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Performance Validity Tests in non-litigant patients with Functional Motor Disorders

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ABSTRACT

Background: Performance Validity Tests (PVTs) are used in neuropsychological assessments to detect patterns of performance suggesting that the broader evaluation may be an invalid reflection of an individual's abilities. Data on Functional motor disorder (FMD) are currently poor and conflicting.

Objectives: We aimed to examine the rate of failure at three different PVTs of non-litigant, noncompensation seeking FMD patients, and we compared their performance to that of healthy controls and controls asked to simulate malingering (healthy simulators).

Methods: We enrolled 29 non-litigant, non-compensation seeking patients with a clinical diagnosis of FMD, 29 healthy controls and 29 healthy simulators. Three PVTs, the Coin in the Hand Test (CIH), the Rey 15-item Test (REY) and the Finger Tapping Test (FTT), were employed.

Results: FMD Patients showed low rates of failure at the CIH and REY tests (7% and 10%, respectively) and slightly higher at the FTT (15%, n=26) test, which implies a motor task. Their performance was statistically comparable to that of healthy controls but statistically different from that of healthy simulators (p<0.001). 93% of FMD patients, 7% of healthy simulators, and 100% of healthy controls passed at least two of the three tests.

Conclusions: PVT performance of non-litigant, non-compensation seeking patients with FMD ranged from 7 to 15%. Patient's performance was comparable to controls and significantly differed from that of simulators. This simple battery of three PVTs could be of practical utility and routinely used in clinical practice.

INTRODUCTION

A cornerstone of neuropsychological assessment is the investigation of patterns suggestive of negative response bias or impaired engagement through performance validity tests (PVTs) [1, 2]. PVTs have historically been described as "effort tests" partly because low scores on tests might suggest poor effort, while scores below chance using a 'forced choice' paradigm may indicate that the subject intentionally chooses not to give the correct answer. Basically, these tests are designed to appear complicated on the surface, but they are, instead, easy to pass. Indeed, in the absence of interfering factors, performance could be expected to be good, even in patients with moderate cognitive or psychiatric impairment [1]. Therefore, failure of PVTs can invalidate the broader neurological or neuropsychological assessment and can also have implications in forensic or medico-legal contexts. Critically, however, the interpretation of failure at PVTs needs to be embedded in a comprehensive evaluation of patient's history and clinical presentation. Moreover, several factors, such as pain, fatigue, apathy, anxiety, or emotional distress as well as involvement in lawsuits or disability claims, might affect performance. Thus, results should always be interpreted with caution.

The literature on PVTs has proliferated in recent years and base rates of performance in several tests have been studied across multiple clinical populations, but mainly in those seeking financial compensation (i.e., disability claimants or litigating subjects) or in mixed clinical and litigant [1]. However, to better understand how these measures might inform standard psychological evaluations, it is nonetheless essential to have data on PVTs performance in different clinical groups, in the absence of known external motivations. Improved understanding of performance validity is especially important in Functional Motor Disorder (FMD), in which patients often experience stigma associated with misperceptions that they may be feigning or exaggerating their symptoms [3]. FMD is a common and disabling neurological condition [4], characterized by movement abnormalities where there is typically evidence of inconsistency of motor performance over time, changing in frequency or amplitude, worsening with attention, and attenuating with distraction [5]. Since FMD involve a disturbance of voluntary movements, it has often been confused with feigning, either as a factitious disorder or malingering [6, 7]. Malingering is defined as the fabrication, feigning or exaggeration of symptoms for an external gain (i.e., financial compensations/avoiding duties), whereas those with factitious disorder feign symptoms for a psychological reason [8]. In both cases, individuals are fully conscious of their behavior and voluntarily simulate symptoms, for example, failing easily achievable tasks. Since malingering might be prosecutable, a confident differentiation from FMD is highly desirable [9]. Unfortunately, to date, no shared and validated tools or biomarkers can differentiate functional disorders from willful exaggeration or simulation. New proposed criteria for neurocognitive malingering include the evidence of an invalid cognitive presentation as an essential criterion to detect malingering

with the demonstration of it achieved either with compelling inconsistencies (documented with written, audio, and video) or using PVTs [10].

