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Short communication

Tremor retrainment as therapeutic strategy in psychogenic (functional) tremor

A.J. Espay [a,b,*], M.J. Edwards [c], G.D. Oggioni [d], N. Phielipp [e], B. Cox [a], H. Gonzalez-Usigli [a], C. Pecina [a], D.A. Heldman [f], J. Mishra [a], A.E. Lang [e]

[a] James J. and Joan A. Gardner Family Center for Parkinson's Disease and Movement Disorders, Cincinnati, OH, USA
[b] UC Neuroscience Institute, Department of Neurology and Rehabilitation Medicine, University of Cincinnati, Cincinnati, OH, USA
[c] Sobell Department of Motor Neuroscience and Movement Disorders, UCL Institute of Neurology, Queen Square, London, UK
[d] A. Avogadro University and Maggiore della Carità Hospital, Novara, Italy
[e] Movement Disorders Clinic and The Edmond J. Safra Program in Parkinson's Disease, University Health Network, University of Toronto, Toronto, Canada
[f] Biomedical Research Division, Great Lakes NeuroTechnologies, Cleveland, OH, USA

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ABSTRACT

Background: Entrainment, the change or elimination of tremor as patients perform a voluntary rhythmical movement by the unaffected limb, is a key diagnostic hallmark of psychogenic tremor.
Objective: To evaluate the feasibility of using entrainment as a bedside therapeutic strategy ('retrainment') in patients with psychogenic tremor.
Methods: Ten patients with psychogenic tremor (5 women, mean age, 53.6 ± 12.8 years; mean disease duration 4.3 ± 2.7 years) were asked to participate in a pilot proof-of-concept study aimed at "retraining" their tremor frequency. Retrainment was facilitated by tactile and auditory external cueing and real-time visual feedback on a computer screen. The primary outcome measure was the Tremor subscale of the Rating Scale for Psychogenic Movement Disorders.
Results: Tremor improved from 22.2 ± 13.39 to 4.3 ± 5.51 ($p = 0.0019$) at the end of retrainment. The benefits were maintained for at least 1 week and up to 6 months in 6 patients, with relapses occurring in 4 patients between 2 weeks and 6 months. Three subjects achieved tremor freedom.
Conclusions: Tremor retrainment may be an effective short-term treatment strategy in psychogenic tremor. Although blinded evaluations are not feasible, future studies should examine the long-term benefits of tremor retrainment as adjunctive to psychotherapy or specialized physical therapy.

1. Introduction

Psychogenic tremor (PsyT) is the most common psychogenic movement disorder. It may be prolonged and disabling and there are no readily available therapies. PsyT varies in amplitude and distribution. Frequency variability is particularly important for diagnosis, especially if tremor completely stops (suppressibility) or entrains in frequency (entrainment or entrainability) to externally cued rhythmical movements of an unaffected body part [1]. Tremor entrainment and suppressibility, with or without the inability to copy simple cued movements with an unaffected body part, are hallmark clinical features of PsyT. Tremor entrainment not only helps define the clinically definite category of diagnostic certainty for PsyT but may also assist the diagnostic debriefing process at the bedside. We hypothesized that it might also be harnessed as a primary or adjunctive biofeedback treatment ('retrainment'). In this manner, teaching tremor retrainment to patients may help in facilitating self-modulation of the frequency and severity of their tremor, opening the possibility of eventual volitional control over their movements. In this proof-of-concept study, we sought to ascertain the feasibility of tremor retrainment as a biofeedback intervention for patients with chronic PsyT.

2. Patients and methods

2.1. System

A modified version of the Crescendo system (Great Lakes NeuroTechnologies, Cleveland, Ohio) (Supplementary Fig. 1A), originally designed for upper extremity motor rehabilitation, was used to apply an external sensory and visual feedback for rate retrainment. The system included inputs for measuring joint angles

* Corresponding author. University of Cincinnati Academic Health Center, 260 Stetson Street, Suite 2300, PO Box 670525, Cincinnati, OH 45267-0525, USA.
E-mail addresses: alberto.espay@uc.edu, aespay@gmail.com (A.J. Espay).

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(electrogoniometers) and outputs for providing functional electrical stimulation (FES). One electrogoniometer was used to measure wrist flexion–extension and one channel of FES was applied to the wrist extensor muscles at a magnitude sufficient to produce a tactile sensation, but below the motor threshold to prevent elicitation of a muscle contraction. A LabVIEW interface was developed for controlling stimulation parameters, processing tremor kinematics, displaying feedback, and saving data (Supplementary Figs. 1B and 2).

