extensor, coactivation index) and hand acceleration with frequency range (1–3 Hz, 3–8 Hz, 8–12 Hz) as an independent variable revealed a main effect of frequency range [flexor: $F_{(2,22)} = 5.7$, $p = 0.01$; extensor: $F_{(2,22)} = 13.6$, $p < 0.01$,

coactivation index: $F_{(2,22)} = 6.9$, $p < 0.01$]. Tukey post hoc analysis revealed a significantly higher power ($p < 0.05$) of amplitude of the 3–8 Hz range compared to the other two ranges (1–3 and 8–12 Hz), respectively, for the extensor and coactivation index. No difference was found for the flexor. No significant difference was found between the 1–3 and 8–12 Hz range.

Of the twelve patients in whom EMG activity of adjacent muscles was assessed, eight patients had type B tremor. Two-way ANOVA for coherence between EMG activity of adjacent muscles (adjacent flexor and extensor, coactivation index) and hand acceleration of type A and type B tremor revealed a main effect for frequency range [flexor: $F_{(2,14)} = 7.6$, $p < 0.01$; extensor: $F_{(2,14)} = 14.7$,

Fig. 3 *Top panel* mean and standard error of the power of the peak frequency for adjacent flexor, adjacent extensor and A–A sum for each frequency band. *Bottom panel* mean and standard error of the coherence between the respective EMG signal of adjacent muscles and tremor acceleration for each frequency band. *AU* arbitrary units. *$p < 0.05$, **$p < 0.01$
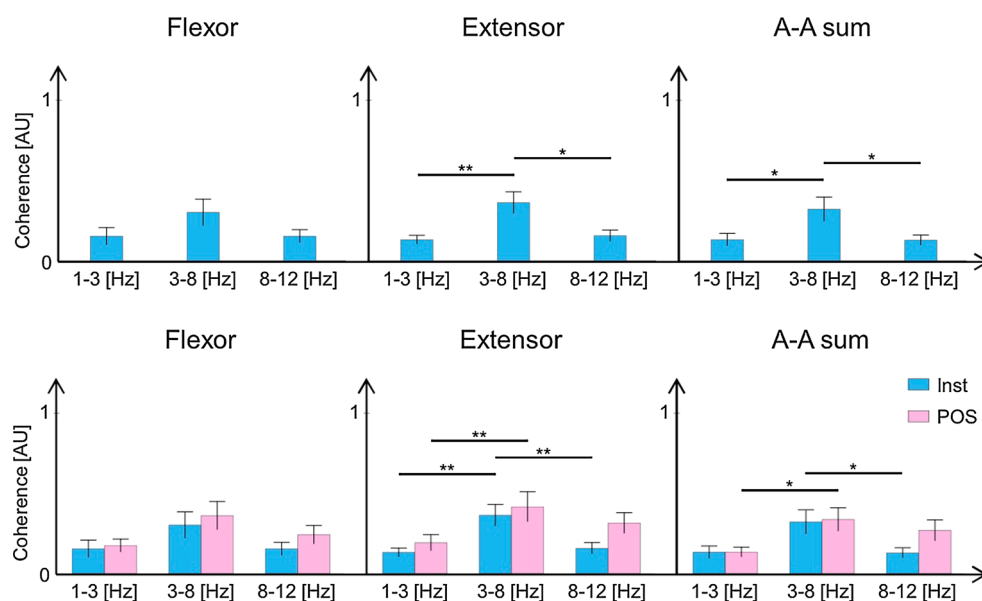
$p < 0.01$; coactivation index: $F_{(2,14)} = 9.1$, $p < 0.01$] but not for task (Fig. 3). Post hoc analysis did not reveal a difference in coherence in type A and type B tremor in any frequency range for either muscle.

Discussion

Primary task-specific tremors have been mostly described in string instrumentalists (Lederman 2012; Lee et al. 2013b). In this paper, we quantitatively assessed and described task-specific tremor in 22 musicians at a variety of instruments. We found a mean tremor frequency of 6.5 ± 0.9 Hz for all patients. This finding is comparable with previous reports of frequencies in PWT of 5-7 Hz (Elble et al. 1990; Bain et al. 1995; Modugno et al. 2002) and slightly faster than 5.8 ± 0.8 Hz in PBT reported before (Lee et al. 2013a). Mean frequency for type B tremor was 5.3 ± 1.5 Hz. In addition, a small standard deviation across patients confirmed consistency of the tremor frequency across different instrumentalists, however, due to the small sample size per instrument general conclusions should be drawn carefully and larger sample sizes are needed.

Power and coherence of tremor-related signals

We found a significantly higher tremor amplitude in the frequency range of 3–8 Hz for antagonist muscular activities as well as for coactivation. This corroborates our previous results of four violinists with PBT (Lee et al. 2013b) and extends it to TST in other instruments and tremor different from wrist flexion–extension tremor. This

is interesting, because although TSTM may occur at different body parts, this does not influence spontaneous tremor frequency. We did not detect tremor at the resonance frequency at those body parts that have a resonance frequency outside the 3–8 Hz frequency range, as e.g. wrist tremor which has a resonance frequency of 8–12 Hz (Elble 1996) or tremor at the shoulder which has a resonance frequency of 0.5–2 Hz (Deuschl et al. 2001).

