





ORIGINAL ARTICLE

Shoulder-Touch test to reveal incongruencies in persons with functional motor disorders

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Abstract

Background and Purpose: Clinical experience suggests that many patients with functional motor disorders (FMD), despite reporting severe balance problems, typically do not fall frequently. This discrepancy may hint towards a functional component. Here, we explored the role of the Shoulder-Touch test, which features a light touch on the patient's shoulders, to reveal a possible functional etiology of postural instability.

Methods: We enrolled consecutive outpatients with a definite diagnosis of FMD. Patients with Parkinson's disease (PD) or progressive supranuclear palsy (PSP) with postural instability served as controls. Each patient underwent a clinical evaluation including testing for postural instability using the retropulsion test. Patients with an abnormal retropulsion test (score ≥ 1) also received a light touch on their shoulders to explore the presence (S-Touch+) or absence (S-Touch-) of an incongruent, exaggerated postural response, defined as taking three or more steps to recover or a fall if not caught by the examiner.

Results: From a total sample of 52 FMD patients, 48 patients were recruited. Twenty-five patients (52%) had an abnormal retropulsion test. Twelve of these 25 patients (48%) had an S-Touch+, either because of need to take two or more steps ($n = 4$) or a fall if not caught by the examiner ($n = 8$). None of the 23 PD/PSP patients manifested S-Touch+. The sensitivity of the S-Touch test was 48%, whereas its specificity was 100%.

Conclusion: The S-Touch test has a high specificity, albeit with a modest sensitivity, to reveal a functional etiology of postural instability in persons with FMD.

KEYWORDS

balance, functional motor disorders, functional neurological disorders, retropulsion test

INTRODUCTION

Patients with functional motor disorders (FMD) might experience balance impairments [1]. Clinical experience suggests that many of these patients do not report recurrent falls, despite severe subjective balance problems [1]. One possible way to identify the functional nature of postural instability is the use of distraction. Distraction has

been explored in both clinical practice [1] and experimental studies using posturography [2, 3] and the results typically show a reduction of postural instability by using motor or cognitive distractive maneuvers. However, this diagnostic procedure is not infallible, as balance impairment is not influenced by distraction in all patients. Therefore, it would be helpful to have access to additional clinical clues that would point towards a functional origin of the balance problems.

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Recently, Coebergh et al. [4] introduced “the shoulder tap test” (i.e., merely applying a gentle tap to the shoulders, which would itself normally be insufficient to cause instability) as a novel diagnostic procedure to test for the presence of exaggerated postural responses in patients with functional gait disorders. Their findings showed such exaggerated responses in around 82% (14 of 17) of the tested individuals with FMD [4]. Conversely, a light touch applied to the patient’s shoulders (which we would define as the “Shoulder-Touch test” [S-Touch]) could be more informative and perhaps provide fewer false-positive results, because a sudden tap could evoke an exaggerated postural response in patients with hyperekplexia or stiff-person syndrome [5].

Therefore, in the present study we explored the role of this new S-Touch, which features a mere light touch applied to the patient’s shoulders, in revealing a possible functional etiology of postural instability in FMD patients. We explored this by applying the S-Touch in FMD patients and in patients with Parkinson’s disease (PD) or progressive supranuclear palsy (PSP) with postural instability.

METHODS

In this observational cross-sectional study, we enrolled consecutive outpatients with a definite diagnosis of FMD [6] between August 31, 2019 and December 31, 2021 attending the Neurology Unit, Department of Neurosciences, Biomedicine and Movement Sciences, University of Verona in Italy. To assess the performance of the S-Touch test, we further recruited 23 patients with PD or PSP, all of whom scored ≥ 1 for the item 3.12 “Postural stability” of the Movement Disorder Society-Sponsored Revision of the Unified Parkinson’s Disease Rating Scale (MDS-UPDRS) [7]. In both groups, exclusion criteria were: (i) cognitive or physical impairments that impeded the patient from properly signing the informed consent form for participation in the study [8] and (ii) concomitant neurological diseases affecting postural control, in particular patients with clinical and neurophysiological (such as electromyography, transcranial magnetic stimulation and electroencephalography) characteristics suggesting hyperekplexia, stiff person syndrome and cataplexy in the setting of narcolepsy.

