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Short communication

# Head tremor in essential tremor: "Yes–yes", "no–no", or "round and round"?

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ABSTRACT

*Introduction:* Essential tremor (ET) is a common yet frequently misdiagnosed movement disorder. One contributing factor may be the dearth of studies that focus on the nuances of clinical phenomenology. A clinical feature that has received relatively little attention is head tremor. Indeed, there is no consensus regarding the predominant direction of head tremor in ET, and no study has examined the clinical correlates of directionality.

*Methods:* We identified 51 ET cases with head tremor enrolled in a clinical-epidemiological study of ET at Columbia University. Each had a videotaped neurological examination. Videotapes were viewed and coded by a movement disorders neurologist for head tremor direction ("no–no", "yes–yes", or mixed) and continuity (continuous, intermittent, or rare). Direction was correlated with a wide range of clinical features.

*Results:* Fourteen cases (27.5%) had "no–no" tremor, 9 (17.6%) had "yes–yes" tremor, and 28 (54.9%) had a mixed tremor. Mixed and "yes–yes" cases were older ($p = 0.004$) and had a longer tremor duration ($p = 0.018$) than "no–no" cases. Tremor severity (arms) was higher for mixed cases than for "yes–yes" and "no–no" cases ($p = 0.04$). More mixed cases had continuously present tremor while more "no–no" cases had rare head tremor ($p < 0.001$).

*Conclusions:* Head tremor in ET seems to start as an infrequent tremor in one direction (esp. "no–no") and becomes more frequent while acquiring additional directionality and a mixed phenotype as the disease progresses. These findings add to our understanding of the clinical spectrum of ET.



## 1. Introduction

Essential tremor (ET) is one of the most common movement disorders; however, according to some estimates, it is also misdiagnosed as much as 30–50% of the time [1]. One contributing factor is the surprising dearth of studies that focus on the nuances of clinical phenomenology.

Head tremor affects many ET patients, and there is a female preponderance [2]. Even though directionality (i.e., horizontal vs. vertical) was observed in early descriptions of ET head tremor, there are few detailed studies of and no consensus regarding the predominant direction of head tremor in ET. Furthermore, the existing reports addressing head tremor direction must be interpreted within the context of several limitations, including small sample sizes (e.g., n = 5, 16, 23), restriction of analyses to familial cases, or the fact that they were conducted prior to modern diagnostic distinctions between ET and dystonic head tremor [3–7]. The most complete description characterized head tremor in 16 familial ET cases; of these, 12 (75.0%) had "no–no" (i.e., horizontal) tremor, 1 (6.25%) had "yes–yes" (i.e., vertical) tremor, and 3 (18.75%) had mixed tremor (i.e., more than one direction) [7]. In another series of patients with mixed movement disorders (23 ET, 7 cervical dystonia), 13 (56.5%) of 23 ET cases had "no–no" tremor, 4 (17.4%) of 23 ET cases had "yes–yes" tremor, and 6 (26.1%) of 23 ET cases had mixed tremor [5].

In this study, we characterize the direction of head tremor and examine the clinical correlates of directionality in a large series of 51 patients with ET who had head tremor. As the largest study to date to determine head tremor directionality and the first to

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correlate clinical features with tremor direction, we hope to add to our clinical understanding of this very common neurological disease.

## 2. Methods

### 2.1. Description of study

Participants were prospectively enrolled in a clinical-epidemiological study of ET, (2009–2014) [8]. ET cases were identified from a computerized billing database at the Center for Parkinson's Disease and Other Movement Disorders, Neurological Institute, Columbia University Medical Center (CUMC), with a search conducted of all patients seen within the three years prior to enrollment. Each of the enrolled ET cases had received a diagnosis of ET from their treating neurologist at the Institute and lived within 2 h driving distance of CUMC. One of the authors (E.D.L.) reviewed the office records of all selected cases; those with diagnoses or physical signs consistent with other movement disorders were excluded. The CUMC Internal Review Board approved of all study procedures. Written informed consent was obtained upon enrollment. Analysis of data was also approved by the Internal Review Board at Yale Medical School.

### 2.2. Evaluation and diagnostic confirmation

A trained research assistant conducted an in-person evaluation, administering demographic and medical history questionnaires. During the in-person assessment, a videotaped neurological examination was performed. This included one test for postural tremor and five for kinetic tremor (12 tests total) as well as assessments of speech and voice (sustained phonation). A neurologist specializing in movement disorders (E.D.L.) used a reliable and valid clinical rating scale, the Washington Heights-Inwood Genetic Study of ET (WHIGET) tremor rating scale, to rate postural and kinetic tremor during each test: 0 (none), 1 (mild), 2 (moderate), 3 (severe), resulting in a total tremor score (range = 0–36) [9]. Diagnoses of ET were re-confirmed by E.D.L. using the videotaped neurological examination and WHIGET diagnostic criteria (moderate or greater amplitude kinetic tremor [tremor rating $\geq$ 2] during three or more tests or a head tremor, in the absence of Parkinson's disease, dystonia or another cause) [9].

