No Exaggerated Tremor Severity Perception in Functional Tremor

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Abstract

Background: Symptoms of functional neurological disorder tend to be variable, yet patients often report them being present constantly and of permanently severe intensity. Furthermore, they typically worsen when they are mentioned or during clinical examination. Such phenomena are sometimes interpreted as indicating symptom exaggeration or even fabrication.

Methods: In order to test the notion of inaccurate symptom perception or reporting, we directly compared subjective to objective tremulousness of reaching movements in people with a functional action tremor, people with an organic action tremor and healthy controls. Identical subjective and objective measures were used, thus eliminating any potential metacognitive confounders. Furthermore, we assessed both immediate perceptual experience with a real-time perceptual task, offering the most direct comparison; and near-time retrospective reports as the latter contribute to peoples´ overall judgement of their condition.

Results: There was no significant difference in subjective compared to objective tremor severity between the three groups for either the real-time or retrospective conditions.

Conclusion: People with functional tremor do not perceive or report their tremor in an exaggerated manner, compared to people with an organic tremor or healthy controls. We propose that symptom exacerbation through attentional mechanisms provides an alternative explanation for findings that are frequently attributed to 'exaggeration'.

Key words: functional neurological disorder, functional movement disorders, exaggeration, perception, prejudice, visual feedback, attention

Introduction

Functional neurological symptoms are often severe and debilitating. Patients frequently demonstrate great difficulties in moving during examination, with marked slowness, weakness, or additional movements such as tremor or jerks. Conversely, with distraction, movements become transiently more fluid and natural (1), and abnormal movements such as tremor may stop altogether. This apparent paradox leads to questions about the voluntariness of functional neurological symptoms, and the concern that patients may be exaggerating or even fabricating their symptoms (2).

One way of exploring this important issue is to determine whether patients with functional movement disorders (FMD) accurately report aspects of their symptoms (e.g., severity, presence) judged against objective recordings. Two previous studies compared objective tremor duration over multiple days to patients' judgement of the duration or its impact. In the first study, functional tremor patients reported tremor durations that exceeded actual tremor durations recorded actigraphically by 65% (compared to 28% in the organic tremor group) (3). The second study found no difference in symptom burden ratings, nor in their association with objective tremor duration, between functional and organic tremor groups (4). In a different study without control group, people with FMD retrospectively rated their symptom severity while they had been filmed, and again while watching the recording. Their retrospective severity ratings were worse than their ratings of the video recordings, which in turn were no different to those of an independent expert (5). Duration estimates are frequently distorted (6), and ratings of symptom burden contain a multitude of metacognitive confounders such as circumstances, expectations, general health, outlook, mood etc. Thus, there is no controlled study of symptom intensity perception in FMD without metacognitive confounders.

At a basic perceptual level, symptom intensity or duration may be genuinely perceived in a manner that is inconsistent with objective reality i.e., there may be an increase in the gap between the actual and perceived symptom, akin to a perceptual gain. The next level is the metacognitive perception of the severity of the symptom - is it rated as mild or severe? A related level is the perceived impact on the quality of life or symptom burden.

A fundamental question which has not been addressed in previous studies is whether there is an abnormality in sensory perception, akin to an abnormal perceptual gain. This would mean that sensory feedback about symptoms/performance is distorted at a basic level and is perceived in an excessive manner. Such a phenomenon has been shown in anorexia nervosa for example, in which patients perceive their body dimensions as larger than they are in reality (7). We set out to explicitly address this question in functional tremor. In order to allow direct comparison between subjective and objective tremor severity, without metacognitive confounders, identical measures were used. Specifically, we recorded patients' reaching trajectories, and quantified the tremulousness of their trajectories by the lateral deviations from a perfectly straight line.

We considered that judgements about one's own movements can involve both an immediate perceptual experience of the movement one is currently making, and also a more synthetic, memory-based, and often re-interpreted assessment of one's previous movement performances in general. The immediate perceptual experience offers the most direct comparison between subjective and objective tremor severity. Retrospective estimates of tremor may be confounded by memory but may nevertheless form the core of people's overall judgement of their condition and their clinical history. We therefore developed a real-time judgement task to probe the immediate perceptual experience of tremor during reaching movement, and a retrospective judgment task across a 2–3-hour episode comprising many movements.