Assessment

Each patient underwent a detailed clinical evaluation and video recording during the neurological examination. We collected clinical and demographic details as described elsewhere [8, 9]. Then, we tested corrective balance responses under two different conditions: (i) during the retropulsion test, a quick forceful pull on the patient’s shoulder while the patient is standing erect with eyes open and feet comfortably apart and parallel to each other [7] and (ii) with the S-Touch test, which features a light touch, instead of forceful pull, on the patient’s shoulders. These conditions were randomly applied and always preceded by one practice pull. A clinical expert in movement

disorders rated the balance correcting responses of both tests as recommended by item 3.12 “Postural stability” of the MDS-UPDRS [7] and described the pattern of the balance-correcting response. Patients with an abnormal retropulsion test (score ≥ 1) were categorized according to whether they displayed a positive S-Touch test (S-Touch+: score ≥ 1 for item 3.12 “Postural stability” of the MDS-UPDRS) or not (S-Touch–: score = 0 for item 3.12 “Postural stability” of the MDS-UPDRS).

Statistical analysis

Data were expressed as mean \pm standard deviation (SD) for continuous variables, and as counts and percentages for categorical variables. Comparisons of demographic and clinical features between FMD patients S-Touch+ and S-Touch– were performed using the One-Way ANOVA or Mann-Whitney U-test for continuous variables and the chi-squared test or Fisher’s exact test (if ≤ 5 expected frequencies) for categorical variables. Moreover, comparisons of demographic and clinical features between patients with and without postural instability using the retropulsion test scored with item 3.12 “Postural stability” of the MDS-UPDRS were also performed using the above tests. The sensitivity of the S-Touch test was calculated as the number of functional patients S-Touch+/number of functional patients with a score ≥ 1 on the retropulsion test, whereas its specificity was calculated as number of non-functional patients with S-Touch–/number of non-functional patients with a score ≥ 1 on the retropulsion test. Statistical analyses were performed using SPSS statistical software (version 20; IBM-SPSS).

RESULTS

We recruited a total of 52 patients with FMD; the data from 4 patients were excluded from analysis because video recordings and/or detailed clinical information were missing, thus leaving 48 patients for the final analysis. Their mean age at onset was 37 ± 16 years; and 41 (85.4%) were women. Twenty-five FMD patients (52%) were not able to recover from the retropulsion test with one or two steps, and therefore had a score of 1 or higher. Of these, 12 (48%) had a S-Touch+, implying that they needed more than two steps to recover from a light touch to the shoulders ($n = 4$) or that they fell ($n = 8$). See the accompanying video for several illustrative examples (Video S1).

We recruited a total of 23 control patients, 9 with PD and 14 with PSP, 13 men and 10 women with a mean age of 68.7 ± 4.8 years. None of these 23 patients with PD or PSP had an S-Touch+. According to these figures, the sensitivity of the S-Touch test was 48%, whereas its specificity was 100%. The comparison between patients with postural instability stratified according to the S-Touch test (i.e., S-Touch+ vs. S-Touch–) only revealed a lower frequency of pain in the former group ($p = 0.039$; Table 1). Finally, the comparisons between FMD patients with and without postural instability (as assessed by the retropulsion test scored with item 3.12 “Postural stability”

TABLE 1 Comparison of clinical and demographical features of functional motor disorders patients with S-Touch+ and S-Touch-

