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Comprehensive, blinded assessment of balance in orthostatic tremor

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ABSTRACT

Introduction: Orthostatic Tremor (OT) is a movement disorder characterized by a sensation of unsteadiness and tremors in the 13–18 Hz range present upon standing. The pathophysiology of OT is not well understood but there is a relationship between the sensation of instability and leg tremors. Despite the sensation of unsteadiness, OT patients do not fall often and balance in OT has not been formally assessed. We present a prospective blinded study comparing balance assessment in patients with OT versus healthy controls.
Methods: We prospectively enrolled 34 surface Electromyography (EMG)-confirmed primary OT subjects and 21 healthy controls. Participants underwent evaluations of balance by blinded physical therapists (PT) with standardized, validated, commonly used balance scales and tasks.
Results: OT subjects were mostly female (30/34, 88%) and controls were majority males (13/20, 65%). The average age of OT subjects was 68.5 years (range 54–87) and for controls was 69.4 (range 32–86). The average duration of OT symptoms was 18 years.
OT subjects did significantly worse on all the balance scales and on most balance tasks including Berg Balance Scale, Functional Gait Assessment, Dynamic Gait Index, Unipedal Stance Test, Functional Reach Test and pull test. Gait speed and five times sit to stand were normal in OT.
Conclusions: Common validated balance scales are significantly abnormal in primary OT. Despite the objective finding of impaired balance, OT patients do not commonly have falls. The reported sensation of unsteadiness in this patient population seems to be out of proportion to the number of actual falls. Further studies are needed to determine which components of commonly used balance scales are affected by a sensation of unsteadiness and fear of falling.

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1. Introduction

Orthostatic Tremor (OT) is a rare movement disorder characterized by a sensation of unsteadiness and 13–18 Hz leg tremors present upon standing. Both the sensation of unsteadiness and the tremors disappear or improve by sitting, walking or leaning [1]. The 1998 consensus statement on tremors from the Movement Disorders Society described OT as a subjective feeling of unsteadiness during stance and only in severe cases during gait, noting that patients rarely fall. Patients typically have no problems when sitting and lying down. Clinical findings are sparse and mostly limited to a visible or auscultated and occasionally, only palpable fine amplitude rippling of the legs. The diagnosis can be confirmed by surface EMG recordings with a fast 13–18 Hz tremor [2]. These are the criteria still used by most movement disorder experts today.

Despite a sensation of unsteadiness as one of the defining features of the disease, falls are relatively uncommon in primary OT

patients. Many OT patients report they do not fall more often because they are very careful, though there have been some reports of falls in OT patients in the literature [3,4]. One large retrospective study reported at least occasional falls in 24% of reviewed cases [5]. It remains unclear if OT patients have a true impairment of balance or only a sensation of instability. Formal balance assessment has not been studied in this patient population. We evaluated balance in primary OT with available standardized scales and tasks in a formal, comprehensive blinded fashion.

2. Methods

In this single blinded prospective case-control study we enrolled subjects with surface EMG-confirmed OT and healthy controls. This study was approved by the University of Nebraska Medical Center (UNMC) Institutional Review Board (IRB) and was part of the large ongoing UNMC OT study project. Informed consent was obtained over the phone due to the subjects coming from large distances and re-consent was obtained at the start of the study visit as approved by the IRB. Record of surface EMG with leg tremor frequency on standing in the 13—18 Hz range was accepted as proof of OT diagnosis and inclusion into the study. Participants with known vestibular or balance problems other than OT were excluded. This exclusion was based on self-reported prior diagnosis that could affect gait or balance. Cases of secondary OT including patients with Parkinsonism were excluded. Evaluations were video recorded.

Subjects underwent standardized balance evaluations by blinded physical therapists who were experienced at performing these scales. These scales have been thoroughly validated and have been shown to predict risk of falling in multiple neurological illnesses [6—13]. The scales were performed in a uniform fashion including the same sequence, procedure of test and patient instructions. The therapists were given folders that only had the participant's number, and the scale forms to be filled out. They did not have access to any other research data on the participants. Physical therapists were not told the diagnostic status of the subject. Subjects were instructed not to discuss their diagnosis with the evaluators and to avoid and language during testing that may tip off their level of discomfort with the testing.