Methods

People with a functional action tremor were compared to two control groups: healthy controls and people with an organic action tremor (each condition comprised mainly patients with a dystonic tremor, except for four patients with an essential tremor and one with Wilson's disease). For detailed recruitment information and methods, please refer to our previous publication (1). The number of included participants in each condition are summarised in Table 1 and Supplementary Table 1. The latter furthermore details the participants´ characteristics. There was no statistically significant difference between the three groups in terms of tremor severity, gender, age, and a non-verbal IQ test. Tremor duration was significantly longer in the organic compared to functional tremor group. The functional tremor group had significantly higher anxiety and depression sub-scores on the Hospital Anxiety and Depression scale than either control group. (Supplementary Table 1) Participant numbers varied between the three conditions because the retrospective conditions depended on participants having successfully performed corresponding reaching movement conditions as described in our previous publication (1). Furthermore, the real-time condition was performed after many conditions and hence omitted in some because of ensuing fatigue, discomfort or time constraints. For both retrospective conditions, four functional tremor participants were excluded because their tremor was initially severe but improved in a linear fashion over time, making their average unrepresentative. The study was approved by the local ethics committee (Reference: 16 ⁄LO ⁄1463) and carried out in accordance with the Declaration of Helsinki (8). Participants gave their written, informed consent.

Participants moved their index finger on a touchpad from a starting position to a target (4.5mm circle) 24cm straight ahead. In the trials with direct visual feedback, subjects had direct vision of the touchpad and their arm (Fig. 1A). In the indirect visual feedback conditions, their arm was hidden underneath a horizontal screen on which the start, the target and a cursor indicating current finger position, were displayed in real-time (Fig. 1A).

