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Research paper

Suggestibility as a valuable criterion for laboratory-supported definite functional movement disorders

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ABSTRACT

Objective: To evaluate the application of suggestibility in electrophysiologic studies as a tool to increase the diagnostic certainty of "laboratory-supported definite" FMD.
Methods: We retrospectively reviewed the electrophysiologic studies performed in our center on patients with FMD. Recordings where suggestibility was included in the test battery were then selected.
Results: We present three cases with equivocal clinical features, but with findings on electrophysiologic studies that were consistent with "laboratory-supported definite" FMD.
Conclusion: When combined with other tests, demonstration of suggestibility in electrophysiologic studies may increase the accuracy in differentiating functional from organic movement disorders.
Significance: This case series is an essential first step in evaluating the applicability of suggestibility as an electrophysiologic criterion to aid in the diagnosis of FMD. Application in a larger cohort, incorporation in a test battery, and validation studies, including quantitative evaluation of suggestibility, are required to assess the reliability and the added value of this test.

1. Introduction

Functional movement disorders (FMD) are common and disabling causes of abnormal movements that cannot be explained by a classic neurological disorder (Galli et al., 2020). There is a broad range of phenomenology in FMD, including tremor (most common), myoclonus, tics, dystonia, parkinsonism, chorea, and hemiballism (Hallett, 2016; Galli et al., 2020). FMD are diagnosed clinically based on positive features on history and physical examination (Thenganatt and Jankovic, 2019). Abrupt onset, a preceding injury, fluctuations or remissions, presence of somatizations, failed therapeutic trials, history of childhood trauma, underlying psychiatric comorbidities, ongoing litigation, and secondary gain are suggestive of FMD (Hallett, 2016). Physical examination findings include variability in frequency and distribution, distractibility with cognitive and motor tasks, entrainability, and suggestibility (Hallett, 2016). Electrophysiologic studies (ES), including surface electromyography (sEMG) and EEG backaveraging, are occasionally needed to support the diagnosis, especially for functional tremor and myoclonus (Chen and Chen, 2020). Thus, ES has been included in the most recent operational criteria for FMD, wherein cases with evidence from ES supportive of a diagnosis of FMD are considered "laboratory-supported definite" (Gupta and Lang, 2009). Despite its known utility, ES is not performed in all cases of FMD; the clinical features (e.g., phenomenology and physical examination findings) and physician's experience and confidence in the clinical diagnosis of FMD determine the need for ES while the availability and cost of ES are major limitations. Further, in selected cases, even when the diagnosis is clinically apparent, ES may be obtained in order to provide additional laboratory support to be used in explaining and supporting the diagnosis when presenting this to the patient and their family. In our center, electrophysiologic evaluation was obtained in about 15% of FMD patients, and about 50% of ES (from 2005 to 2019) were performed for the evaluation of FMD.

Although there have been many ES in FMD, testing centers have different study protocols. To date, a standardized battery of tests has only been proposed for functional tremor (Table 1). We retro-

Abbreviations: EEG, electroencephalography; EMG, electromyography; ES, electrophysiologic studies; FMD, functional movement disorders; sEMG, surface electromyography.

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Table 1
Summary of proposed criteria and/or electrophysiologic findings in common functional movement disorders.