| Variable | Total (n = 25) | S-Touch- (n = 13) | S-Touch+ (n = 12) | Group comparison |
|--|----------------|-------------------|-------------------|--------------------------|
| Sex, female, n (%) | 22 (88) | 11 (84.6) | 11 (91.7) | 1000 ^F |
| Age, years, mean (SD) | 42.1 (15.3) | 39 (14.5) | 45.4 (16.1) | 0.306 ^A |
| Time lag from onset of symptoms to FMD diagnosis, years, mean (SD) | 2.9 (3.8) | 1.8 (1.8) | 4.2 (5) | 0.060 ^M |
| FMD phenotype, n (%) | | | | |
| Weakness | 24 (96) | 13 (100) | 11 (91.7) | 0.480 ^F |
| Gait disorders | 21 (84) | 11 (84.6) | 10 (83.3) | 1000 ^F |
| Tremor | 17 (68) | 10 (76.9) | 7 (58.3) | 0.411 ^F |
| Dystonia | 4 (16) | 3 (23.1) | 1 (8.3) | 0.593 ^F |
| Jerks | 2 (8) | 1 (7.7) | 1 (8.3) | 1000 ^F |
| Facial movement disorders | 4 (16) | 2 (15.4) | 2 (16.7) | 1000 ^F |
| Parkinsonism | 1 (4) | 0 | 1 (7.7) | 1000 ^F |
| Self-reported non-motor symptoms, n (%) | | | | |
| Fatigue | 22 (88) | 12 (92.3) | 10 (83.3) | 0.593 ^F |
| Pain | 21 (84) | 13 (100) | 8 (66.7) | 0.039^F |
| Headache | 16 (64) | 7 (53.8) | 9 (75) | 0.411 ^F |
| Anxiety | 13 (52) | 7 (53.8) | 6 (50) | 1000 ^C |
| Insomnia | 13 (52) | 7 (53.8) | 6 (50) | 1000 ^C |
| Depersonalization/derealization | 9 (36) | 6 (46.2) | 3 (25) | 0.411 ^F |
| Panic attacks | 5 (20) | 3 (23.1) | 2 (16.7) | 1000 ^F |
| Neurological comorbidities, n (%) | 6 (24) | 1 (7.7) | 5 (41.7) | 0.073 ^F |
| Non-neurological comorbidities, n (%) | 11 (44) | 6 (46.2) | 5 (41.7) | 1000 ^F |
| Psychiatric comorbidities, n (%) | 3 (12) | 1 (7.7) | 2 (16.7) | 0.593 ^F |
| Associated FND, n (%) | | | | |
| Sensory functional symptoms | 16 (64) | 9 (69.2) | 7 (58.3) | 0.688 ^F |
| Non-epileptic seizures | 6 (24) | 4 (30.8) | 2 (16.7) | 0.645 ^F |
| Visual functional symptoms | 7 (28) | 3 (23.1) | 4 (33.3) | 0.673 ^F |
| Cognitive functional symptoms | 10 (40) | 6 (46.2) | 4 (33.3) | 0.688 ^F |
| Fibromyalgia | 4 (16) | 1 (7.7) | 3 (25) | 0.322 ^F |
| Functional bowel syndrome | 3 (12) | 1 (7.7) | 2 (16.7) | 0.593 ^F |
| Precipitating factors, n (%) | | | | |
| Surgery | 6 (24) | 2 (15.4) | 4 (33.3) | 0.378 ^F |
| Physical trauma | 4 (16) | 1 (7.7) | 3 (25) | 0.322 ^F |
| Psychological trauma | 1 (4) | 1 (7.7) | 0 | 1000 ^F |
| General anesthesia | 3 (12) | 1 (7.7) | 2 (16.7) | 0.593 ^F |
| Type of trigger, n (%) | | | | |
| Exercise/movement | 15 (60) | 9 (69.2) | 6 (50) | 0.428 ^F |
| Emotional | 1(4) | 0 | 1 (8.3) | 0.480 ^F |
| Visual | 4 (16) | 2 (15.4) | 2 (16.7) | 1.000 ^F |
| Touch | 0 | 0 | 0 | - |
| Auditory | 0 | 0 | 0 | - |

Note: Statistical testing (denoted by superscript letters): One-Way ANOVA (A), Mann-Whitney U-test (M), Kruskal-Wallis test (K), Chi-square test (C), Fisher's exact test (F); significant values at $p < 0.05$ in bold.

Abbreviations: FMD, functional motor disorders; FND, functional neurological disorders; SD, standard deviation; S-Touch, Shoulder-Touch test; S-Touch+, a positive Shoulder-Touch test with Movement Disorder Society-Sponsored Revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS) score for postural instability ≥ 1 ; S-Touch-, no postural instability with MDS-UPDRS score for postural instability rated zero.

of the MDS-UPDRS; Table S1) showed that the former were older ($p = 0.023$), had more frequent functional weakness ($p = 0.044$) and fewer emotional triggers ($p = 0.020$) than the latter.