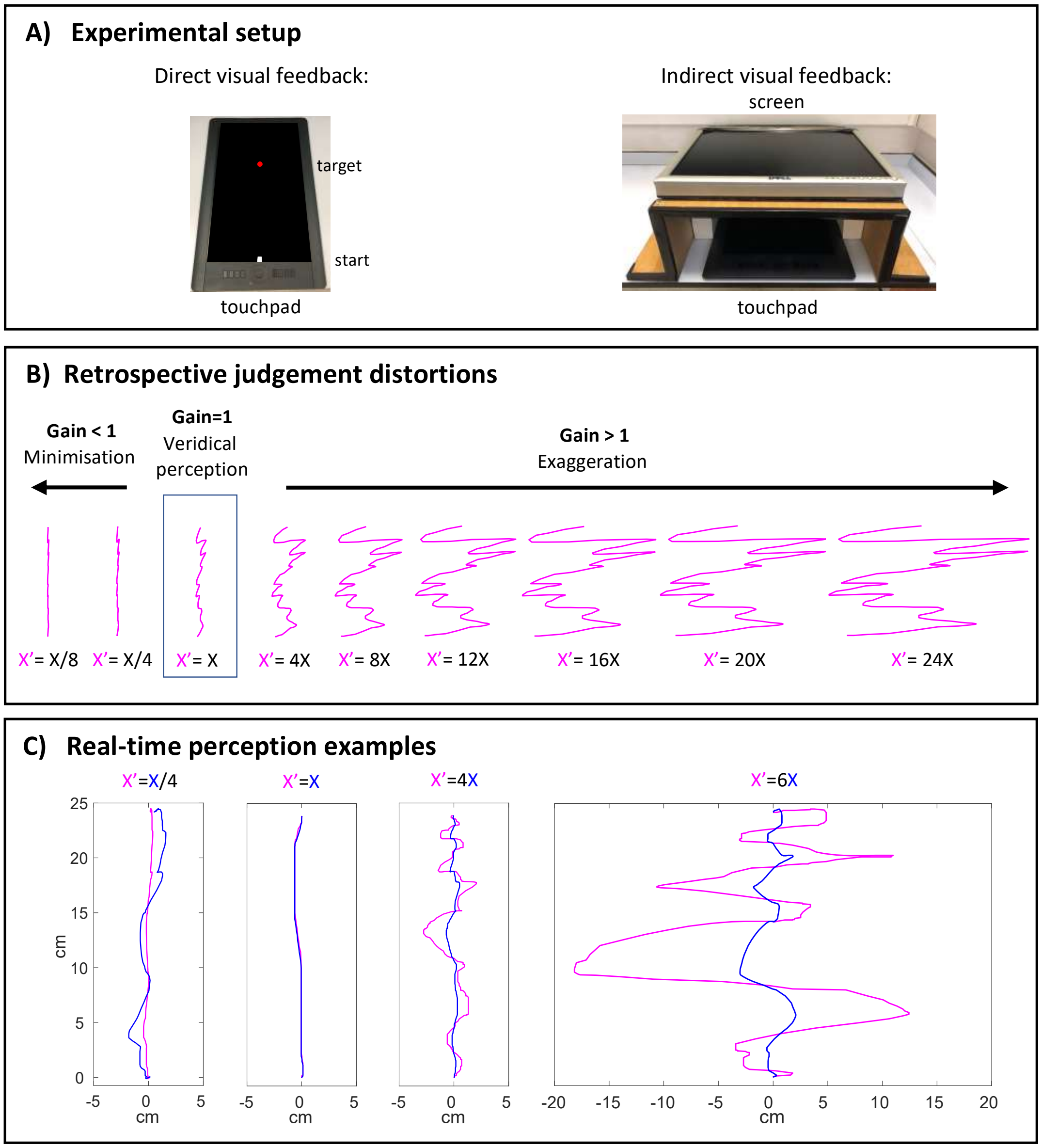

Fig. 1 Experimental setup and examples
A) Equipment (1). The direct path from start to target was 24 cm. B) The near-average trajectory, two straightened out distortions (each x coordinate divided by 4 and 8 respectively) and 6 shakier distortions (each x coordinate multiplied by 4, 8, 12, 16, 20 and 24 respectively) were shown. Participants chose the trajectory they thought corresponded to their average. C) Real-time distortion examples. Purple line: trajectory of the cursor provided as visual feedback on the screen (only cursor movement visible to the subject), blue line: actual finger trajectory on the touchpad (invisible to the subject).

Retrospective tremor judgements: Participants performed over 250 reaching movements described previously (1), over a time period of 2-3 hours. Subsequently, the trajectory corresponding most closely to the individual´s average trajectory, both in terms of path length and maximum lateral deviation from a perfectly straight line was selected. The lateral deviation of this near-average trajectory was then distorted with different gains as illustrated in Fig. 1B. Participants chose the trajectory they thought corresponded most closely to their average of all previous trials. A distortion or gain of 1 indicates no distortion and hence a correct judgement of one's movements; a gain >1 indicates exaggerated judgement and thus overestimation of tremulousness / non-straightness; and a gain <1 an underestimation. The real-time condition below was performed with indirect visual feedback. In order to ensure that the indirect visual feedback did not alter people's perception, the retrospective tremor judgements were performed twice, once for the trajectories with indirect and once for those with direct visual feedback.

Real-time tremor perception: The real-time indirect visual feedback on the screen of the participants´ finger movement on the touchpad, moved either exactly as their finger did (visual feedback X coordinate identical to the finger position X coordinate), or it moved in a shakier or less shaky manner. For shakier visual feedback, all X coordinates of the finger position on the touchpad were displayed on the screen after having been multiplied by 2, 4, 6, 8 or 10 respectively, thus increasing the lateral deviations of the visual feedback. Conversely, straightened-out visual feedback was created by dividing all X coordinates of the finger position by 2, 4, 6, 8 or 10 respectively, thus decreasing the lateral deviations of the visual feedback. (Fig. 1C). Participants decided during their reaching movement, if the visual feedback moved as their finger did, or in a shakier or straighter manner. After each trial, they adjusted the shakiness / gain of the feedback up or down by one distortion level until they felt that the visual feedback moved exactly as their finger did. This was performed eleven times, starting once, in random order, with each visual distortion level (X/10, X/8, X/6, X/4, X/2, X, 2X, 4X, 6X, 8X, 10X).

