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Sensitivity and Specificity of the Finger Tapping Task for the Detection of Psychogenic Movement Disorders

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SHORT TITLE: Finger Tapping Task in Psychogenic Movement Disorders

BACKGROUND

Psychogenic movement disorders (PMD) represent 3% of all movement disorder clinic patients

[1]. While this represents a small percentage of the total clinic population, PMD patients are

diagnostically challenging cases and can require a disproportionate amount of clinic resources

and health care dollars [2,3]. In 1988, Fahn and Williams [4] proposed criteria for PMD

categorizing patients based upon their clinical history and exam findings. This classification has

been revised by subsequent authors [5-7] but to date there are no reliable, objective means of

identifying a potential PMD in a clinic based setting.

Finger tapping tests (FTT) provide an objective measure of upper extremity fine motor skills and

are a core component of neuropsychiatric testing for a variety of neurological illnesses including

movement disorders, psychogenic conditions, and malingering [8-12]. FTTs also have a long

history of use in movement disorders [13-16]; however, they have primarily been used to

quantify the upper extremity impairment in patients with idiopathic Parkinson disease (IPD).

Multiple authors have demonstrated FTTs inversely correlate with United Parkinson Disease

Rating Scale III (motor) subscale (UPDRS III) [17] scores when adjusted for age [14,15,18]. No

studies to date have looked at FTT scores in patients with PMD.

In neuropsychiatric testing, high variability and inconsistency between trials are considered an

indication of malingering or psychogenicity [11,19,20]. Malingering is defined as purposefully

exaggerating a physical symptom for a clear goal while psychogenicity concerns a broader group

which may include malingering patients but also those with somatoform and conversion

disorders demonstrating non-organic symptoms with no clear secondary objective [21]. FTTs are consistently reduced and more variable in both malingering and psychogenic disorders [8-11]. Arnold et al. compared FTT scores in subjects with suspected malingering to subjects with a variety of neurological illnesses including closed head injury, dementia, and depression. They found subjects with suspected malingering performed the FTT more slowly than their comparison group counterparts regardless of the neurological diagnosis [8]. Similarly, another study found naïve and coached malingerers performed significantly slower than control subjects on FTT [9]. Even pseudoseizure patients demonstrated scores in the impaired range on the FTT component of the Halstead-Reitan Neuropsychological Test Battery [11,22]. Matheson et al. devised a unique technique to measure the inconsistency in patients with non-organic symptoms and suspected malingering by examining the coefficient of variation (CV) between test trials [20]. Given the slow and highly variable FTT scores across groups with psychogenic disorders and malingering, we hypothesized that PMD patients will similarly demonstrate lower FTT scores and large CVs between trials when compared to patients with other common movement disorders.

METHODS

Subjects-This study was approved by the Washington University School of Medicine Human Research Protection Organization. Subjects were recruited from the Washington University in St. Louis Movement Disorder Center between June 2006 and October 2008 and signed informed written consent. The sample consisted of 325 individuals divided into five groups: (a) IPD, (b) essential tremor (ET), (c) dystonia, (d) PMD, and (e) healthy adult controls. All subjects were evaluated and diagnosed by a movement disorder specialist. IPD patients were all classified as

probable Parkinson's disease according to the United Kingdom Brain Bank clinical criteria [23].

Dystonic subjects demonstrated the following patterns of primary dystonias: 18 cervical, 1

oromandibular, 4 blepharospasm, 2 craniocervical, 2 generalized, and 5 brachial (writer's cramp)

[24]. All PMD patients were categorized as clinically established psychogenic movement

disorders according to the Fahn and Williams classification defined as inconsistency (we

included distractibility) or incongruity in the movement with one of the following: other

neurologic signs that are definitely psychogenic, multiple somatizations, or an obvious

psychiatric disturbance [4]. Within the PMD group 53.0% of our subjects demonstrated action

tremor, 46.2% had rest tremor, 15.4% dystonia, 30.8% bradykinesia, 30.8% myoclonus, 15.4 %

chorea, and 7.6% had tics using the criteria proposed by Hinson et al. [25]. In addition, three

patients with paroxysmal movements also had video EEG monitoring with no electrographic

correlate to their movements. All tremor patients were categorized as classic essential tremor

using criteria established by the consensus statement of the Movement Disorder Society on

Tremor [26]. Control subjects were recruited from the healthy spouses and family members of

patients seen in the Movement Disorder Center. All controls were screened for tremor and

parkinsonism by a movement disorder specialist using the UPDRS III. Control subjects were

excluded if they had a total UPDRS III score >3 or rest, postural, or action tremor $\geq$ 1 on the

UPDRS III.