### 2.3. Final sample with head tremor and re-review of videotaped examination

Among the 141 enrollees, we identified 51 (36.2%) with head tremor on videotaped neurological examination. These 51 were selected for additional analyses. The videotaped neurological examination was re-reviewed by a movement disorders neurologist (D.R.) who coded clinical features. At the start of the re-review, a 25% sample of these ET cases was co-reviewed with the senior movement disorders neurologist (E.D.L.) to ensure agreement.

During the videotaped neurological examination, the patient's head was visible during the following segments: 1) sitting at rest with arms in lap, 2) sitting with arms outstretched, 3) pouring water from one cup into another, 4) bringing a spoon to the mouth, 5) finger-to-nose testing, 6) speech and sustained vocalization, 7) rapid alternating movements, 8) walking normally and tandem gait, 9) writing, and 10) reading a passage out loud.

Head tremor was coded as horizontal ("no–no"), vertical ("yes–yes"), or mixed. The mixed category included (1) tremor that was sometimes "yes–yes" and sometimes "no–no" and (2) tremor in an axis other than horizontal or vertical (e.g., diagonal). In the former, a certain percentage of head tremors were predominantly "yes–yes" or predominantly "no–no" (i.e., a direction that was present for more than half the time the tremor was visualized), and this was also noted. Tremor was also categorized according to constancy and was labeled as continuously present (i.e., present whenever the head was visible during the videotaped neurological examination), intermittently present, or rarely present (i.e., present during few segments of the videotape).

**Table 1**
Clinical characteristics of 51 ET cases with head tremor on examination.

| Characteristic | Data |
|---|---|
| Age (years) | 74.5 ± 10.9 |
| Female gender | 35 (68.6) |
| Tremor duration (years)[a] | 38.3 ± 19.3 |
| Reported one or more 1st degree relative with ET | 17 (33.3) |
| Prescribed tremor medications | 35 (68.6) |
| Tremor in voice (examination) | 19 (37.3) |
| On examination, the head tremor is: | |
| Continuous | 20 (39.2) |
| Intermittent | 15 (29.4) |
| Rare | 16 (31.4) |
| Total tremor score (0–36)[b]: | |
| 0–12 | 9 (18.0) |
| 13–24 | 30 (60.0) |
| 25–36 | 11 (22.0) |

All values represent mean ± standard deviation or number (percentage).
[a] Data unavailable for 4 ET cases.
[b] Data unavailable for 1 ET case.

### 2.4. Data analyses

Data were analyzed in SPSS (Version 21.0), comparing groups using Student t tests or chi-square tests. For some analyses (e.g., age category, tremor duration, total tremor score), data were stratified into categories.

## 3. Results

Fifty-one ET cases had head tremor (Table 1). The group included a preponderance of women (68.6%, Table 1). Head tremor was continuous in 39.2% and intermittent or rare in the remainder (Table 1).

In terms of directionality, 14 cases (27.5%) had "no–no" head tremor, 9 (17.6%) had "yes–yes" head tremor, and 28 (54.9%) were of the mixed type. The mixed types were as follows: 6 of 28 were predominantly "no–no", 3 of 28 were predominantly "yes–yes", 10 alternated between "yes–yes" and "no–no" to an equal extent, and 9 involved axes other than horizontal.

We examined the clinical factors that correlated with tremor direction (Table 2). Mixed and "yes–yes" cases tended to be older ($p = 0.004$) and have a longer tremor duration ($p = 0.018$) than cases with "no–no" tremor. More than half the "no–no" cases had a tremor duration of less than 20 years, where only 11.1% of the mixed and none of the "yes–yes" cases had such short tremor duration. We also found an association between total tremor score and directionality ($p = 0.04$); scores tended to be higher for mixed cases than for "yes–yes" and "no–no" cases. The continuity of the tremor was correlated with directionality ($p < 0.001$): more mixed cases had continuously present tremor while more "no–no" cases had rare head tremor, and "yes–yes" cases displayed an intermediate distribution. A marginally higher proportion of the mixed tremor cases were female (82.1%) ($p = 0.058$). The proportion of mixed and "yes–yes" tremor cases who had voice tremor was higher than those with "no–no" tremor, a difference that was marginally significant ($p = 0.07$, Table 2).