Results

When *retrospectively* estimating their tremor severity / trajectory straightness, all three groups overestimated their tremulousness / non-straightness, but there was no significant difference between the three groups, neither for direct, nor for indirect visual feedback conditions. Additional analyses of covariance (ANCOVA), with covariates of anxiety and depression scores, did not change inferences about group main effects. (Table 1) The estimates for the retrospective direct versus indirect visual feedback conditions did not significantly differ in either group (only including participants having performed both conditions; one-sample t-test on the difference between the log10 of the perceived gains in the functional tremor group: $t(16)=-1.48$, $p=.16$; organic tremor group: $t(18)=-1.58$, $p=.13$; and healthy controls: $t(19)=-1.02$, $p=.32$). Hence the indirect visual feedback was unlikely to be a confounder in the real-time tremor perception condition.

In the *real-time* tremor perception condition, a significant group effect was found in ANOVA, but disappeared when covarying for anxiety and depression scores. The healthy controls, who given the absence of any tremor had to perceive a smaller lateral movement signal, underestimated their non-straightness. Both tremor groups showed no significant misperception of their actual tremor. (Table 1)

	Functional tremor mean [95% CI]	**Organic tremor** mean [95% CI]	**Healthy controls** mean [95% CI]	**Statistical analyses**	
Retrospective tremor judgement (indirect visual feedback)					
N	19	21	23		
Perceived gain	2.54 [0.46-14.01]	2.74 [0.41-18.43]	2.06 [0.30-14.37]	ANOVA *p*=.59 η^2=.018	ANCOVA Group *p*=.75 HADA *p*=.20 HADD *p*=.84
				Perceived gain versus no gain: FT: signed-rank **p_{corr}=.002**, *r*=.75 OT: t-test **p_{corr}=.0004**, *d*=1.04 HC: t-test **p_{corr}=.002**, *d*=0.73	
Retrospective tremor judgement (direct visual feedback)					
N	17	19	20		
Perceived gain	3.34 [0.75-14.89]	4.17 [0.66-26.24]	2.83 [0.32-25.06]	ANOVA *p*=.45 η^2=.030	ANCOVA Group *p*=.20 HADA *p*=.95 HADD ***p*=.030**
				Perceived gain versus no gain: FT: t-test **p_{corr}=.00002**, *d*=1.58 OT: signed-rank **p_{corr}=.0006**, *r*=.83 HC: signed-rank **p_{corr}=.002**, *r*=.69	
Real-time tremor perception					
N	21	19	23		
Perceived gain	1.04 [0.36-3.02]	0.82 [0.28-2.40]	0.69 [0.37-1.30]	ANOVA ***p*=.022** η^2=.119	ANCOVA Group *p*=.38 HADA *p*=.91 HADD *p*=.24
				Perceived gain versus no gain: FT: signed-rank p_{corr}=.61, *r*=-.11 OT: t-test p_{corr}=.25, *d*=-0.36 HC: signed-rank **p_{corr}=.001**, *r*=-.73	

Table 1: Results of the retrospective judgement and real-time tremor perception conditions
The gain or distortion factor is the factor by which each X coordinate is multiplied. A gain of 1 signifies no distortion. Statistical analyses were performed on the $\log_{10}$ of the perceived gain, in order to linearise the data. One-way ANOVA was performed if the assumption of homogeneity of variance was met (Levene's test). Additional analyses of covariance (ANCOVA) were performed in order to exclude influence of covariates such as anxiety and depression. One-sample t-tests (t-test), or Wilcoxon signed-rank test (signed-rank) in case of non-normal distributions, evaluated if each group´s average perceived gain significantly varied from no gain (gain=1). The significance level for all tests was set at 0.05, two-tailed. Significant results are highlighted in bold. Šidák-Holm corrections adjusted for multiple comparisons (p_{corr}). Effect size estimates were based on eta squared (η^2) Cohen's *d* and Pearson´s *r*. 95% CI = 95% confidence interval, N=number of participants, FT= functional tremor, OT=organic tremor, HC=healthy controls.

Discussion

The present study evaluated subjective tremor severity both in real-time and in retrospect relative to objectively-measured tremor severity. Both the subjective and objective assessment used the same dependent variable, namely lateral deviation during reaching movements, allowing direct comparison and excluding potential confounding metacognitive factors. While in the retrospective conditions all groups overestimated the tremor/non-straightness of their reaching trajectories, there was no significant difference between the three groups. Overall, the results did not show any significant difference in tremor severity perception between functional tremor patients and either people with an organic tremor or healthy controls, neither when estimating their tremor severity in retrospect, nor in real-time. Interestingly, the functional tremor group's real-time perceptual gain of their attended movement was 1.04, i.e., virtually perfect, given a gain of 1 indicates no distortion. Thus, in a situation in which people with FMD attended to their movement, there was normal real-time sensory perception with a normal sensory perceptual "gain".