Procedure-Handedness was determined from patient self-report. Finger tapping equipment

consisted of a counter with two levers spaced 20 cm apart [27]. Each subject completed three-30

second trials for each hand starting with the dominant side and then alternating between hands.

For each trial subjects were instructed to use the index finger of the indicated hand to alternate

tapping between the two levers as many times as possible in the 30 second period. Scores were recorded for each 30 second trial. Mean tapping scores were calculated by averaging the 30 second trial scores. The CV was calculated as the quotient of the standard deviation divided by the mean. Mean tapping scores and CVs were calculated for dominant hand trials, non-dominant hand trials, and the combination of both hands (combined scores).

Statistical Analysis-

The difference in age between the five categories was analyzed using ANOVA. If the ANOVA demonstrated an overall significance at $p < 0.05$, a Scheffe test was used to examine the differences between diagnostic groups. A Fisher's Exact test was used to analyze the differences in gender between diagnostic categories. The relationship between age and FTT scores and age and CV were examined by Pearson's correlation. If the overall Pearson's correlation was significant at $p < 0.05$, correlations were analyzed for individual subgroups. The overall effect of gender on FTT score and CV was examined by Student's T-test. If the Student's T test was significant at $p < 0.05$, t-tests were performed on individual subgroups. Associations between the Hoehn and Yahr stages [28] (H &Y stage), UPDRS III, FTT scores, and CV within the IPD group were analyzed by Pearson's correlation. ANOVA analysis was used to examine the differences in FTT scores between H & Y stages. If the ANOVA was positive at $p < 0.05$, Tukey's HSD post-hoc analysis was used to further examine the relationship between stages. The ANOVA and Tukey's analysis were also used to examine differences in FTT scores and the CV between the five diagnostic categories. Analysis of covariance (ANCOVA) using age as a covariate was further employed to analyze statistical differences among groups for FTT and CV. Specificity and sensitivity cutoff values for the PMD category were determined by visual

inspection of the data and confirmed through ROC analysis. ROC analysis was performed on the

raw FTT score and the ratio of expected to predicted FTT scores. The predicted FTT score was

determined from the regression equation for control subjects based upon age. The statistical

software SPSS for Widows v16.0 (Chicago, IL) was used for data analysis.

RESULTS

There was a significant difference in age between the diagnostic groups ($p < .001$). Subjects in

the PMD (49 ± 11.50 years) were younger than participants in the IPD, ET, dystonia and healthy

adult groups. The difference between the PMD and dystonia groups was not statistically

significant ($p = 0.176$). Gender was unevenly distributed between diagnostic categories ($p <

0.001$) with women over-represented in the PMD and dystonia categories. (Table 1) When all

groups were included, average combined FTT scores negatively correlated with age ($r = -0.274$,

$p < 0.001$). Subgroup analysis revealed combined FTT scores did not correlate with age for the

PMD or dystonia groups but did correlate with age in the IPD ($r = -0.422$, $p < 0.001$), ET ($r = -

0.480$, $p < 0.001$) and normal control group ($r = -0.338$, $p < 0.001$). The correlation between age

and FTT scores in the normal control data indicated the FTT score would be expected to

decrease by 0.338 taps for each additional year of age. Gender had no effect on FTT scores ($t =

0.735$, $p = 0.476$). Within the IPD category, combined FTT scores were negatively correlated

with both the H &Y stage ($r = -0.406$, $p < 0.001$) and UPDRS III scores ($r = -0.528$, $p < 0.001$).

ANOVA analysis revealed a significant difference between the H & Y stages ($F = 12.22$, $p \leq

0.001$). The Tukey's HSD showed that stage 1 subjects performed significantly more taps than

stages 3-5. Means and standard deviations for combined FTT in IPD patients are reported by H

&Y score in Table 2. The combined FTT means, standard deviations, adjusted combined FTT

means, and 95% confidence intervals for the five diagnostic categories are reported in Table 3. There was a significant difference in combined mean FTT between the diagnostic categories (F= 31.72, p <0.001.) A Tukey's HSD post hoc comparison indicated that the PMD group performed fewer taps than the IPD, ET, dystonia, and normal control groups. The difference in mean combined FTT between the diagnostic categories persisted after controlling for age (F=36.37, p < 0.001). Subgroup analysis demonstrated PMD subjects had a significantly lower average FTT score than the IPD subjects with a difference of 17.06 taps (p < .001) between the groups.