Phenomenology	Test Battery/Electrophysiologic findings suggestive of FMD
Tremor[a]	A total score of ≥ 3 points is suggestive of functional tremor in the proposed test battery, which includes: 1. Incorrect tapping performance at 1, 3, and 5 Hz (3 points) 2. Entrainment, suppression, or pathologic frequency shift during tapping at 1, 3, 5 Hz (3 points) 3. 50% reduction in tremor amplitude with contralateral ballistic movements (1 point) 4. Tonic coactivation 300 msec before onset of tremor burst (1 point) 5. Significant coherence in bilateral tremors (1 point) 6. > 130% increased tremor amplitude with weight loading (1 point)
Myoclonus or jerks[b]	1. well organized triphasic pattern of activation of agonist and antagonist muscles 2. long burst duration (>70 msec) 3. variable and long latency (>100 msec) of onset of jerks if with stimulus sensitivity 4. habituation with repeated stimulation (for stimulus-sensitive jerks) 5. variable order of muscle recruitment 6. absence of short (20–40 msec) latency cortical discharge with EEG back averaging 7. presence of Bereitschaftspotential (Bereitschaftspotential has not been documented in organic myoclonus)
Propriospinal myoclonus or jerks[c]	1. absence of a typical rostral and caudal recruitment pattern 2. isolated muscle activity in a single muscle (e.g. rectus abdominis muscle) 3. EMG burst duration longer than 1,000 msec 4. less consistent bursts, i.e., variable first muscle where EMG bursts are noted initially 5. presence of facial movements (particularly eye blinks) or vocalization together with the axial jerks 6. rapid conduction velocities (>16 m/sec) 7. presence of Bereitschaftspotential (Bereitschaftspotential has not been documented in organic myoclonus) 8. presence of event-related desynchronization (ERD) of the beta (13–30 Hz) or mu rhythms (8–12 Hz) from the sensorimotor area (C3/C4)
Tics or movements resembling tics[d]	1. inconsistent pattern of muscle activation 2. presence of variability, entrainment and distractibility of the motor bursts 3. presence of late Bereitschaftspotential (although short duration Bereitschaftspotential can be seen in a proportion of patients with organic tics)

[a] Adapted from Schwingenschuh et al., 2011, 2016; [b] Adapted from Brown and Thompson, 2001; Pal, 2011; Chen and Chen, 2020; [c] Adapted from Kang and Sohn, 2006; van der Salm et al., 2010, 2014; Erro et al., 2013; Chen and Chen, 2020; [d] Adapted from Vial et al., 2019.

spectively reviewed the ES performed in our center on patients with FMD. Recordings where suggestibility was included in the test battery were then selected. Herein, we describe three cases of FMD, where suggestibility on ES increased the diagnostic certainty. Suggestibility was documented on sEMG recording, as demonstrated by an alteration of the movements with the suggestion to the patient that the application of pressure (e.g., using a vibrating turning fork or deep palpation) could either induce, worsen, improve or resolve the movements. Table 2 summarizes the clinical features and the findings on ES.

2. Case series

2.1. Case 1: Functional tremor

A 56-year-old woman complained of right leg tremor three days after a motor vehicle accident. She also reported severe neck pain and generalized weakness and was bedridden for almost three months. The weakness gradually improved, but the right leg tremor persisted, along with back pain, numbness, and paresthesia. There was no autonomic dysfunction, and repeated diagnostic workup was unremarkable. The tremor was present at rest and worsened with maintaining posture (e.g., right leg extended and held off the floor) and movement. After undergoing intensive physiotherapy for two years, she was able to walk with assistive devices, but only for short distances. On examination, she had continuous, semi-rhythmic right leg tremor, with variable frequency and amplitude (**Supplementary Video 1**). The tremor mainly involved the distal right lower extremity, and at times the proximal muscle groups, especially when the leg was raised off the examination bed. There was also a co-contraction sign, and she had to position her heel off the floor to maintain clonus-like rhythmicity. Distractibility and entrainability were difficult to elicit. Although the history and physical examination were suggestive of a functional tremor, ES was requested to support the diagnosis. On sEMG, the tremor was not distractible with cognitive tasks and contralateral ballistic movements. She performed correctly contralateral foot-tapping at 1, 2, and 2.5 Hz, but the tremor amplitude was only reduced at the latter two frequencies and with the right leg raised off the ground. Contralateral ballistic movements did not alter the tremor frequency and amplitude. The only ES findings that supported a diagnosis of FMD were the contralateral foot tapping but with the right leg extended, and the increased tremor amplitude with weight loading. Suggestibility with deep palpation and application of a vibrating tuning fork were then added to the battery of tests, which increased tremor amplitude (Fig. 1**A, 1B**).

