## RESEARCH ARTICLE

# Impaired Metacognition of Voluntary Movement in Functional Movement Disorder

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ABSTRACT: Background: Motor symptoms in functional movement disorders (FMDs) are experienced as involuntary but share characteristics of voluntary action. Clinical and experimental evidence indicate alterations in monitoring, control, and subjective experience of self-performed movements.

**Objective:** The objective of this study was to test the prediction that FMDs are associated with a reduced ability to make accurate (metacognitive) judgments about self-performed movements.

Methods: We compared 24 patients with FMD (including functional gait disturbance, functional tremor, and functional tics) with 24 age- and sex-matched healthy control subjects in a novel visuomotor-metacognitive paradigm. Participants performed target-directed movements on a graphics tablet with restricted visual feedback, decided which of two visually presented trajectories was closer to their preceding movement, and reported their confidence in the visuomotor decision. We quantified individual metacognitive performance as participants' ability to assign high confidence preferentially to correct visuomotor decisions.

Results: Patients and control subjects showed comparable motor performance, response accuracy, and use of the confidence scale. However, visuomotor sensitivity in the trajectory judgment was reduced in patients with FMD compared with healthy control subjects. Moreover, metacognitive performance was impaired in patients, that is, their confidence ratings were less predictive of the correctness of visuomotor decisions. Exploratory subgroup analyses suggest metacognitive deficits to be most pronounced in patients with a functional gait disturbance or functional tremor.

Conclusions: Patients with FMD exhibited deficits both when making visuomotor decisions about their own movements and in the metacognitive evaluation of these decisions. Reduced metacognitive insight into voluntary motor control may play a role in FMD pathophysiology and could lay the groundwork for new treatment strategies. © 2023 The Authors. *Movement Disorders* published by Wiley Periodicals LLC on behalf of International Parkinson and Movement Disorder Society.

**Key Words:** functional movement disorder; metacognition; motor control; visuomotor

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## Introduction

Motor symptoms in functional movement disorders (FMDs) are experienced as involuntary but exhibit fundamental characteristics of voluntary movement: motor symptoms typically aggravate with attention and improve with distraction, phasic pathological movements have been shown to be preceded by a readiness potential,<sup>2</sup> and functional tremor is entrained by concurrent rhythmic movement of another limb.<sup>3</sup> These findings set FMD apart from non-FMDs and serve as positive diagnostic criteria. They also suggest that the subjective experience of movement is altered in FMD. In line with this, experimental signatures of voluntary motor control, such as action-effect binding<sup>4</sup> and sensory attenuation of action effects, 5 have been found to be reduced in FMD compared with healthy control participants (HCs).

Besides changes in subjective experience, exaggerated self-directed attention and expectation of conscious control over motor actions are hypothesized to play an important role in the genesis and maintenance of FMD.<sup>6,7</sup> In a visuomotor monitoring paradigm, patients with FMD showed increased sensitivity for task-irrelevant and reduced sensitivity for task-relevant perturbations compared with HCs; moreover, motor performance improved, in patients with FMD only, when movements were performed without attention.<sup>8</sup> Despite the detrimental effect of self-directed attention, patients with FMD have been found to show increased gaze toward the affected limb during clinical examination.<sup>9</sup>

The clinical and experimental evidence reviewed earlier suggests alterations in control and subjective experience of movement in patients with FMD, likely affecting both the ability to make accurate judgments about selfgenerated movements and the metacognitive insight into these judgments. However, to our knowledge, only two studies have investigated metacognition in patients with FMD experimentally. Patients with FMD showed lower performance compared with HC in a visuomotor perturbation detection task, but trial-per-trial confidence ratings did not indicate metacognitive deficits in patients with FMD, 10 possibly because of a lack of statistical power. A second study used a visual contrast task<sup>11</sup> and demonstrated positive evidence (using Bayesian statistics) for intact metacognitive ability in patients with FMD. This speaks against a general metacognitive deficit in FMD. However, given that metacognitive abilities are, to some extent, domain-specific, 12,13 the question of a metacognitive deficit related to motor actions remains open. Addressing this question is important for a better understanding of FMD and has the potential to inform novel therapeutic approaches.

In this study, we compared visuomotor sensitivity and metacognitive ability in self-generated hand movements between patients with FMD and HCs. We developed a task combining visuomotor judgments about self-performed movements with metacognitive ratings: participants perform goal-directed movements on a graphics tablet with restricted visual feedback, then decide which of two visually presented trajectories is closer to their own preceding movement, and finally report their confidence in the preceding visuomotor decision. Metacognitive insight into the visuomotor decision is assessed using established measures relating confidence ratings to response accuracy on a trial-by-trial basis. <sup>14</sup> Based on the clinical and experimental evidence discussed earlier, we predicted that, relative to HCs, patients with FMD would exhibit both a reduced visuomotor sensitivity in trajectory decisions and lower metacognitive performance.