The combined CV for each subject across all trials did not correlate with age for any of the diagnostic categories (r = 0.063, p =0.258). There was also no effect from gender on the combined CV (t = 1.129, p = 0.260). The combined CV did not correlate with either the UPDRS III (r = -0.128, p =0.202) or H &Y stage (r = -0.118, p = 0.238) in IPD subjects. ANOVA/ANCOVA analysis did not demonstrate a significant difference in the combined CV between the diagnostic categories with the CV for all diagnostic categories falling between 6.10-11.2% respectively. ANCOVA for the dominant and non-dominant hands revealed a similar pattern of difference in combined FTT scores and CV between the diagnostic categories.

An ROC analysis was performed for combined mean FTT scores unadjusted for age with the area under the curve = 0.773, p = 0.001. Simple linear regression analysis was performed on scores from the normal control group to create a regression equation for predicted FTT scores based upon age in years. The regression equation was as follows:

Predicted FTT score = 96.84 - 0.388 (age in years)

ROC analysis was performed on the ratio of actual FTT score/predicted FTT score for age resulting in an area under the curve = 0.817, $p \leq 0.001$. Acceptable cutoff scores were defined as those with a specificity $\geq 80\%$ for the diagnostic category of PMD. A combined FTT score of 52.16 or less yielded an 80% specificity and a 69.2% sensitivity for the diagnosis of PMD. The ratio of actual FTT/predicted FTT scores was associated with a specificity of 89.1% and sensitivity of 76.9% for the diagnosis of PMD at the ratio score of ≤ 0.670 or $\leq 67.0\%$ of the predicted FTT score. Mean FTT and ratio scores were also analyzed for the dominant and non-dominant hand but there was no improvement in the specificity or sensitivity.

DISCUSSION

Our study demonstrates that the FTT is not only a valid method of quantifying the motor impairment in IPD, but it also provides an objective tool to aid in the identification of PMD patients within a movement disorder clinic. Consistent with previous reported FTTs [14,29], mean combined FTT scores inversely correlated with both H & Y stage and UPDRS III scores in subjects with IPD. The H & Y stage and FTT scores were strongly correlated, however there was a difference between stage 1 and stages 3 thru 5 as demonstrated on post-hoc testing. This may reflect an underlying functional difference between the stages or the relatively small number of subjects in these groups. FTT scores also demonstrated a strong inverse correlation with UPDRS III scores. These results validate that our method of FTT administration provides an objective means of measuring upper extremity motor impairment in IPD independent of physical examination. Our method has the advantage of providing a validated, inexpensive, and simple

objective measure of IPD impairment with the portability not available in more complex FTT methods [14-16,29].

While clinical assessments are the standard practice in diagnosis of PMD, the difference in FTT scores between the diagnostic categories in our study was remarkable. PMD subjects had significantly lower combined FTT scores when compared to the other diagnostic groups. When examining the unadjusted FTT means, PMD patients had an average of six fewer taps than the IPD group regardless of that group's H &Y stage or UPDRS III score. The difference was even more remarkable considering the IPD group was on average 20 years older. Correlations between age and the FTT indicated that scores decreased by 0.338 taps for each year of age or one tap for every three years of life. PMD patients would have been expected to perform approximately six more taps on average than the IPD group. Not surprisingly, when the analysis was controlled for age, the difference between the PMD and IPD subjects was even larger (17.06 taps) with the IPD subjects having the lowest scores of the four diagnostic categories available for comparison.

ROC curves suggested combined average FTT scores of less than 52.16 have a reasonable specificity and specificity for categorization into the PMD group. However these raw FTT scores were not corrected for age. The ratio of actual/predicted FTT scores included a correction for age and appears to be a better predictor of inclusion into the PMD group. The actual/predicted score of ≤ 0.670 was associated with a specificity of 89.1% and sensitivity of

76.9%. Assuming a 3% prevalence of PMD in a movement disorder clinic [1], the positive predictive value (PPV) and negative predictive value (NPV) for a ratio score of ≤ 0.670 (89.1 % specificity) would be 18.5% and 99.3% respectively. Applying these numbers to an average movement disorders clinic where PMD patients are relatively uncommon, it is not unexpected that the FTT would be associated with a low PPV of 18.5%. Conversely, the low disease prevalence is associated with relatively high NPV of 99.3%. This indicates that any patient with a FTT ratio greater than 0.670, has a 99.3% chance of not having a PMD.