Coherence between EMG signals and tremor acceleration was highest at the frequency range of 3–8 Hz corroborating findings in PBT (Lee et al. 2014b). Likewise, Elble et al. found a flexion–extension tremor of the forearm in PWT with a frequency of 5–7 Hz and alternating as well as synchronous EMG activity. This coactivation was described as dystonia with increasing intensity as writing continued (Elble et al. 1990). A novel approach of this paper with regard to coactivation is the application of coactivation index (Inoue et al. 2011; Ariga et al. 2012) which is associated with coactivation of a movement (Ariga et al. 2012). In our previous paper (Lee et al. 2013b), coactivation was obtained by calculating the time-varying coactivation of the wrist agonist and antagonist muscles by computing the overlap of the waveforms of these muscles (Furuya et al. 2012). The advantage of coactivation index over the coactivation is that it is linearly correlated to the joint stiffness (Ariga et al. 2012) i.e., a higher coactivation index is equivalent to a higher joint stiffness caused by coactivation. It therefore constitutes a quantitative assessment of coactivation that can furthermore be applied to model human limb movement (Ariga et al. 2012). These advantages over our previous method and the confirmation of our first hypothesis strongly support the notion that coactivation is directly related to task-

specific tremor and that peripheral mechanisms do not play a major role in the genesis of task-specific tremor. This suggests that tremor at different instruments and locations shares similar pathophysiological mechanisms and probably originates from the same entity. The higher coherence in the high frequency range for the flexor and coactivation index to our understanding reflects the fact that amplitudes above 8 Hz also contribute to TSTM.

Comparison between type A and B tremors

We did not find a difference in any frequency range between type A and type B tremor for either tremor power or coherence between EMG activity and tremor acceleration. This indicates that cocontraction plays a similar role for both tremor types and suggests that both tremor types share common mechanisms. With regard to frequency ranges, tremor power and coherence between EMG activity and tremor acceleration were highest for the frequency range 3–8 Hz. Coherence was significantly higher compared to the low frequency range. However, the difference between the middle and the high frequency was not significant for all conditions. This again might indicate that frequencies above 8 Hz contribute to tremor, not only for type A tremor, as mentioned above, but also for type B tremor. Interestingly, it has been shown for physiological tremor that in small hand muscles tremor at frequencies above 8 Hz can be found during isometric finger muscle contraction (Farmer et al. 1993; McAuley et al. 1997). It is therefore possible that our finding reflects these mechanisms, although in the aforementioned studies healthy individuals were assessed. Furthermore, our finding of a possible contribution of higher frequencies of type A tremor cannot be explained by this phenomenon, since type A tremor is not isometric.

Tremor frequency was slightly higher for type A tremor (6.5 Hz) as compared to type B tremor (5.3 Hz). Likewise, a higher tremor frequency has been reported for type A tremor (7.3 Hz) as compared to type B tremor (5.5 Hz), in PWT (Bain et al. 1995). In contrast to the movement-specific type A tremor, an isometric contraction was necessary for type B tremor to hold the affected body part in a position for playing the instrument. Our finding therefore is interesting, because it is contrary to what has been found in physiological hand or finger tremor. Several studies could show that tremor frequency is lower in a task that involves movement than during an isometric task. Lakie et al. found that the frequency of movement-related tremor is 2 Hz lower than static tremor (Lakie et al. 2012) and Daneault et al. found a decrease in tremor frequency from 9.7 Hz (movement) to 6.45 Hz (postural) (Daneault et al. 2011). Reynolds and Lakie assessed a change in tremor frequency after a voluntary hand movement was stopped and found an increase in hand tremor frequency from 7.2 to 8.0 Hz (Reynolds and Lakie 2010). This effect was explained by muscle thixotropy, i.e., the reduction of muscle stiffness during voluntary movement. Since tremor frequency is proportional to muscle stiffness by

$$f \sim \sqrt{(K/I)},$$

where K is stiffness and I is inertia, the conclusion was that mechanical properties play an important role in physiological tremor. We did not find an increase in frequency in type B tremor, similar to what is known from PWT (Bain et al. 1995) and consider this finding as another indication that TST (unlike physiological tremor) does not depend on mechanical properties but is mainly centrally driven. Our finding differs from findings in PWT in that coactivation was found only in type B (and not in type A) tremor in PWT (Bain et al. 1995), whereas in our sample coactivation occurred in both tremor types. The prevalence of almost 50 % found in 21 patients with PWT (Bain et al. 1995), is comparable to that of 43 % in our sample.