## Subjects and Methods

## **Participants**

Twenty-four participants with a diagnosed FMD (15 female, 39.7 ± 16.0 years old [mean ± standard deviation], range 16–68 years, 2 left-handed) and 24 HCs (15 female, 40.3 ± 16.3 years old, range 16–68 years, 2 left-handed) were included in this study. Participants from the two groups were matched pairwise with respect to sex and age (±6 years). Data from one additional patient with FMD and two HCs were not used for data analysis for these reasons: the patient with FMD strongly deviated from the target accuracy level in the trajectory judgment (52%; target level, 71%; range of other participants, 65.1%–74.5%), one HC misunderstood the trajectory judgment task, and one HC took antidepressant medication at the time of the experiment.

Patients with FMD were recruited from the outpatient clinics of the Departments of Neurology and the Center for Rare Diseases of the University Hospital Schleswig-Holstein, Campus Lübeck, and the Department of Neurology of the University Hospital Hamburg-Eppendorf, Hamburg, Germany. The clinical diagnosis was made according to standard published criteria. All patients underwent a detailed videorecorded clinical assessment and a subsequent video rating of symptoms by a movement disorders specialist (A.W.) using the Clinical Global Impression-Severity scale (CGI-S<sup>15</sup>;  $4.0 \pm 1.0$ ) and the Simplified-Functional Movement Disorders Rating Scale (S-FMDRS<sup>16</sup>;  $10.0 \pm 6.5$ ). Age at disease onset (34.7  $\pm$  15.6 years) and disease duration (5.0  $\pm$  5.1 years) were noted. In addition, patients with FMD were categorized according to their main motor symptoms; functional gait disturbance (n = 16), functional tremor (n = 9), functional tics (n = 5), or other functional symptoms (n = 2). Eight patients had both a functional gait disturbance and functional tremor. Intelligence quotient (IQ) was estimated using a validated short form of the Wechsler Adult Intelligence Scale. <sup>17</sup> IQ could not be assessed in one patient because of severe fine motor impairment (but no apparent cognitive deficit during clinical assessment and experiment). At the time of participation, none of the patients with FMD had clinically relevant psychiatric or additional functional symptomatology. HCs were recruited through an announcement at the billboard of the University Clinic of Schleswig Holstein, Campus Lübeck. See Supporting Information Tables S1 and S2 for individual characteristics.

Participants took part after written informed consent according to the Declaration of Helsinki. The study was approved by the local Ethics Committee (reference: 20–136).

## Visuomotor-Metacognitive Paradigm

The experimental paradigm comprised three task components on each trial (see Fig. 1). Participants made center-out movements with a pen on the graphics tablet, controlling the movement of a cursor on the screen from a starting area through a target presented at one of eight possible positions in the upper right quadrant (upper left for participants using their left hand). The cursor was shown continuously between trials and during initial task familiarization. During the main experiment, target and cursor disappeared immediately after movement onset. Participants were then shown two candidate trajectories on the screen, and they moved the pen on the tablet to choose the one closer to their preceding movement. With limited visual feedback during the motor task, this trajectory judgment had to rely on an internal visuomotor representation of the preceding movement. One of the trajectories corresponded to their actual movement (corrected by the estimated visuomotor bias; see Supporting Information, section 2.2); the alternative trajectory was a left- or right-rotated copy of the actual trajectory. The direction of the deviation (left/right) was randomized in subblocks of eight trials, to ensure an even distribution across the experiment. Task difficulty, that is, the angular deviation between trajectories, was adjusted using a two-down one-up adaptive procedure (step size 2°), targeting 71% response accuracy. Two separate adaptive procedures were used for leftward and rightward deviations to account for potential visuomotor bias (ie, angular deviation between the actual and the subjective mapping between tablet and screen). Finally, participants rated their confidence in the preceding trajectory judgment on a continuous visual analog scale, again by moving the pen on the tablet.

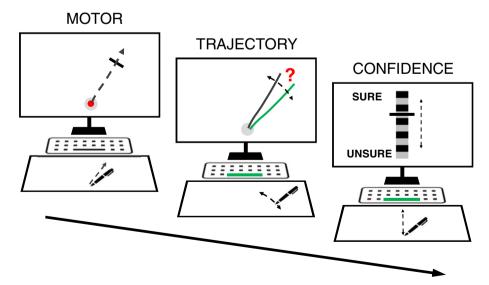
The three task components were gradually introduced for familiarization with the experimenter sitting next to the participant for instruction. The main experiment with all three components consisted of 192 trials (three blocks of 64 trials). Details about the stimuli and procedure for each task component are provided in the Supporting Information (section 2). The total duration of the experiment, including familiarization, was about 50 minutes.