Contrary to our hypothesis, PMD patients did not have a greater variability between trials than our four control groups. The minimal variance in all groups ($< 12\%$) may be explained by the short task period (30 seconds). Alternatively, in this study we did not attempt to separate malingering from other psychogenic conditions. It is possible these conditions have different pathophysiologies and therefore do not reflect the previous studies by Matheson [20] which focused solely on patients with suspected malingering. Lastly, in an effort to keep this task simple and easily reproduced, we were unable to evaluate the time interval between taps. A more sophisticated FTT such as the one used by Jobbágy et al. [16] may have been able to detect an interval variation but with more expense and sacrificing portability.

The main limitation of this study was the small number of subjects with clinically established PMD, reflecting the low prevalence of PMD patients within movement disorders clinics. The thirteen subjects with PMD in this study represent every prospectively ascertained, clinically

established PMD patient seen from our large referral base in two years. With a prevalence of 3%

this corresponds to over 400 movement disorders patients screened. We could have increased

the number of PMD patients by including less stringently defined PMDs, but for a validation

study that would be inappropriate. The lack of statistically significant difference in CV between

the diagnostic groups may be a type II error. However, the mean FTT scores differed very

significantly between the PMD group and every other clinical group, including being over 30%

lower than even the much older IPD group. The chance that this is a false positive finding is less

than 1 in 1000 ($p < 0.001$).

In conclusion, the FTT has clinical applications within a movement disorder clinic beyond

quantifying the upper extremity impairment in IPD. The FTT has a good specificity and

sensitivity as a diagnostic tool for supporting the diagnosis of PMD. While PMD patients

represent a small percentage of the clinic, their care can consume a disproportionate amount of

clinic resources and health care dollars [2,3]. The FTT may provide an objective tool to aid to the

clinical diagnostic criteria set forth by Fahn and Williams [4] for identifying patients with a

PMD.

ACKNOWLEDGMENTS

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Table 1. Demographic data by Diagnostic Category.

		Normal Control (n = 130)	IPD (n = 101)	ET (n = 49)	Dystonia (n = 32)	PMD (n = 13)	p value
Handedness	Right	116	92	41	30	10	
	Left	11	7	6	1	2	
	Ambidextrous	3	2	2	1	1	
Gender	Male	47	57	15	5	3	<0.001[a]
	Female	83	44	34	27	10	
Age (yr)[b]		64 $\pm$ 11.09	69 $\pm$ 9.02	61 $\pm$ 16.02	58 $\pm$ 10.28	49 $\pm$ 11.50	<0.001[c]

Table 2. Clinical Characteristics of PMD subjects (n =13).

	Action Tremor	Rest Tremor	Dystonia	Bradykinesia	Myoclonus	Chorea	Tics
Subject 1						Generalized	
Subject 2						Generalized	
Subject 3	Bilateral UE[a]						
Subject 4	Right UE	Right UE		Left UE, LE[b] > Right UE, LE			
Subject 5						Generalized	
Subject 6			Bilateral UE				
Subject 7	Bilateral UE	Jaw, Right LE		Bilateral UE	Bilateral LE		
Subject 8	Bilateral UE	Bilateral UE	Bilateral UE				
Subject 9	Bilateral UE	Right UE, Bilateral LE		Left UE, LE > Right UE, LE			
Subject 10						Generalized	
Subject 11	Right > Left UE	Right UE		Right UE, LE > Left UE, LE			
Subject 12	Head, Bilateral UE	Left > Right UE					Head, Bilateral UE
Subject 13						Generalized	

Table 3. Idiopathic Parkinson combined FTT averages and UPDRS III score by Hohn and Yahr Stage.

H & Y Stage	n	Combined FTT		UPDRS3	
		Mean	SD	Mean	SD
1.00	6	71.25	15.68	12.50	3.11
2.00	72	58.64	12.24	25.68	8.85
3.00	13	48.55	10.75	40.12	7.58
4.00	4	41.50	8.01	36.75	5.24
5.00	6	48.17	15.10	44.33	3.84

Table 4. Combined FTT Mean, SD, and Adjusted Mean by Diagnostic Category.

	n	Mean	SD	Adjusted Mean	Std. Error	95% Confidence Intervals	
						Lower Bound	Upper Bound
Normal control	130	74.31	11.61	74.31	1.120	72.11	76.51
Dystonia	32	68.37	12.30	66.31	2.269	61.84	70.77
Essential tremor	49	61.23	16.42	60.10	1.824	56.16	63.34
Idiopathic Parkinson disease	101	56.79	13.55	58.78	1.303	56.16	61.26
PMD	13	50.42	15.89	41.72	3.643	33.77	48.16