2.2. Case 2: Functional movement disorder resembling tics

A 36-year-old woman presented with a two-year history of intermittent, slow, writhing movements of her neck and shoulders, worsening as the day progresses, and occasionally spreading to other body parts. She denied having childhood tics but claimed that she had an urge and a sensation of relief and that she could actively suppress the movements. She also had occasional clicking sound in her throat. On examination at her clinic visits, she had intermittent vocalizations, but no abnormal movements were observed. Homemade videos showed very slow writhing movements of her trunk and shoulders. The character of the movements and the presence of vocalizations were suggestive of tics. FMD was also considered given the age at onset, prolonged duration and variable distribution. During the ES, she had alternating shoulder elevation, followed by slow writhing movements of her neck (**Supplementary Video 2**). The movements were noticeably more frequent on physical examination, and were rare in occurrence otherwise. There were no audible vocalizations or palatal tremor. Cognitive and motor tasks of the upper limbs abated the movements. Entrainment with finger tapping was not observed, but

Table 2
Clinical features and findings on electrophysiologic testing of three patients with functional movement disorders (FMD).

Case No.	1	2	3
Clinical features			
Sex	Female	Female	Male
AAO (years)	52	34	55
Acute onset	Yes	Yes	Yes
Psychiatric comorbidity	Depression	Schizoaffective disorder	None
Other somatizations	Pain in the neck and lower back; numbness; paresthesia	Pain in both knees	None
Phenomenology	Rest, postural and action tremor	Slow, writhing movements with urge, relief and suppressibility (tics vs. FMD)	Jerky movements
Body distribution	Right leg	Neck, shoulders	Upper extremities, shoulder
Variability	Present	Present	Absent
Findings on electrophysiologic testing supportive of FMD			
Distractibility with cognitive tasks	Absent	Present	Present
Distractibility with motor tasks	Absent; i.e., no reduction in tremor amplitude with ballistic movements	Present	Present
Entrainment	Present, but only with higher tapping frequencies and when coupled with a more complex task	Absent	Present at higher frequencies
Loading test	Present; i.e., weight loading increased the tremor amplitude	Not performed	Not applicable
Coactivation	Could not be assessed in the sEMG recordings; tremor was continuous	Not applicable	Not applicable
Suggestibility	Present; increased tremor amplitude with deep palpation and application of a vibrating tuning fork	Present; movements were induced with deep palpation and application of a vibrating tuning fork	Present: Cessation of the jerky movements with application of a vibrating tuning fork
Others	–	(+) Bereitschaftspotential	(+) Bereitschaftspotential

AAO= age at onset; FMD = functional movement disorders; sEMG = surface electromyography.