A recent systematic review has reported that failure of single PVTs is common in several clinical groups, for example, Epilepsy and Parkinson's disease, with rates exceeding 25% in some groups with failure rates no higher in functional disorders than in other clinical conditions [1]. However, most studies have been conducted in patients with functional seizures [1, 11], while data on FMD patients are poor. Few previous studies addressed performance validity in the context of neuropsychological assessment in the FMD population, and results were conflicting [12, 13, 14]. In one study, valid test performance was present in most patients [12]. Low rates of failure of effort tests were shown in a mixed population of patients with medically unexplained symptoms compared to the performance of mild and strong simulators [13]. Conversely, patterns compatible with non-credible performance were found in psychogenic jerky movement disorder patients, but patients involved in lawsuits were not excluded [14]. To date, no studies have addressed non-litigant or non-seeking compensation FMD patients, and measures for motor effort have never been assessed in this population. In the present study, we aimed at assessing rates of failure of non-litigant no compensation-seeking FMD patients on three different PVTs, the Coin in the Hand Test (CIH), the Rey Fifteen-item Test (REY) and a test involving a motor task, the Finger Tapping Test (FTT). We compared the performance of FMD patients to that of a group of matched healthy controls asked to simulate cognitive malingering (healthy simulators) and healthy controls who did not receive specific instructions.

METHODS

Participants

Twenty-nine patients with a clinically established diagnosis of FMDs [15], aged \geq 18 years, were recruited at the FMD Clinic of the Parkinson's Disease and Movement Disorders Unit in Verona (Italy). Exclusion criteria were involvement in litigations or disability claims; prominent non-epileptic seizures; prominent and severe arm weakness; the presence of cognitive decline scored <70 on the Wechsler Memory Scale [16]; the presence of pain at the Numerical Rating Scale > 3; the presence of severe anxiety (Beck Anxiety Inventory score > 22) [17], and severe depression (Beck Depression Inventory-II scores > 19 for women and > 17 for men) [18]. Involvement in disability claims or litigations was detected through a structured interview. We also enrolled two groups of healthy controls (each, n = 29), selected through an advertisement among university students and healthcare personnel, matched for sex and education level. All participants gave their written, informed consent to participate in the study, which was carried out under the Helsinki Declaration tenets (2013). Approval

was obtained from the Institutional Ethics Committee (Project Number: University of Verona, Prog. 1757CESC).

Performance Validity Tests

FMD patients, healthy simulators, and healthy controls were assessed with three different PVTs: the CIH [19], the REY [20], and the FTT [21]. The three tests were administered in a counterbalanced order within each group.

The CIH is a well-known, easy-to-administer bedside forced-choice test, initially developed to detect malingering among patients with memory disorders [19]. The examiner shows participants a 2-euro coin in one hand for approximately 2 seconds and then closes his hands. Subjects are asked to close their eyes and count loudly backward from ten to one, then point to the examiner's hand where the coin is held. The task must be repeated for ten trials with the sequence R L L R R L R L R L, where R stands for right and L for left [22]. This forced-choice paradigm test also allows for scoring the chance level that is the one expected from random responding [19]. Scoring below the chance level is so low that blind guessing would have resulted in a better score, and it is considered a meaningful metric to suggest feigning.

The REY is a non-forced choice recognition memory performance two-sessions test. The first session is the "recall trial," in which a sheet with 15 items (including letters, numbers, and figures) arranged in five rows is shown for approximately 15 seconds [20]. Subjects are then given a blank sheet where they should reproduce previously seen items. In the second session, called "recognition phase", they are asked to circle the items previously seen in a sheet of 30 different figures with letters and numbers. The final performance is scored as follows: the number of correct recalled items + (correct recognition items – false positive) [20, 23].

Finally, given the nature of FMDs, we chose the FTT since it requires a motor task. The FTT is a non-forced choice test that assesses randomly, unrealistically slow, or erroneous responses, highlighting inconsistency of response patterns compared to the performance of well-documented neurological disorders. In the FTT, participants are asked to place both hands on the computer keyboard and tap one key (key B of a qwerty keyboard) as often as possible with the index finger until the examiner stops them [21]. Participants undergo five consecutive trials, each lasting 10 seconds, with the dominant and then with the other hand. A 10-seconds rest break is given after each trial, and a 30-seconds break after each third trial. The score consists of the total number of presses in a given time (10 sec), and the final performance is as follows: FTTscore = dominant hand average score + non - dominant hand average score.