## **Data Analysis**

Experimental data were processed in Matlab, using custom-written code and publicly available Matlab code (http://www.columbia.edu/~bsm2105/type2sdt/) for type-2 signal- detection-theoretic analysis.<sup>20</sup>

#### Motor Task

Motor performance was quantified in terms of movement duration and movement error (angular deviation between target direction and movement direction when



**FIG. 1.** Setup and task components. Participants made center-out, target-directed movements on a graphics tablet with limited visual feedback onscreen, decided which of two trajectories was closer to their actual movement by moving the pen on the tablet, and rated their confidence in the trajectory judgment on a visual analog scale. [Color figure can be viewed at wileyonlinelibrary.com]

crossing the task radius). Both the average signed  $(\pm)$  and absolute movement error were used to quantify movement accuracy.

### Trajectory Judgment

Response accuracy was quantified as the percentage of correct trajectory judgments and the signal-detection measure d', which controls for response bias and scaling artefacts (ie, compression near the boundaries of the 0%–100% scale). With response accuracy stabilized by the adaptive procedure, the mean task difficulty (absolute deviation between trajectories) is the main performance measure at this level. Better performance, that is, higher visuomotor sensitivity to discriminated self-generated from alternative trajectories, is indicated by a smaller angular deviation. The mean absolute visuomotor bias estimated by the adaptive procedure provides an additional measure of the accuracy of the visuomotor representation.

## **Confidence Ratings**

Confidence ratings were not an outcome measure of interest by themselves. Still, mean and range of reported confidence were determined to test for extreme response patterns (outliers) and to assess potential group differences, which might affect measures of metacognitive performance.

#### Metacognitive Performance

Metacognitive sensitivity was quantified by meta-d' introduced by Maniscalco and Lau,<sup>20</sup> using the maximum likelihood estimator approach. In brief, meta-d' is estimated by determining the (hypothetical) d' value that maximizes the likelihood of the observed type-2 (confidence ratings) hit and false alarm rates under a signal-theoretic model assuming full use of the available type-1 (trajectory judgment) information. 20 Because this analysis is based on discrete confidence ratings, the continuous (0–100) confidence ratings were binned by dividing each participant's total confidence range (minimum-maximum) into four equal-sized intervals. To control for a potential influence of type-1 accuracy on metacognitive sensitivity,<sup>20</sup> we computed metacognitive efficiency as the ratio (M-ratio) meta-d'/d'. This was our main measure of metacognitive performance. An M-ratio of 1.0 indicates that the full information used for the trajectory judgment is used for the confidence rating. Smaller values indicate that not all the information is used, whereas larger values indicate that more information is used for the metacognitive than for the trajectory judgment. Thus, larger values indicate better metacognitive performance.

In addition, a nonparametric measure of metacognitive sensitivity, meta-area under the receiver operating curve (AUROC relating confidence to response accuracy) was computed.<sup>14</sup> Meta-AUROC quantifies to what extent confidence ratings discriminate between incorrect and correct responses. Values range between 0 and 1, with larger values indicating better metacognitive performance. Meta-AUROC is known to be affected by type-1 accuracy<sup>14</sup>; however, unlike meta-d' and the M-ratio, meta-AUROC makes use of the full confidence scale (no discretization required) and, as a nonparametric measure, does not rely on model assumptions being satisfied and also avoids potential problems with model fitting.

## Statistical Analysis

Because this was the first study using this visuomotor-metacognitive paradigm, meaningful sample size calculations were not possible. Our final sample size of 24 per group exceeds those of most previous studies on action control and awareness in FMD.<sup>4,5,8,10</sup>

Statistical analyses were performed in R 4.1.2.<sup>21</sup> Dependent variables were compared between groups by two-sample Welch t tests (no equal variance assumed). The nonparametric Mann–Whitney test was used instead when data from one or both groups deviated from normal distribution (as assessed by the Shapiro–Wilk test). Effect sizes are reported using a robust version of Cohen's d, denoted  $d_R$ .<sup>22</sup> Bayes factors quantifying the evidence in favor of the alternative hypothesis (BF<sub>10</sub>) were computed.<sup>23,24</sup> All statistical tests were two-sided. The criterion for statistical significance testing was P < 0.05. A Bayes factor BF<sub>10</sub> > 3 was interpreted as substantial and BF<sub>10</sub> > 10 as strong evidence against the null hypothesis.

Associations between individual characteristics (age, disease onset and duration, clinical scales and symptoms) and the main experimental measures (visuomotor sensitivity and metacognitive efficiency) were assessed within the group of patients with FMD using Pearson correlation tests and exploratory subgroup analyses.

# **Data Sharing**

Original data and analysis scripts (Matlab, R) are available from a public OSF repository (https://osf.io/qc5uf/).