2.2. Participants and assessments

Ten patients with chronic PsyT were studied (5 women, mean age, 53.6 ± 12.8 years; mean disease duration 4.3 ± 2.7 years) as necessary from 1 to 3 consecutive days (Table 1). All patients were recruited at the University of Cincinnati movement disorders center. The diagnosis of PsyT was made by documenting variability of hand tremor frequency and amplitude, as well as by demonstrating entrainability or, in its absence, clear suppressibility during performance of complex tasks in an unaffected limb. Clinical assessments of tremor severity were performed prior to and immediately at the completion of the retrainment session. Longer-term outcome was determined by telephone follow up between 3 and 6 months from completion of retrainment. Baseline entrainability and suppressibility were scored offline by blinded investigators using video from a standardized recording protocol (0–2: 0, none; 1, partial; 2 complete, for each). Tactile stimulation with visual feedback was delivered synchronously by Crescendo at approximately 2/3 of the baseline frequency of the affected limb for approximately 60 min, and then at 1/3 of the baseline frequency for another 60 min. Patients were instructed to make flexion–extension wrist movements of the most affected hand matching the device stimulation for the duration of the feedback. Intrusion of baseline tremor was discouraged during the performance of the externally paced wrist movements. To that end, training of up to 10 min prior to the biofeedback session was conducted to ensure optimal matching of the given rate with minimal intrusion of the baseline tremor. If the tremor was not substantially improved at the end of a single 2-h retrainment session, the patient participated in a second and, if necessary, a third consecutive-day session. All patients were recruited from the University of Cincinnati Movement Disorders Center. The institutional review board approved the study and patients provided informed consent.

2.3. Data analysis

The primary outcome measure was the combined severity, duration factor, and incapacitation scores of the Tremor subscale of the Rating Scale for Psychogenic Movement Disorders (0–12 per each limb and head [maximum for all five regions: 60]) [2]. A paired *t* test was used to compare the scores before and after retrainment.

3. Results

At baseline, suppressibility was more common than entrainability in this cohort (1.5 ± 0.5 vs. 0.6 ± 0.66, $p = 0.009$), but both were present in half the cohort (Video 1). Entrainment was unfeasible in 5 subjects due to complete suppressibility. Nevertheless, tremor improved across subjects with predominant suppressibility (in whom entrainability was precluded) as well as in those whose tremor was both entrainable and suppressible (tremor score changed from 22.2 ± 13.4 to 4.3 ± 5.5; $p = 0.0019$) (Table 1) (Fig. 1) (Video 2). Three subjects achieved tremor freedom, two of whom had complete suppressibility on examination at baseline (i.e., were non entrainable) (Video 3). Two of these subjects, however, had partial or complete entrainment in another limb or the head.

Supplementary data related to this article can be found online at http://dx.doi.org/10.1016/j.parkreldis.2014.02.029.

A 2-h retrainment session was sufficient for four subjects whereas two consecutive 2-h sessions were needed for five subjects. Only one subject (10 years disease duration) required a third day of retrainment to substantially improved (Video 2). The benefits were maintained for at least 1 week and for up to 6 months, at last follow up. By six months, four patients had experienced relapse whereas six remained markedly improved, including one free of tremor.

4. Discussion

Tremor retrainment appears to be an effective treatment strategy for PsyT patients. Currently used exclusively to establish the diagnosis, entrainment or suppressibility, may be exploited for therapeutic purposes.

As recently proposed, sharing diagnostic signs such as entrainment with patients during the debriefing process serves to illustrate how the diagnosis is made and may be a powerful way of persuading them about the nature of their illness and the futility of proceeding with extensive neurological investigations [3]. Although explaining the diagnosis of a psychogenic movement disorder (or, as we prefer, a “functional movement disorder” [4]) to a patient can be a difficult task, such ability may be assisted by the affirmation of a feature that carries with it both diagnostic and therapeutic implications. Therefore, discussing the presence of entrainment/suppressibility (and its potential as a therapeutic tool) would be expected to facilitate acceptance of the diagnosis, arguably the most important factor in determining good outcome [5].

The retrainment intervention examined here adds further strength to the role of rehabilitation protocols for psychogenic/functional movement disorders. A recent experience was reported for an outpatient, one-week intensive rehabilitation protocol based on “motor reprogramming” applied to patients with functional gait disorder [6]. It is possible, however, that physical therapy- and biofeedback-based treatment protocols may only be associated with sustained long-term benefits when paired with cognitive behavioral therapy [7]. The growing evidence supporting the use of this treatment modality in patients with psychogenic nonepileptic seizures [8,9] is expected to be translatable to patients with psychogenic/functional movement disorders [10].

A number of limitations of our pilot study must be highlighted. By design, a study examining the concept of retraining needed to be open-label and could not be controlled (e.g., by a “noisy” external

Table 1
Demographics, baseline tremor scores, and outcome in patients studied.

Subject	Age	Gender	Duration (years)	Training (days/h)	Tremor scores		Baseline S & E		Outcome
					Before	After	S	E	
1	55	F	8	2/4	42	19	1	1	Relapsed after 2 weeks
2	66	F	10	3/6	7	0	2	0	Relapsed after 6 months
3	61	F	4	2/4	12	8	1	1	Relapsed after 3 months
4	36	M	2	2/4	24	2	1	0	Remained tremor-free at 5 months
5	36	F	5	1/2	36	0	1	0	Relapses only with stress at 3 months
6	75	M	3.5	2/4	45	2	2	1	Relapsed after 4 months
7	57	F	5	1/2	18	4	2	0	Remained improved after 4 months
8	41	M	3	1/2	7	0	1	1	Remained tremor-free after 4 months
9	64	M	1.5	2/4	18	6	2	2	Remained tremor-free after 4 months
10	45	M	0.8	1/2	13	2	2	0	Remained improved after 6 months

S = Suppressibility; E = Entrainability.
No additional therapies recommended or received during the follow up period for any of the subjects.