While FMD patients and the healthy control group were asked to complete the test to the best of their ability without further specific instructions, subjects in the healthy simulators group were deliberately asked to deceive [24]. Healthy simulators were explicitly told: "I'd *like you to play a role. You have been involved in a car accident during which you reported a serious head trauma; you lost conscious-ness for some minutes and have been hospitalized for some days. Six months afterward, you still suffer from headaches, irritability, difficulty remembering things, staying focused for more than a few seconds, and your mood is always low. It is difficult for you to remember what you read. Your insurance company asks you to undergo a medical examination. Be aware that the more you appear compromised, the higher the probability of receiving the compensation you deserve. So, it is in your interest that the doctor understands that your symptoms are serious".*

Statistical analysis

Statistical analyses were performed through SPSS 24 software for Mac (Chicago, IL). Data were summarized as mean and standard deviation (SD). A one-way ANOVA test was used to compare the performance at the PVTs with 'group' as a between-subject factor. *Post hoc* analyses for multiple comparisons were also performed. The significance level was set at $p \le 0.05$ and the Bonferroni correction was applied for multiple comparisons.

In the absence of an absolute reference cut-off score for the three PVTs, we first computed cut-offs based on the performance of the healthy control group. We employed a criterion cut-off point using the formula "mean – (2 x standard deviation)". Scores lower than these cut-offs were used to classify subjects as having failed the test. To assess the performance of each group at the combination of the three tests, we used the criterion of passing at least two over three tests (percentage of participants who scored above cut-offs in at least two tests).

RESULTS

We enrolled 29 FMD patients [23 females, age years median (interquartile range, IQR) 43 (18-62); education years median (IQR) 13 (10-18)], 29 healthy simulators [22 females, age years median (IQR) 44 (28-68); education years median (IQR) 13 (13-19)] 29 healthy controls [23 females, age years median (IQR) 43 (20-60); education years median (IQR) 13 (8-20)]. FMD patients had a mean disease duration of 4.6 (\pm 3.6) years and predominant clinical phenotypes of functional weakness (72%, n=21), tremor (48%, n=14), and dystonia (45%, n=13).

Three FMD patients did not complete the FTT due to physical limitations: one reported both hands being too weak to perform the test, one suffered from paresis of the left hand, and one presented worsening motor symptoms while performing the test. Most participants were right-handed (23 right-handed FMD; 28 right-handed healthy simulators; 25 right-handed healthy controls). Frequency of failure and 'fail' cut-off scores based on the healthy control group performance (CIH = 9; REY = 20; FTT = 62) are reported in **Table 1**. Mean (and SD) scores of performances of the three groups and group comparisons are reported in **Table 2**.

Patients with FMD had a failure frequency of 10% on the CIH test (3 out of 29), 7% (2 out of 29) on the REY test, and 15% (4 out of 26) on the FTT (of note, three patients did not complete the FTT for physical problems, as specified above). One patient (3%), no healthy controls, and 12 (41%) healthy simulators scored below the chance to the CIH, a forced-choice test. When comparing the groups, as shown in **Table 2**, FMD patients' performance at CIH, REY, and FTT was statistically different from that of the healthy simulator group (all, p < 0.001), while it did not significantly differ from that of the healthy control group (**CIH p =.860; REY p=1.00; FTT p=.367**) (**Table 2; Fig. 1a-c**). Also, between the healthy simulator and the healthy control group, we found significantly different performances in all three tests (all, p < 0.001) (**Table 2; Fig. 1 a-c**). Considering the three tests together, according to the criterion of passing at least two tests, overall, 93% of patients, 7% of healthy simulators, and 100% of healthy controls passed the tests.