## Results

Sample data from two representative participants are shown in Fig. 2. These participants had comparable basic performance in the motor and trajectory task, as well use of the confidence scale. However, the relation between confidence ratings and response accuracy in the trajectory task, shown in the right panel of Fig. 2, is markedly different. In the HC participant (Fig. 2, top panels), the relation between correct and incorrect responses changes from low confidence (more incorrect

METACOGNITION IN FUNCTIONAL MOVEMENT DISORDER

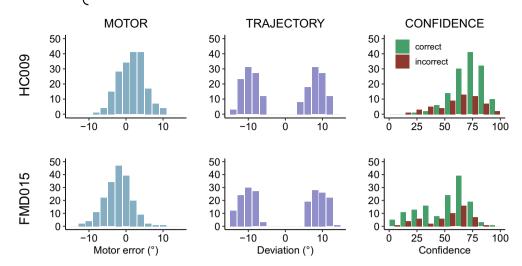


FIG. 2. Sample data from participants (HC009: healthy control; FMD015: patient with functional movement disorder) with comparable performance in task components but higher (top) versus lower (bottom) metacognitive performance. Motor: angular movement error. Trajectory: angular deviation between central and deviant trajectories. Confidence: ratings for trials with correct versus incorrect trajectory judgments. [Color figure can be viewed at wileyonlinelibrary.com]

responses) to high confidence (more correct responses), as expected for "well-calibrated" metacognition. In contrast, the relation between correct and incorrect responses remains stable across the entire confidence range in the patient with FMD (Fig. 2, bottom panels), suggesting lower metacognitive performance.

#### **Motor Task**

No systematic group differences were observed concerning angular movement error or movement duration (Table 1, Supporting Information Fig. S1).

## **Trajectory Judgment**

For the trajectory judgment, the adaptive procedure resulted in comparable accuracy levels in both groups (Fig. 3A), although response accuracy tended to be low in patients with FMD compared with HCs (see Table 1 for details). Quantified using d' (Fig. 4A), the group difference was more pronounced, emphasizing the importance of correcting metacognitive sensitivity (meta-d') for type-1 performance.

Importantly, the adaptive procedure resulted in larger deviations between alternative trajectories (Fig. 3B), that is, lower difficulty levels, in patients with FMD compared with HCs, indicating a reduced visuomotor sensitivity in patients with FMD compared with HCs. This finding is strengthened by the fact that, despite lower task difficulty, response accuracy was lower in patients with FMD. The absolute trajectory bias was larger for patients with FMD as well (Supporting Information Fig. S2A), providing additional evidence of a less accurate visuomotor representation of self-performed movements. Bayes factors >3 indicate substantial evidence for group differences in these measures, with medium to large effect sizes.

## **Confidence Rating**

Participants varied substantially in mean and range of confidence ratings (Supporting Information Fig. S3), but no significant group differences were observed for these measures (Table 1).

## Metacognitive Performance

A significant group difference was found for *meta-d'* (Fig. 4A; see Table 1 for details), with lower metacognitive sensitivity in patients with FMD compared with HC. This group difference persisted after correcting for type-1 performance using the M-ratio (meta-d'/d', Fig. 4B), confirming lower metacognitive efficiency in patients with FMD compared with HCs. A significant group difference in the same direction was found for meta-AUROC (Supporting Information Fig. S4). This convergent evidence from an independent nonparametric measure of metacognitive performance, using the full confidence scale, indicates that the observed group difference in metacognition is robust and not due to a particular choice of analysis method or potential problems with model fitting. Bayes factors >3 indicate substantial evidence for group differences in metacognitive measures, with large effect sizes ( $|d_R|$ between 0.76 and 0.96).

### **Control Analyses**

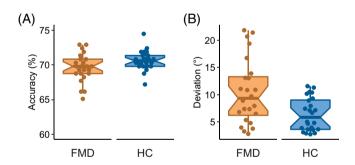
Apparent outliers were noted for each of the three task components (motor, trajectory, confidence; Supporting Information Results, section 2.2 and Figs. S2–S4). Repeating the main analyses after excluding these outliers (along with their matched participants) in all cases confirmed the results reported earlier (Supporting Information, Tables S3–S5). In fact, group differences in