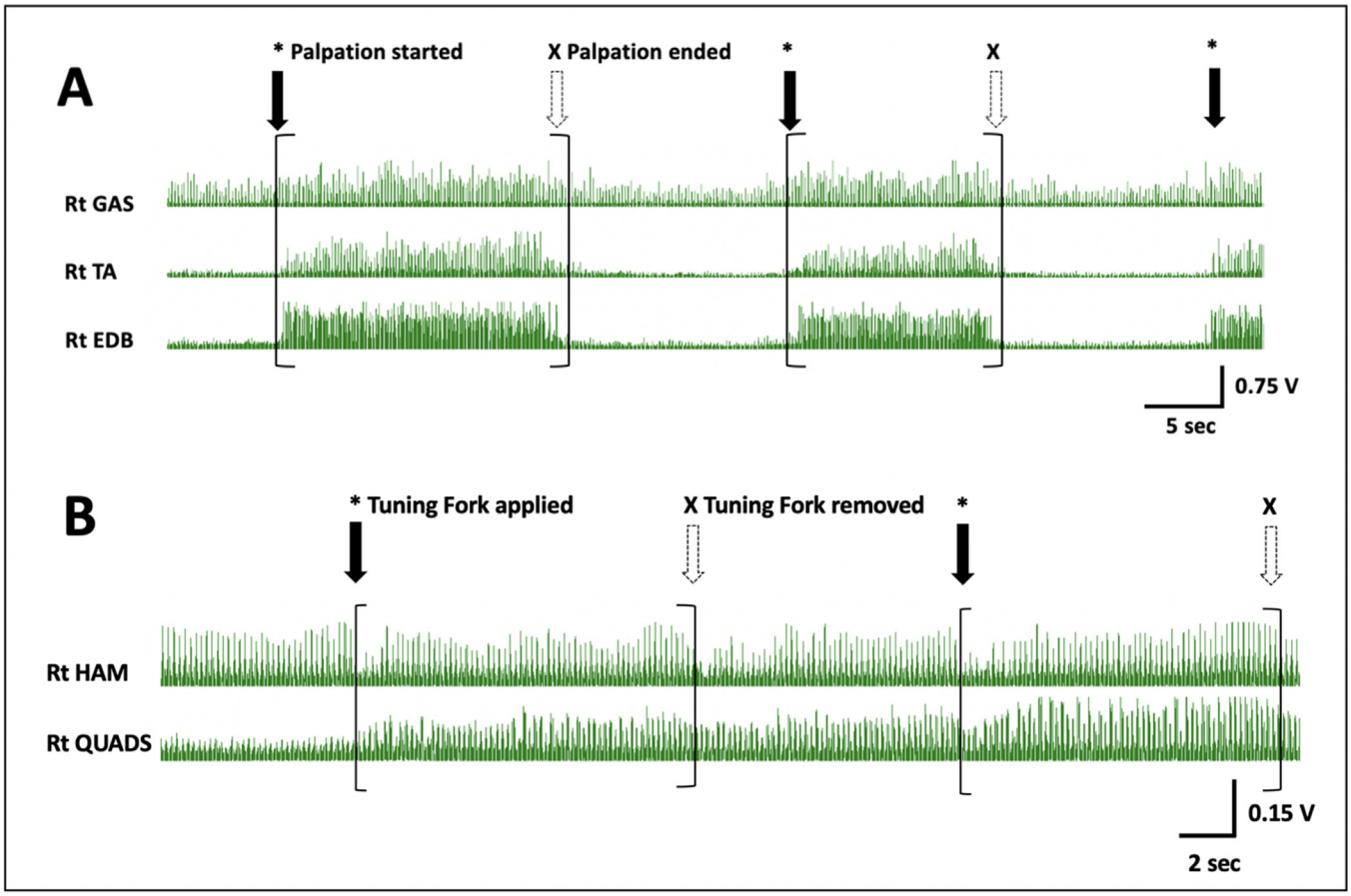

Fig. 1. Case 1. Demonstration of suggestibility in functional tremor. A. Deep palpation on the right ankle markedly increased the tremor amplitude, which was then reduced back to baseline upon cessation of deep palpation. B. Application of a vibrating tuning fork on the right knee increased the tremor amplitude continuously throughout the recording, even with removal of the instrument in between trials. EDB = extensor digitorum brevis; GAS = gastrocnemius; HAM = hamstring muscles; Lt = left; QUADS = quadriceps femoris; Rt = right; TA = tibialis anterior.