DISCUSSION

In this study, we aimed to assess whether FMD patients exhibit abnormal performance in a battery of three tests that are typically used to evaluate validity of performance in comparison to a group of healthy simulators and a group of healthy controls matched for age, sex, and educational level. Hence, we wanted to investigate whether non-litigant, non-compensation seeking FMD patients have performances that could raise the suspicion of poor effort or symptom exaggeration, according to a proposed algorithm of three tests. We found that FMD patients generally performed well at PVTs, with a low rate of failure of 10% at the CIH, 7% at the REY, and a slightly higher rate of failure (15%) at the FTT, which involves a motor task. FMD patients' performance was statistically different from that of healthy simulators but not from healthy controls. Conversely, healthy simulators scored clearly under the cut-offs, confirming that they correctly followed the instructions for simulating cognitive malingering. Of note, the performance of healthy simulators was statistically different from that of FMD and healthy controls in each test. We also found that most FMD patients passed a combination of the three tests. Indeed, 93% of patients passed at least two tests over three.

To the best of our knowledge, this is the first study that has specifically tested PVTs in nonlitigant, non-compensation seeking FMD patients. Our results expand the existing evidence of functional patients showing low failure rates at PVTs [12,13], which aligns with their reasonably expected performance capacity and with the nature of functional symptoms that are not

intentionally feigned or exaggerated [6, 25]. Moreover, our results implement previous attempts to demonstrate the value of PVTs in assessing functional patients [12, 13, 14]. Kemp et al. found that patients with medically unexplained symptoms and somatoform disorders had a low rate of test failure of cognitive PVTs (only 11%) in a sample of 43 patients [13]. Additionally, they found that patients performed differently compared to two groups of "mild" and "strong" cognitive impairment simulators. In keeping with these results, Věchetová et al. reported a 10% failure rate of PVTs in 30 FMD patients undergoing neuropsychological assessment [12]. Similarly, our sample showed a frequency of failure ranging from 7 to 15%, with the highest proportion of patients failing at the FTT, a measure of motor effort. Given the nature of FMD, our study's proposed test battery integrates effort-related measures for both motor and cognitive domains. FMD patients, indeed, often show a complex phenomenology, including motor and non-motor symptoms (including cognitive complaints) that are considered to rely on the same pathophysiological mechanisms. Conversely, another study by Heintz and collaborators found that patients with psychogenic myoclonus exhibited a worse performance at cognitive PVT than patients with Gilles de la Tourette syndrome, who served as a patient control group and healthy control subjects [14]. However, some of the patients in their sample were involved in lawsuits or other financial compensation matters. The authors could not exclude the possibility that this accounted for their failure at PVT. Additionally, they employed only one test to detect non-credible performance, whereas using at least two tests is soundly recommended in the literature [14].

Cochrane *et al.* (1998) demonstrated that participants who are asked to simulate make errors at the CIH test compared to patients with memory disorders, who do not make errors [26]. In our study, the median of the performance at the PVTs of the healthy simulators group was lower than that of the healthy control and FMD groups, confirming the higher number of errors made by the healthy simulators group.

Importantly, given the lack of published reference cut-offs, we calculated novel cut-offs based on the performance of our sample of healthy controls. In previous reports, the CIH test has demonstrated high sensitivity (86%) in distinguishing simulators from healthy non-simulators and brain-injured patients with a cut-off of < 8.5 [27] and even higher employing a cut-off of < 9 to identify 95% of volunteers asked to feign memory problems [28]. Our results were based on a similar cut-off of 9. Similarly, the REY is a fast and easy-to-administer PVT used in clinical settings. It has shown excellent specificity (92%) and good sensitivity (71%) when used to differentiate patients with a suspect effort from neurological patients, learning-disabled students, and controls using a cut-off of < 20 [20]. We found the same cut-off score of < 20 based on our controls' performance. The FTT has been previously used to differentiate simulators from head-injured patients [29]. In our study, we found a cut-off score of < 62 that has been previously employed to distinguish people meeting the criteria for

definite malingered neuropsychological dysfunction from individuals with a history of moderate to severe brain injury [29]. Similar scores were obtained in other studies evaluating FTT, confirming our results [21].