 TABLE 1
 Group comparisons for main dependent measures

Variable	FMD (mean $\pm$ SD)	HC (mean $\pm$ SD)	Statistics <sup>a</sup>
Motor performance			
Motor error (°)	$2.68 \pm 5.80$	$1.10 \pm 3.80$	$W = 320.0, P = 0.519, d_R = 0.20, BF_{10} = 0.36$
Absolute error (°)	$5.77 \pm 4.12$	$4.29 \pm 1.87$	$W = 378.0, P = 0.065, d_R = 0.57, BF_{10} = 1.14$
Duration (s)	$0.17 \pm 0.05$	$0.19 \pm 0.06$	$t(44.4) = -1.47, P = 0.149, d_R = -0.36, BF_{10} = 0.69$
Trajectory judgment			
Accuracy (%)	$69.60 \pm 2.02$	$70.64 \pm 1.39$	$t(40.7) = -2.08, P = 0.044, d_R = -0.69, BF_{10} = 1.61$
Accuracy (d')	$1.05 \pm 0.15$	$1.14 \pm 0.10$	$t(39.2) = -2.52, P = 0.016, d_R = -0.87, BF_{10} = 3.49$
Deviation (°)	$10.48 \pm 5.94$	$6.39 \pm 3.01$	$W = 409.0, P = 0.012, d_R = 0.69, BF_{10} = 3.98$
Absolute bias (°)	$6.05 \pm 5.14$	$3.02 \pm 2.61$	$W=416.0, P=0.008, d_R=0.61, { m BF}_{10}=6.57$
Confidence ratings			
Mean confidence	$58.24 \pm 19.17$	$53.56 \pm 16.26$	$t(44.8) = 0.91, P = 0.366, d_R = 0.24, BF_{10} = 0.40$
Confidence range	$76.00 \pm 23.31$	$82.21 \pm 19.75$	$W = 237.5, P = 0.302, d_R = -0.31, BF_{10} = 0.43$
Metacognitive perform	ance		
Meta-d'	$0.44 \pm 0.37$	$0.78 \pm 0.44$	$t(44.8) = -2.86, P = 0.006, d_R = -0.92, BF_{10} = 6.96$
M-ratio	$0.42 \pm 0.36$	$0.67 \pm 0.36$	$t(46.0) = -2.45, P = 0.018, d_R = -0.76, BF_{10} = 3.09$
Meta-AUROC	$0.58 \pm 0.05$	$0.62 \pm 0.06$	$t(45.4) = -2.47, P = 0.017, d_R = -0.96, BF_{10} = 3.18$

Abbreviations: FMD, functional movement disorder; HC, healthy control; SD, standard deviation;  $d_R$ , robust version of Cohen's d; AUROC, area under the receiver operating curve.

<sup>&</sup>lt;sup>a</sup>Two-sample t test or Mann–Whitney test comparing the two groups (FMD and HC). Bayes factor BF<sub>10</sub> indicates evidence *against* the null hypothesis. All tests are two-sided. Significant effects (P < 0.05) are in boldface.



**FIG. 3.** Performance in the trajectory judgment. (A) Response accuracy. (B) Mean absolute trajectory deviation. Larger deviations indicate lower performance at a given accuracy level. Individual data and box plots, showing median, interquartile range (IQR), and whiskers 1.5 IQR beyond the boxes. FMD, functional movement disorder; HC, healthy control. [Color figure can be viewed at wileyonlinelibrary.com]

visuomotor sensitivity and metacognitive performance became numerically more pronounced (larger absolute effect sizes) when excluding outliers.

Variability in trajectory deviations across the experiment was larger in patients with FMD compared with HCs (W = 391, P = 0.034; Supporting Information Fig. S2B). Because increased type-1 variability has been shown to result in inflated measures of metacognitive performance, <sup>25</sup> the effects in metacognitive performance reported earlier likely underestimate the actual group differences.

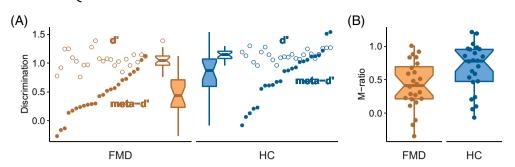
Two patients with FMD performed the motor task with their nondominant left hand, which could affect their ability to gauge self-performed movements. Repeating the main analyses after excluding these participants, along with their matched HCs, corroborated the main results, with lower visuomotor sensitivity and metacognitive performance in patients with FMD compared with HCs (Supporting Information Table S6).

IQ scores tended to be lower in patients with FMD  $(97.7 \pm 10.2)$  than in HCs  $[103.0 \pm 7.9; t$  (41.3) = -1.83; P = 0.074], which may be partially explained by fine motor impairments. Still, we wanted to control for a potential confounding effect of IQ. Including only matched participant pairs with IQ between 85 and 115 (n = 19 + 19) successfully equalized IQ between groups  $(P = 0.48, BF_{10} = 0.34)$  but preserved significant group differences in visuomotor sensitivity and metacognitive efficiency (Supporting Information Table S7).

## **Association with Clinical Characteristics**

Individual characteristics and clinical measures (age, disease onset, disease duration, CGI, S-FMDRS) did not show significant correlations with visuomotor sensitivity or metacognitive efficiency (Supporting Information Table S8).