Adjacent muscular activity

We found a main effect of "frequency range" on coherence between EMG activity of adjacent muscles (flexor, extensor or coactivation index, respectively) and tremor acceleration. This confirms the hypothesis of non-identical means between the different frequency ranges, even though post hoc tests did not find a significant difference. The highest coherence between EMG activity of adjacent muscles and tremor acceleration was found in the medium frequency range, where TSTM can be found as shown under hypothesis 1 and in previous studies (Lee et al. 2014b). Importantly, coherence between coactivation of adjacent muscles and tremor acceleration was significantly higher in the middle frequency range, as well, which indicates that coactivation of adjacent antagonist muscles plays a role in TSTM, as well and that an overflow of muscular activity occurs in TST, as well. This is contrary to what has been reported in PWT (Bain et al. 1995). In that paper, the proportion of patients who had rhythmic activity in the adjacent muscles was compared between patients and controls and no difference was found. However, no direct relationship between muscular activity and tremor acceleration was measured and therefore cannot be ruled out. Overflow of muscular activity to adjacent muscles and coactivation of antagonist muscles are characteristic findings in focal dystonia (Cohen and Hallett 1988; van der Kamp et al. 1989; Berardelli et al. 1998). The common explanation for the excessive motor output is a disinhibition at the spinal and cortical level (Sohn and Hallett 2004; Torres-Russotto and Perlmutter 2008) and loss of independent movement selection (Berardelli et al. 1998).

Surround inhibition is impaired in musician's dystonia as well as in writer's cramp (Sohn and Hallett 2004; Furuya and Altenmüller 2013). Furthermore, alterations in short intracortical inhibition have been repeatedly found in task-specific dystonia, including musician's dystonia (Chen et al. 1997; Espay et al. 2006). Finally, a shortened silent period (SP) after transcranial magnetic stimulation was reported in focal dystonia (Chen et al. 1997; Espay et al. 2006). Likewise, in TST, a reduced intracortical inhibition (Byrnes et al. 2005) as well as a shortened SP (Ljubisavljevic et al. 2006) was found that has also been reported in PWT (Byrnes et al. 2005). Our results suggest that disinhibition does lead to an overflow of muscular activity to antagonist muscles that are adjacent to the affected muscles in TSTM, which is a known phenomenon in focal dystonia. During clinical examination, we ruled out dystonic phenomena. We therefore believe that our findings are further evidence that TST are a form of dystonia and share a common pathophysiology. Further findings suggesting that TST and dystonia share a common pathophysiology come from functional brain imaging studies. Increased activity in the sensorimotor area and in the cerebellum was found in task-specific dystonia (Odergren et al. 1998; Haslinger et al. 2010) as well as in PWT (Berg et al. 2000). A recent study could show an alteration in the functional connectivity within the cerebello-basal-ganglia-cortical loop at rest in focal dystonia (Dresel et al. 2014). To our knowledge, the functional connectivity in PWT has not been investigated so far, however we would hypothesize that a similar alteration becomes apparent.

Different studies have discussed TST as a form of dystonic tremor (Gironell and Kulisevsky 2009; Elble and Deuschl 2011), which is defined as a tremor in the body part affected by dystonia (Deuschl et al. 1998). Little, however, is known about the pathophysiology of dystonic tremor and it is mainly discussed in the context of dystonia itself (Deuschl and Bergman 2002; McAuley and Rothwell 2004) as a reduced inhibition (Berardelli et al. 1998). Therefore, according to the main phenomenological definition, none of our patients in this study had dystonic tremor, since dystonic symptoms were an exclusion criteria. However, if increasing evidence suggests that TST and task-specific dystonia share a common pathophysiology, it may be worth discussing, to make it two entities: (1) dystonic tremor with dystonic symptoms and (2) dystonic tremor without additional dystonic symptoms (TST). A recent study investigating dystonic tremor in musician's dystonia of the fingers did not find dystonic tremor (Lee et al. 2014a). It therefore remains an open question, why in some cases dystonia in musicians manifests itself as an involuntary cramping and in others as an involuntary oscillation of the affected body part.

One clinical implication of this finding may be to consider a treatment of adjacent muscles with Botulinum toxin in TSTM if treatment in the affected muscles does not yield a satisfactory improvement. Further studies have to address this question.

Conclusion

We found that in TSTM, neuromuscular activity of flexor and extensor muscles as well as their coactivation not only of the affected muscles but also of muscles adjacent to the affected muscles contributes to tremor. These findings corroborate the notion that coactivation is related to TST and that an overflow of muscular activity to adjacent muscles occurs in TST, which in turn indicates that TST are related to dystonia. We furthermore assessed type B TST and compared it to type A tremor. As expected, no difference was found with regard to EMG activity. In contrast to physiological tremor, positional tremor was not slower than movement-related tremor. Thus, the direct relation of neuromuscular output to tremor and the lack of tremor at the resonance frequency for hand tremor supports the hypothesis that mechanical reflex properties play a minor role in genesis of TST and that it is a centrally driven phenomenon. Further studies have to assess, whether a treatment of adjacent muscles with Botulinum toxin is beneficial in TSTM.

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