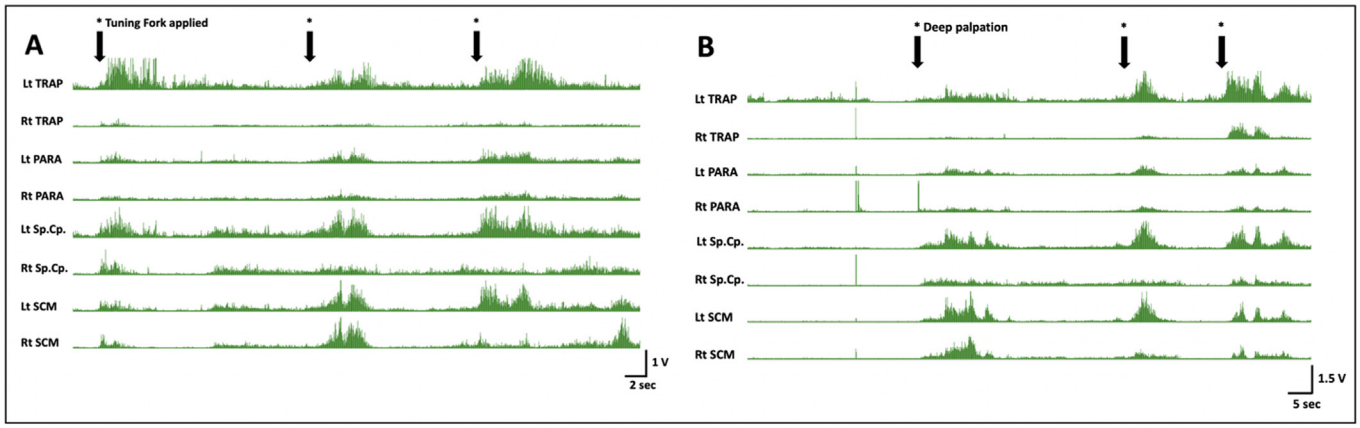

Fig. 2. Case 2. Demonstration of suggestibility in functional movement disorder resembling tics. Application of a vibrating tuning fork (A) and deep palpation (B) on the left shoulder induced the slow, writhing movements. Lt = left; PARA = paraspinal muscles; Rt = right; SCM = sternocleidomastoid; Sp.Cp. = splenius capitis; TRAP = trapezius.

the movements became less frequent. Application of a vibrating tuning fork and deep palpation on the left shoulder, but not on the forehead or right shoulder, induced pain and the abnormal movements. The EMG bursts had variable duration (average of 10–15 s), with a well-organized pattern of activation of antagonistic muscle pairs, occurring at a frequency of 2–3 per minute. The EMG activities were distractible with cognitive and motoric tasks. The movements were also induced by tuning fork vibration and deep palpation on the left shoulder, but not on the forehead or right shoulder (Fig. 2**A, 2B**). EEG back averaging from the left trapezius muscle revealed a pre-movement potential (Bereitschaftspotential).

2.3. Case 3: Functional jerks

A 57-year-old right-handed electrician complained of bilateral hand action tremors four months after a motor vehicle accident, complicated by bifrontal subdural hematoma. During the first consultation visit, the patient had very mild rest tremor affecting the right thumb. He also had mild postural and action tremors bilaterally, reminiscent of a cerebellar outflow tremor (Supplementary Video 3, Segment 1). Cup pouring test, spiral drawings, writing, and gait examination were unremarkable. At his next visit, there were rare very mild upper limb tremors, but he had brief, intermittent jerky movements affecting both arms synchronously, sometimes combined with the trunk and shoulders (**Supplementary Video 3, Segment 2**). Gait examination was normal. FMD was considered given the history, variability, together with ongoing assessment by his insurance company. ES showed evidence for variable, distractible, and inducible jerky movements of the bilateral upper extremities, lasting 300–500 msec. The frequency of the jerky movements was significantly reduced with the application of a vibrating tuning fork on the glabella and occiput (Fig. 3**A, 3B**). Bereitschaftspotential was also present.

3. Discussion

With the increased recognition of FMD, there has been a growing interest in utilizing ES, especially in unclear cases where classic examination findings of FMD may not be present. ES is most helpful to support the diagnosis of functional tremor and myoclonus (Gupta and Lang, 2009), but these could also be performed for other types of movement (Table 1). A combination of tests is preferred because no single test adequately differentiates functional from organic movements (Schwingenschuh et al., 2011). To date, criteria for laboratory-supported definite FMD have only been proposed and validated for functional tremor (Schwingenschuh et al., 2011, 2016). However, there are instances when characteristics of FMD are difficult to demonstrate in ES as in case 1, where the tremor was "overtrained" and not easily distractible or entrainable.