**FIG. 4.** Metacognitive performance. (**A**) Response accuracy (d') and metacognitive sensitivity (*meta-d*') per participant, ordered by *meta-d*'. (**B**) Metacognitive efficiency (M-ratio), which normalizes *meta-d*' with respect to d'. For each measure, larger values indicate higher performance. FMD, functional movement disorder; HC, healthy control. [Color figure can be viewed at <u>wileyonlinelibrary.com</u>]

Patients with FMD were broadly (and nonexclusively) categorized as exhibiting functional gait symptoms (n = 16), functional tremor (n = 9), functional tics (n = 5), or other functional symptoms (n = 2). Participant numbers, overlap between categories (n = 8 had both gait symptoms and tremor), and the imbalance in group sizes precluded a formal statistical comparison between subgroups. We explored potential influences of the kind of motor symptoms by considering the following subgroups (patients with their matched HCs): gait, tremor, and tics (see Supporting Information Figures S6 and S7). Restricting the analyses to patients with FMD with gait disturbance, as well as their matched HCs (n = 16 + 16), showed qualitatively similar and numerically more pronounced group differences in visuomotor sensitivity (P = 0.003;  $d_R = 1.18$ ;  $BF_{10} = 15.8$ ) and metacognitive efficiency (P = 0.001;  $d_R = -1.62$ ; BF<sub>10</sub> = 28.69), with very large effect sizes and strong Bayesian evidence for a group difference (Supporting Information Table S9). Similarly, restricting the analysis to patients with functional tremor (n = 9)+ 9) resulted in numerically more pronounced group differences in visuomotor sensitivity and metacognitive efficiency (Supporting Information Table S10). Importantly, these subgroup analyses confirmed the absence of group differences in basic motor performance (P > 0.3; $|d_R|$  < 0.3; BF<sub>10</sub> < 0.5 in all cases).

## **Discussion**

We investigated the ability of patients with FMD to make accurate visuomotor and metacognitive judgments about self-performed movements. As hypothesized, patients with FMD exhibited lower visuomotor sensitivity and reduced metacognitive performance compared with HCs. Exploratory subgroup analyses suggest metacognitive deficits to be most pronounced in patients with functional gait disturbance or functional tremor.

When making visuomotor judgments about their own preceding hand movements, patients with FMD

needed larger deviations (ie, lower task difficulty) between alternative trajectories compared with HCs to reach a comparable response accuracy level. This is in line with a previous study showing a reduced sensitivity in patients with FMD compared with HCs to detect visuomotor perturbations. These findings suggest that visuomotor representations of self-generated movements are less precise in patients with FMD compared with HCs. Future studies should test whether this is also the case for other sensory modalities, for example, the somatosensory or proprioceptive domain. 26

Metacognitive performance was quantified by relating confidence ratings to response accuracy of visuomotor decisions on a trial-by-trial basis.<sup>14</sup> Metacognitive efficiency quantified by the M-ratio, 20 which controls for interindividual variation in response accuracy, was significantly reduced in patients with FMD compared with HCs. This was corroborated by a complementary analysis using a nonparametric measure of metacognitive sensitivity. Control analyses ensured group differences in metacognitive performance were not attributable to outliers or potential confounds. The adaptive procedure resulted in comparable accuracy levels but increased variability of task difficulty in the trajectory judgment task in patients with FMD compared with HCs, which has been shown to result in inflated measures of metacognitive performance.<sup>25</sup> Thus, our results likely underestimate the actual group differences in metacognitive performance in this task.

Correlation of experimental measures with clinical characteristics did not show any significant associations. This was not unexpected, because clinical measures assess the severity of motor impairments rather than potential changes in (meta)cognition associated with FMD. The absence of significant correlations, along with the fact that groups did not differ in basic motor task performance, strongly suggests that the observed visuomotor and metacognitive deficits in FMD cannot be explained by motor impairments alone. Because subgroups of patients with functional gait disturbance, functional tremor, or functional tics were unbalanced in size and overlapping, a formal statistical comparison of subgroups was not warranted.

However, restricting the group comparison to patients with a functional gait disturbance or functional tremor (along with their matched HC) resulted in numerically more pronounced group differences, with very large effect sizes and strong Bayesian evidence for lower visuomotor and metacognitive performance in FMD. Thus, in contrast with motor symptom severity, categorical differences in motor symptomology may be associated with differences in the ability to make accurate judgments about one's own movements. This preliminary result should be followed up by studies with more balanced subgroups.

It has been proposed that FMD may be associated with an "excessive expectation of control," leading to "excessive conscious monitoring" of one's own voluntary movements, and ultimately—given that conscious access to sensory and motoric details of actions remains limited—resulting in a perceived loss of voluntary control. This proposal is motivated by experimental findings of more accurate perception of action-related sensory events in patients with FMD compared with HCs. For instance, patients with FMD showed a significant reduction of physiological attenuation of self-generated force stimuli<sup>5</sup> and a reduced compression of the perceived time interval (temporal binding) between a motor action and a resulting sensory consequence.<sup>4</sup> The results of our study indicate that, if indeed present in FMD, over-accuracy with respect to motoric or perceptual details does not benefit visuomotor sensitivity or metacognitive insight related to self-generated movements. On the contrary, these processes may undermine the integration of sensory, motor, and intentional signals characteristic of voluntary action. Interestingly, the explicit sense of agency during voluntary finger tapping has been found to be intact in FMD,<sup>27</sup> suggesting a dissociation from the visuomotor and metacognitive processes investigated in the present study. This is in line with recent evidence that judgments of agency do not rely on the same computational mechanisms as confidence judgments about one's own performance.<sup>28</sup>