These findings suggest neither the real-time immediate perception of tremor, nor the near-time memory-based retrospective judgement of tremor are particularly exaggerated or unusual in functional tremor patients, relative to the other groups.

How do our findings fit with previous studies? It is important to recognise the different questions asked in the controlled studies. Parees et al. asked participants how much of the day they had experienced tremor. Based over a whole day period, patients significantly over-estimated the amount of time compared to people with organic tremor (3). In the Kramer et al. study participants reported five times a day how much the tremor had "bothered" them in the interim. This metacognitive judgement was similar between people with functional and organic tremor (4). Our current study assessed the more immediate sensory perceptual aspects of tremor.

It seems likely that the Parees et al. study result is mediated primarily by attentional effects and a resulting metacognitive bias. Functional symptoms typically manifest with attention and improve or disappear with distraction (1). Functional tremor manifests whenever patients' attention is directed towards their arm, including when they check for tremor presence. Conversely, when attention is focused elsewhere, the tremor largely disappears, but the person remains unaware of it. This produces a general bias that the tremor is always or often present. The video recording study summarised above (5) was uncontrolled. Thus, retrospective symptom judgements may not be exaggerated solely in FMD, but also in control groups, as in the present study and hence this may be considered normal. However, as for the duration overestimation, attentional effects offer the most plausible explanation – whenever the person's attention was focused on their symptoms, symptoms were present, yet they were unaware of them being milder or even absent whenever their attention was focused elsewhere. Thus, retrospective ratings overestimated symptom severity. However, when viewing recordings of their movements, people with FMD realised that their symptoms were milder, and rated them similarly to an independent expert.

Many clinical observations can also be explained by symptom exacerbation through attentional mechanisms rather than exaggeration. Three common examples are (1) patients reporting that their symptoms are present constantly when in fact observation reveals them being intermittent, (2) symptoms appearing when they are mentioned, and (3) symptom exacerbation during clinical examination, compared to when the same movement is performed implicitly.

The current finding of intact immediate and near-time symptom reporting in functional tremor is in line with further previous data. Pain ratings in functional neurological disorder have previously been shown to be not dissimilar (9) or even decreased (10) compared to healthy controls. People with FMD report similar degrees of self-consciousness about their abnormal movements, compared to people with corresponding organic movement disorders (11). Although such measures contain many metacognitive confounders, they again point towards the absence of exaggeration.

In conclusion, the present study directly compared subjective tremor severity with actual tremor severity, using both real-time perceptual assessments, and near-time retrospective judgements. There was no evidence for abnormal reporting of performance in people with functional tremor. Other pathophysiological mechanisms, such as symptom amplification through attention, might be the underlying reason for clinical observations which are frequently misinterpreted as exaggeration.

Statements and Declarations

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Conflict of interest statement

On behalf of all authors, the corresponding author states that there is no conflict of interest.

Data availability

Our ethics agreement prevents data being openly available, but individual researchers may request anonymised data from the corresponding author.

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Authorship

Anne-Catherine Huys: conceptualised, designed and programmed the study, acquired funding and ethics approval, recruited participants, collected and analysed the data, wrote the first draft of the paper and its final version.

Patrick Haggard: verified data analysis, critically reviewed and edited the paper.

Kailash Bhatia: critically reviewed and edited the paper.

Mark Edwards: critically reviewed and edited the paper.