Demonstration of suggestibility on physical examination has been recommended (Kenney et al., 2007; Gupta and Lang, 2009; Thenganatt and Jankovic, 2014), but its incorporation in ES has not yet been described or evaluated. In a study that objectively assessed the commonly employed physical examination maneuvers in the evaluation of FMD cases, suggestibility with vibrating tuning fork application was predictive of functional tremor ($p = 0.04$, sensitivity of 42%); suggestibility with hyperventilation was less reliable ($p = 0.06$, sensitivity of 50%) (Kenney et al., 2007). Unlike distractibility and entrainability, where there is always an improvement of the movements, suggestibility could either worsen (e.g., cases 1and 2) or resolve (e.g., case 3) the movements. In case 1, suggestibility was not apparent on clinical examination, but was demonstrated on sEMG. Moreover, suggestibility could also be applied in other types of FMD, including functional myoclonus and jerks (e.g., case 3) and functional movement disorder resembling tics (e.g., case 2) (Baizabal-Carvallo and Jankovic, 2014; Baizabal-Carvallo and Fekete, 2015). It is sometimes difficult to separate psychogenic jerks and functional movement disorder resembling tics from organic tics and myoclonus. Although patients with Tourette syndrome could have suggestibility, it is still more common in patients with functional tics and jerks (Baizabal-Carvallo and Jankovic, 2014; Baizabal-Carvallo and Fekete, 2015). In all three cases, suggestibility was demonstrated on ES. Combining it with the history, physical examination and other ES findings increased the diagnostic certainty of FMD. Table 3 presents the suggested protocol for the incorporation of suggestibility in the ES test battery for FMD. At an individual level, we propose at least a 50% decrease or increase in amplitude (for tremor) or frequency (for jerks, tics, and functional movement disorder resembling tics) of the mean amplitude or frequency calculated in the pre-test segment, similar to what was proposed for the effect of ballistic movements (Kumru et al., 2004; Schwingenschuh et al., 2011). We recommend performing two maneuvers, vibrating tuning fork application and deep palpation, as the changes may not be as profound with one maneuver only (e.g., Fig. 1). Indeed, the need for more than one type of electrophysiologic assessment is well established for other functional disorders such as tremor and myoclonus. The quantitative effect of suggestibility has to be comprehensively assessed and compared with a control group, such as patients with essential tremor or Parkinson's disease. Moreover, although some clinicians may con-