The generality of our findings concerning deficient motor metacognition in FMD should be addressed by future studies. For instance, does it generalize to other body parts, movement types, and sensory modalities? Future studies should use (eg, purely visual) control paradigms to separate expected deficits in motor metacognition in FMD from a more general metacognitive deficit. Also, it would be valuable to compare patients with FMD with neurological patients with similar phenomenology but known structural origin of motor symptoms. This would allow clarifying whether observed deficits are specific to FMD or might partly be explained by the experience of (involuntary) pathological movements per se. A recent study including such a nonfunctional control group indicated that misdirection of spontaneous attention and effects of an attentional manipulation on reaching movements are specific to FMD.<sup>8</sup>

A more homogeneous patient sample (eg, only gait disturbance or only functional tremor) may have resulted in more homogeneous experimental outcomes but would have made our sample less representative of the diversity of motor symptoms occurring in FMD. Also, it would have precluded exploratory subgroup analyses. Our groups of patients with FMD and HCs were carefully matched with respect to sex and age, and a control analvsis confirmed group differences in visuomotor and metacognitive performance are not attributable to small group differences in IQ. Performance accuracy in the trajectory judgment ("type-1") task was standardized by an adaptive procedure, because metacognitive measures are affected by type-1 accuracy. 14 In addition, our main measure of metacognitive performance, the "M-ratio," is designed to correct for remaining variation in type-1 accuracy.<sup>20</sup> This is of critical importance for the interpretability of our results. For instance, a recent meta-analysis on metacognition in schizophrenia concluded that reported (and plausibly expected) metacognitive deficits are inflated by (or even fully attributable to) nonequated response accuracy in the primary task.<sup>29</sup>

FMD is a common condition. Up to 20% of patients seen in movement disorders clinics have FMD. 30,31 However, the pattern of care is highly inconsistent, and many patients feel dissatisfied with the treatment. 32,33 If misdirected attention and exaggerated expectation of conscious control are "higher-level" pathophysiological processes in different patients with FMD regardless of clinical phenomenology, then behavioral approaches aiming at flexibly refocusing attention and reexamining potentially harmful metacognitive beliefs might be beneficial. A clinical feasibility study using this approach as an intervention is currently underway. 35

To conclude, a reduction of metacognitive insight into voluntary motor control appears to play a crucial role in the pathophysiology of FMS. This finding is a starting point for the development of novel mechanism-based behavioral interventions.

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#### **Data Availability Statement**

Original data and analysis scripts (Matlab, R) are available from a public OSF repository (https://osf.io/qc5uf/).

## References

 Gasca-Salas C, Lang AE. Neurologic diagnostic criteria for functional neurologic disorders. Handb Clin Neurol 2016;139:193–212.

- Beudel M, Zutt R, Meppelink AM, et al. Improving neurophysiological biomarkers for functional myoclonic movements. Parkinsonism Relat Disord 2018;51:3–8.
- McAuley J, Rothwell J. Identification of psychogenic, dystonic, and other organic tremors by a coherence entrainment test. Mov Disord 2004;19(3):253–267.
- Kranick SM, Moore JW, Yusuf N, et al. Action-effect binding is decreased in motor conversion disorder: implications for sense of agency. Mov Disord 2013;28(8):1110–1116.
- Pareés I, Brown H, Nuruki A, et al. Loss of sensory attenuation in patients with functional (psychogenic) movement disorders. Brain 2014;137(11):2916–2921.
- Edwards MJ, Adams RA, Brown H, Parees I, Friston KJ. A Bayesian account of "hysteria.". Brain 2012;135(11):3495–3512.
- Stenner MP, Haggard P. Voluntary or involuntary? A neurophysiologic approach to functional movement disorders. Handbook of Clinical Neurology. Vol. 139. Amsterdam, NL: Elsevier; 2016: 121–129.
- Huys ACML, Haggard P, Bhatia KP, Edwards MJ. Misdirected attentional focus in functional tremor. Brain 2021;144(11):3436– 3450.
- 9. van Poppelen D, Saifee TA, Schwingenschuh P, et al. Attention to self in psychogenic tremor. Mov Disord 2011;26(14):2575–2576.
- Bègue I, Blakemore R, Klug J, et al. Metacognition of visuomotor decisions in conversion disorder. Neuropsychologia 2018;114: 251–265.
- Matthews J, Nagao K, Ding C, Newby R, Kempster P, Hohwy J. Raised visual contrast thresholds with intact attention and metacognition in functional motor disorder. Cortex J Devoted Study Nerv Syst Behav 2020;125:161–174.
- Arbuzova P, Maurer LK, Filevich E. Metacognitive domains are not aligned along a dimension of internal-external information source. Published online May 4, 2022. doi:https://doi.org/10.1101/2022.05. 03.490468
- Rouault M, McWilliams A, Allen MG, Fleming SM. Human metacognition across domains: insights from individual differences and neuroimaging. Personal Neurosci 2018;1:2–13.
- Fleming SM, Lau HC. How to measure metacognition. Front Hum Neurosci 2014;8:443.
- Guy W. ECDEU Assessment Manual for Psychopharmacology. Rockville, MD: US Department of Health, Education, and Welfare, Public Health Service; 1976.
- Nielsen G, Ricciardi L, Meppelink AM, Holt K, Teodoro T, Edwards M. A simplified version of the psychogenic movement disorders rating scale: the simplified functional movement disorders rating scale (S-FMDRS). Mov Disord Clin Pr 2017;4(5): 710–716.
- Donnell AJ, Pliskin N, Holdnack J, Axelrod B, Randolph C. Rapidly-administered short forms of the Wechsler adult intelligence scale—3rd edition. Arch Clin Neuropsychol 2007;22(8):917–924.
- Leek MR. Adaptive procedures in psychophysical research. Percept Psychophys 2001;63(8):1279–1292.
- Charles L, Chardin C, Haggard P. Evidence for metacognitive bias in perception of voluntary action. Cognition 2020;194:104041.
- Maniscalco B, Lau H. A signal detection theoretic approach for estimating metacognitive sensitivity from confidence ratings. Conscious Cogn 2012;21(1):422–430.