DISCUSSION

In this study we found that 25 of 48 FMD patients (52%) made three or more steps or fell in response to the standard retropulsion test, which is indicative for postural instability. Additionally, we found that among these 25 patients, 12 (48%) showed a positive response to the S-Touch test, suggesting incongruency. Given that none of the included patients with PD or PSP had an S-Touch+, this indicates that the S-Touch test has a very high specificity for revealing a functional etiology of postural instability in FMD patients. Comparison between FMD patients with S-Touch+ and S-Touch- did not reveal any differences (with the exception of pain), which suggests that this test does not identify a particular subtype of patients with FMD, but is only useful as a clinical complement to the neurological examination to reveal incongruencies.

Several other tests have previously been introduced to reveal incongruencies pointing to a functional nature of the disorder, including a marked reduction of postural sway during distractive motor tasks [2] or cognitive tasks [3, 10].

Similar to our S-Touch test, none of these other tests has a perfect sensitivity. The S-Touch is therefore a useful addition to the clinical test repertoire to reveal possible incongruencies. The test should not be used in isolation, but always in combination with other gait and balance tests [1].

Our findings are in line with a previous study in which exaggerated (i.e., incongruent) postural responses (postural stability score ≥ 1 for item 3.12 "Postural stability" of the MDS-UPDRS) were described in patients with functional gait disorder using the shoulder tap test [4]. However, drawbacks of this latter study were the relatively small sample size ($n = 17$) and the fact that a vigorous tap on patients' shoulders, unlike the S-Touch test, could evoke an exaggerated postural response not only in individuals with FMD, but presumably also in patients with hyperekplexia or stiff-person syndrome – such a test result might then incorrectly be classified as an incongruent response [4]. This argument, however, remains speculative because in our study we did not compare the two tests in the same population of patients.

Comparisons between FMD patients with and without postural instability did not reveal gross differences in terms of clinical features. Patients with postural instability were older than those without, possibly suggesting that they perceived age-related declines in postural stability [11]. Interestingly, we showed that postural instability did not cluster necessarily with presence of a functional gait disorder, as assumed in earlier literature [4], but also not with other FMDs, especially functional weakness.

We acknowledge some methodological limitations. First, because of the scope of our work, we stratified our population according to a score ≥ 1 for item 3.12 "Postural stability" of the MDS-UPDRS,

regardless of the presence of a self-reported balance questionnaire or more specific balance outcomes. Future studies could relate a positive outcome of the S-Touch test to perceived balance impairment. Second, we evaluated the S-Touch only in patients with an abnormal retropulsion test but it is possible that patients with normal retropulsion test and/or without a subjective complaint of postural disability could have a S-Touch+. Future studies should evaluate these aspects using also specific self-reported balance measures.

Third, FMD and organic disorders can co-exist [12, 13]. It is therefore possible that a positive retropulsion test in FMD patients (combined with a negative S-Touch test) was due to comorbidities (although major neurological comorbidities were an exclusion criterion) impacting on the calculated sensitivity.

In summary, our study shows that postural instability is relatively frequent in FMD and that the S-Touch test, despite not being sensitive, has a very high specificity to reveal its functional etiology.

AUTHOR CONTRIBUTIONS

Christian Geroin: Conceptualization (lead); data curation (lead); formal analysis (lead); investigation (lead); methodology (lead); writing – original draft (lead); writing – review and editing (lead). **Jorik Nonnekes:** Conceptualization (equal); data curation (equal); writing – review and editing (equal). **Roberto Erro:** Resources (equal); writing – review and editing (equal). **Serena Camozzi:** Conceptualization (equal); data curation (equal); writing – review and editing (equal). **Bastiaan R. Bloem:** Conceptualization (equal); data curation (equal); writing – review and editing (equal). **Michele Tinazzi:** Conceptualization (lead); data curation (lead); investigation (lead); methodology (lead); supervision (lead); writing – original draft (lead); writing – review and editing (lead).

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CONFLICT OF INTEREST

None of the authors has any conflicts of interests to declare.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICAL APPROVAL

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The study was approved by the Institutional Ethics Committee of the Azienda Ospedaliera Universitaria Integrata Verona, Prog. 1757CESC.

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SUPPORTING INFORMATION

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