This study presents some limitations. First, instead of assessing real simulating patients, we asked healthy subjects to simulate. It is an issue of nearly all studies on willful exaggeration that it is hard to find a gold standard for malingering, especially in relation to functional motor disorders. It could be that our healthy simulators were performing much worse than practiced simulators would do in real life. Secondly, the lack of comparison to another clinically relevant group might preclude conclusions on the diagnostic utility of the battery of PVTs used in this study, and the use of novel cut-off scores calculated on the performance of our groups of healthy controls could limit the generalizability of results. However, our cut-offs are comparable to those reported in previous studies [20,21, 29]. Moreover, although a motor task could be expected to be altered in FMD, testing performance across multiple domains is warranted, as FMD is commonly affected by cognitive symptoms. Nevertheless, we excluded patients with cognitive decline or prominent arm weakness. Additionally, when instructed to feign cognitive impairment, all healthy simulators scored below cut-offs for FTT, suggesting that it's a test that may be more generally applicable to performance. Finally, it could be argued that excluding patients with cognitive impairment could represent a selection bias. However, even in the presence of poor cognition, performance at PVT is not impaired.

These limitations notwithstanding, our study indicates that FMD patients do not exhibit abnormal performance at PVTs and supports the hypothesis that symptoms exaggeration and poor effort is are not common in non-litigant, non-compensation seeking functional patients. These findings also suggest that the proposed battery of PVTs has some use as an adjunctive tool in studying performance validity in FMD patients. This simple battery of three PVTs may yield a reliable tool that should be employed routinely in neuropsychological testing of FMD, thus being of practical utility in evaluating patients. Failure rates of these and other PVTs among FMD patients involved in litigation and disability claims compared to non-litigant FMD patients would be of interest, as well as a greater emphasis on below-chance performance that more accurately determines the presence of willful exaggeration.

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Table 1. Rate of failure on each Performance Validity Test and frequency of passing the combination of tests

	Cut-off for fail- ure [*]	FMD (n=29**)	HS (n=29)	HC (n=29)
Coin in Hand	<9	10% (n=3)	93% (n=27)	0%
Rey Fifteen Item Test	<20	7% (n=2)	72% (n=21)	0%
Finger Tap- ping Test	<62	15% (n=4)	86% (n=25)	0%
Above cut off in 2/3 tests		93% (n=27)	7% (n=2)	100% (n=29)

Legend: FMD: functional motor disorders; HS: healthy simulators; HC: healthy controls. *Cut-off calculated on our HC group performance as mean – 2SD; **26 for Finger Tapping Test

	Performance score mean ± SD	Three group comparison	Two group com- parison	Two group com- parison	Two group comparison
		(One-way ANOVA)	FMD vs HS	FMD vs HC	HS vs HC
Coin in the hand test	FMD: 9.4 ± 1.4 HS: 4.5 ± 2.6 HC: 9.9 ± 0.3	F (2,86) = 87,4 p < .001	p <.001	<i>p</i> = .860	p <.001
REY 15-item test	FMD: 26.4 ± 4.3 HS: 13.9 ± 5.9 HC: 26.5 ± 3.5	F(2,86) = 68,5 p < .001	p <.001	<i>p</i> = 1.00	p <.001

Table 2. Performance scores and group comparisons at the Performance Validity Tests.

Finger tapping test	FMD: 90,7 ± 29 HS: 33,8 ± 21,5 HC: 100.6 ± 19.3	F (2,83) = 67,8 p < .001	p <.001	<i>p</i> = .367	p < .001
test	HC: 100.6 ± 19.3	p <.001			

Legend: Mean and standard deviation (SD) of the performance scores at the PVTs with the relative p-value for the comparison between the groups. **PVTs**: Performance Validity Tests; **FMD**: functional motor disorder, **HS**: healthy simulators; **HC**: healthy controls.

Figure 1a.



Legend: Healthy controls (HC), healthy simulators (HS) and functional motor patients (FMD). Continuous line: cutoff calculated as mean – 2SD; *p < 0.001





Legend: Healthy controls (HC), healthy simulators (HS) and functional motor patients (FMD). Continuous line: cutoff calculated as mean – 2SD; *p < .001



Fig. 1c.

Legend: Healthy controls (HC), healthy simulators (HS) and functional motor patients (FMD). Continuous line: cutoff calculated as mean – 2SD; *p < .001 **1a**) Scattered plot of performance of the three groups at the Coin in the Hand Test; **1b**) Scattered plot of performance of the three groups at the Rey Fifteen-item Test; **1c**) Scattered plot of performance of the three groups at the Finger Tapping Test