- 21. R Core Team. R: A Language and Environment for Statistical Computing. Published online 2022. Vienna, Austria: R Foundation for Statistical Computing. https://www.R-project.org
- Algina J, Keselman HJ, Penfield RD. An alternative to Cohen's standardized mean difference effect size: a robust parameter and confidence interval in the two independent groups case. Psychol Methods 2005;10(3):317–328.
- Morey RD, Rouder JN. BayesFactor: Computation of Bayes Factors for Common Designs. Published online 2018. https://CRAN.Rproject.org/package=BayesFactor
- 24. van Doorn J, Ly A, Marsman M, Wagenmakers EJ. Bayesian rankbased hypothesis testing for the rank sum test, the signed rank test, and Spearman's ρ. J Appl Stat 2020;47(16):2984–3006.
- Rahnev D, Fleming SM. How experimental procedures influence estimates of metacognitive ability. Neurosci Conscious 2019; 2019(1):niz009.
- Tinazzi M, Marotta A, Zenorini M, Riello M, Antonini A, Fiorio M. Movement perception of the tonic vibration reflex is abnormal in functional limb weakness. Parkinsonism Relat Disord 2021:87:1–6.
- 27. Marotta A, Bombieri F, Zampini M, et al. The moving rubber hand illusion reveals that explicit sense of Agency for Tapping Movements is Preserved in functional movement disorders. Front Hum Neurosci 2017;11:1–15. Accessed November 2, 2022. https://www.frontiersin.org/articles/10.3389/fnhum.2017.00291.
- Constant M, Salomon R, Filevich E. Judgments of agency are affected by sensory noise without recruiting metacognitive processing. Elife 2022;11:e72356.
- Rouy M, Saliou P, Nalborczyk L, Pereira M, Roux P, Faivre N. Systematic review and meta-analysis of metacognitive abilities in individuals with schizophrenia spectrum disorders. Neurosci Biobehav Rev 2021;126:329–337.
- Edwards MJ, Bhatia KP. Functional (psychogenic) movement disorders: merging mind and brain. Lancet Neurol 2012;11(3): 250–260.
- Williams DT, Ford B, Fahn S. Phenomenology and psychopathology related to psychogenic movement disorders. Adv Neurol 1995;65: 231–257.
- 32. Barnett C, Davis R, Mitchell C, Tyson S. The vicious cycle of functional neurological disorders: a synthesis of healthcare professionals' views on working with patients with functional neurological disorder. Disabil Rehabil 2022;44(10):1802–1811.
- Nielsen G, Buszewicz M, Edwards MJ, Stevenson F. A qualitative study of the experiences and perceptions of patients with functional motor disorder. Disabil Rehabil 2020;42(14):2043–2048.
- Wells A. Metacognitive Therapy for Anxiety and Depression. NY City: Guilford Press; 2011.
- Weissbach A. Metacognitive Therapy and Neuro-Physiotherapy as a Treatment for Functional Movement Disorders - a Randomized, Observer-Blinded Feasibility Trial. clinicaltrials.gov; 2022, Bethesda, MD: US National Library of Medicine. Accessed September 14, 2022. https://clinicaltrials.gov/ct2/show/NCT05323344

# **Supporting Data**

Additional Supporting Information may be found in the online version of this article at the publisher's web-site.

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