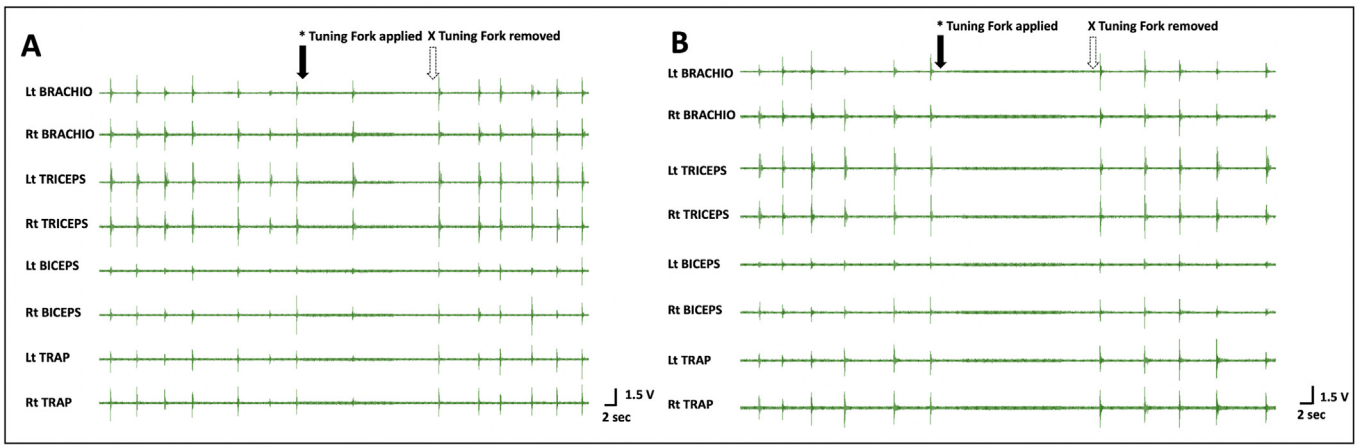

Fig. 3. Case 3. Demonstration of suggestibility in functional jerks. The frequency of the jerky movements was significantly reduced with the application of a vibrating tuning fork on the glabella (A) and occiput (B). BRACHIO = brachioradialis; Lt = left; Rt = right; TRAP = trapezius.

Table 3
Proposed protocol for suggestibility in electrophysiology studies.*

	Description
Channel recordings	• sEMG should be recorded from selected muscles, similar to the muscles chosen for the other procedures in the test battery.
Technical procedure#	1. For functional tremor: obtain the baseline tremor frequency and amplitude for at least 30 s. For functional jerks, tics, and movement disorder resembling tics: obtain the baseline frequency for at least 30–60 s (longer if necessary). 2. Apply a vibrating 128 Hz tuning fork (or an electronic vibration stimulator when available) on at least two body parts and inform the patient that the vibrating stimulus may alter the movement, i.e., worsen, induce, improve or resolve the movements. Repeat this procedure for at least 5–10 trials. 3. Apply deep pressure on at least two body parts and inform the patient that deep pressure may alter the movement, i.e., worsen, induce, improve or resolve the movements. Repeat this procedure for at least 5–10 trials. 4. The examiner explains the procedure to the patient at the end of the test battery (see text for discussion).
Proposed interpretation	• At an individual level, we propose at least a 50% decrease or increase in amplitude (for tremor) or frequency (for jerks, tics, and movement disorder resembling tics) of the mean amplitude or frequency calculated in the pre-test segment in at least 7 out of 10 trials (or in at least 2/3 of the total number of trials). • Note that suggestibility should be included in a test battery and should not be used as the sole basis to classify the patient as having laboratory-supported definite FMD.

*Adapted from the procedures for contralateral ballistic movements response in FMD as outlined in Kumru et al., 2004; Schwingenschuh et al., 2011, 2016. #The authors recommend performing suggestibility at the latter end of the test battery to avoid the perception of deception right at the beginning of the electrophysiologic studies. FMD = functional movement disorders; sEMG = surface electromyography.

sider suggestibility a form of placebo for diagnostic purposes, this is ethically justified when disclosed and explained to the patient afterward to avoid the perception of deception (Rommelfanger, 2013).

Arriving at a correct diagnosis of FMD is crucial for various reasons. Treatment begins when the physician is confident with the diagnosis and is ready to explain it to the patient. Recognizing FMD early in the the patient's course ends the diagnostic workup saga, reduces healthcare costs, and avoids unnecessary, potentially harmful medication trials. Given the lack of a structural lesion, FMD patients have good chances of recovery especially when diagnosed and treated promptly. Indeed, one of the best predictors of a good outcome of FMD is the early diagnosis (Gelauff et al., 2014).

In summary, we presented three cases of FMD with equivocal clinical features, but the ES findings supported the diagnosis of FMD. When combined with other tests, demonstration of suggestibility may increase the accuracy in differentiating functional from organic movement disorders. We recommended incorporating this simple and easily performed test not only on physical examination but also in the battery of tests in ES. The procedure should be explained to the patient afterward for ethical reasons but also in hopes that this could increase the likelihood of acceptance of the diagnosis (Stone and Edwards, 2012). This series is an essential first step in evaluating the applicability of suggestibility as an electrophysiologic criterion to aid in the diagnosis of FMD. Application in a larger cohort, incorporation in a test battery, and validation studies, including quantitative evaluation of suggestibility, are required to assess the reliability and the added value of this test in differentiating functional from organic movement disorders.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary material

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