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	Tremor Duration Mean *(SD)*	**Tremor Severity**	**Age** Mean *(SD)* (range)	**M:F**	**HAD-Anxiety** Mean *(SD)*	**HAD-Depression** Mean *(SD)*	**Raven´s matrix** Mean *(SD)*
Retrospective (indirect visual feedback)							
FT N=19	6.8y *(4.9)*	mild: 9 moderate: 6 severe: 4	51.8 *(15.9)* (21-75)	9:10	9.6 (5.2)	7.9 (4.6)	8.8 (2.9) (N=18)
OT N=21	23.3y *(17.0)*	very mild: 3 mild: 13 moderate: 5	53.6 *(17.4)* (21-78)	11:10	6.3 (3.1)	3.8 (2.2)	9.9 (2.3)
HC N=23	-	-	42.7 *(15.3)* (21-68)	9:14	4.0 (2.4)	1.9 (2.5)	10.3 (1.7)
Statistics	t-test **p=.0002**	Chi^2 p=.052	ANOVA p=.064	Chi^2 p=.67	KW **p=.0004**	ANOVA **p<.0001**	KW p=.18
	Post-hoc tests: Anxiety: FT vs OT*: **p_{corr}=.025,** FT vs HC*: **p_{corr}=.0007,** HC vs OT: **p_{corr}=.015** Depression: FT vs OT: **p_{corr}=.001,** FT vs HC: **p_{corr}=.00004,** HC vs OT: **p_{corr}=.001**						
Retrospective (direct visual feedback)							
FT N=17	6.7y *(5.1)*	very mild: 1 mild: 7 moderate: 7 severe: 2	53.1 *(14.8)* (23-75)	8:9	9.5 (5.4)	8.0 (4.8)	8.6 (3.1) (N=16)
OT N=19	23.6y *(17.1)*	very mild: 3 mild: 13 moderate: 3	53.3 *(17.7)* (21-78)	10:9	6.2 (3.1)	3.6 (2.0)	9.7 (2.4)
HC N=20	-	-	44.0 *(16.0)* (21-68)	9:11	4.0 (2.3)	2 (2.6)	10.1 (1.7)
Statistics	t-test **p=.0004**	Fisher p=.12	ANOVA p=.14	Chi^2 p=.89	KW **p=.002**	KW **p=.0001**	KW p=.38
	Post-hoc tests: Anxiety: FT vs OT*: **p_{corr}=.041,** FT vs HC*: **p_{corr}=.003,** HC vs OT: **p_{corr}=.041** Depression: FT vs OT: **p_{corr}=.002**, FT vs HC: **p_{corr}=.0003,** HC vs OT: **p_{corr}=.003**						
Real-time							
FT N=21	6.5y *(4.8)*	mild: 11 moderate: 6 severe: 4	52.2 *(15.3)* (21-75)	9:12	9.0 *(5.1)*	7.4 (3.7)	8.2 (3.4) (N=20)
OT N=19	23.8y *(16.9)*	very mild: 3 mild: 12 moderate: 4	55.9 *(16.3)* (28-78)	10:9	6.4 (2.9)	3.6 (2.0)	9.8 (2.4)
HC N=23	-	-	42.7 *(15.3)* (21-68)	9:14	4.0 (2.4)	1.9 (2.5)	10.3 (1.7)
Statistics	t-test **p=.0001**	Chi^2 p=.061	ANOVA **p=.022**	Chi^2 p=.67	KW **p=.001**	ANOVA **p<.0001**	KW p=.076
	Post-hoc tests: Age: FT vs OT: p_{corr}=.46, FT vs HC: p_{corr}=.11, HC vs OT: **p_{corr}=.026** Anxiety: FT vs OT*: p_{corr}=.052, FT vs HC*: **p_{corr}=.001,** HC vs OT: **p_{corr}=.013** Depression: FT vs HC: **p_{corr}=.0002,** FT vs OT: **p_{corr}=.001,** HC vs OT: **p_{corr}=.001**						

Supplementary Table 1: Number of participants with their characteristics and statistical analyses

Tremor severity was assessed clinically. Raven´s progressive matrices measure non-verbal IQ, range 0-12. Post-hoc multiple pairwise comparisons (two-sample t-tests or the non-parametric Wilcoxon rank sum test) are Šidák-Holm corrected (p_{corr}). M:F = male to female ratio, HAD = Hospital Anxiety and Depression scale – Anxiety / Depression sub-scores, FT = Functional Tremor, OT = Organic Tremor, HC = Healthy Controls. ANOVA = one-way ANOVA, t-test = two-sample t-tests, *unequal variances assumed in the two-sample t-test, Chi^2= Chi-square goodness of fit, KW = Kruskal-Wallis test with ties, Fisher = Fisher's exact test. Statistically significant results are highlighted in bold. One functional tremor patient did not complete the Raven´s matrices and was thus excluded from the Raven´s group average in the three conditions.