The assessments included the Berg Balance Scale (BBS), Dynamic gait index (DGI), Functional gait assessment (FGA), Unipedal stance test (UST), functional reach test (FRT), pull test (PuT), gait speed (GS) and five times sit to stand (FTSS) test. The Berg Balance Scale is a 14-item test that uses ordinal scoring from 0 to 4 for each item and is designed to measure static and dynamic standing balance [6]. Dynamic Gait Index consists of 8 tasks with similar scoring as Berg Balance Scale, but testing both stance balance and gait [7]. The Functional Gait Assessment is an extension of Dynamic Gait Index, with an additional 3 items plus 7 of 8 previous items for a 10-item scale with enhanced assessment of gait [8]. Unipedal Stance Test, Functional Reach Test and Five Times Sit to Stand are balance tasks that were performed and scored as per known standard protocols [9—13]. The Pull Test is a test of postural stability and was performed as follows. Subjects were instructed that the investigator would pull them backwards and they should try not to fall, taking a step back if necessary. Taking more than 3 steps back, or falling unless caught, was considered an abnormal result. Gait speed was measured both in habitual and brisk walking over 6 m. We used existing normative data for cut-off values in the elderly population to compare the balance performance in our study on these tests [9—14]. These scales have Standard Error of measurements and Minimal Detectable Change data published. However, the Minimally Clinically important difference has not been established.

2.1. Statistical analysis

We compared the functional performances between cases and controls by quantitative statistical methods, based on the original scores or results of tests. The data is not normally distributed, thus we conducted Mann-Whitney tests for most measures, except Gait speed. We conducted independent *t*-test for Gait speed. Significance level was set at 0.05.

3. Results

Thirty-four surface EMG-confirmed primary orthostatic tremor subjects and 21 healthy controls were included in the study. The majority (88.2%, n = 30) of the OT subjects were female. The controls were 35% female (n = 7). The average age in the OT group as of evaluation date was 68.5 years (range 54—87) and 69.4 years in the control group (range 32—86). The mean age of onset of OT symptoms was 50.8 years (range 24—71). The mean age at diagnosis of OT was 61.1 (range 49—82). The average peak tremor frequency in the OT group as documented on surface EMG was 15.6 Hz (range of 12 Hz—18 Hz).

Nine of the 34 OT subjects reported a history of at least one fall and 5 subjects reported more than one fall attributed to OT symptoms. Thirty-two of the OT subjects reported a sensation of unsteadiness when standing in place and 15 of these also reported a sensation of unsteadiness while walking. Twenty-seven OT subjects reported difficulty walking in a straight line slowly and only 4 also reported difficulty walking fast. Thirty-one reported difficulty walking in crowded places and 20 reported difficulty going down stairs. The majority of the OT subjects, 17 of 34, reported leg tremor onset just seconds after standing. Nine of 34 reported tremors in legs immediately on standing and 6 of 34 min after standing. Two subjects did not subjectively feel tremors. Twenty of the OT subjects reported sensation of arm tremors in addition to leg tremors.

We found slight increase in upper extremity tone in 16 of the OT subjects and mild bradykinesia in 19 subjects. There was postural tremor in 13 OT subjects and reduced arm swing noted in 8 of the 34. None of the participants included in the study met full criteria for diagnosis of Parkinson's disease.

OT subjects performed significantly worse compared to healthy controls on all balance scales and most balance tasks while having normal gait speeds and FTSS test (see Table 1).

4. Discussion

A strong sense of instability and associated fear of falling is commonly reported in OT, affecting quality of life and social functioning [15—17]. Actual falls are comparatively uncommon in OT and this has raised the question if these patients have a true impairment of balance or only a sensation of unsteadiness and associated fear of falling as a disease symptom [18,19].

We found OT subjects to have significant abnormalities on all the balance scales and most of the balance tasks tested. However, the gait speed was normal when compared to healthy controls and to standardized normative data. Overall tasks requiring standing in place were more difficult for the OT subjects compared to tasks involving walking. One anomaly, was the FTSS test which was normal in OT subjects and is a test of balance during stance. This normal result could be related to the short duration of stance in between sitting, as many OT patients have some latency to symptom onset upon standing.

All three balance scales (BBS, DGI and FGA) showed worse performance by OT subjects compared to healthy controls. There were significantly more OT subjects in the high risk category on BBS and DGI scales (9 and 8 respectively). The largest difference from

Table 1
Performance on balance scales and tasks.

Test	OT subjects mean, (SD)	Controls mean, (SD)	Normal reference value for mean age of our participants	P value
Berg balance scale	44.3, (10.57)	53.5, (5.56)	>53	<0.001
Dynamic gait index	20.1, (3.8)	22.1, (3.63)	<19 predictive of falls in the elderly and 19 to 22 mild risk of falls	0.005
Functional gait assessment	22.3, (5.42)	26.3, (5.35)	>26	0.001
Functional reach test (cm)	21.1, (9.16)	28.3, (4.71)	>25 cm	<0.001
Unipedal stance test (seconds)	3.8, (3.83)	14.7, (10.42)	14-22 s	<0.001
Pull test	67.9% (n = 23) abnormal	28.6% (n = 6) abnormal	Normal defined as less than 3 steps back when pulled	<0.001
5 times sit to stand (seconds)	13.2, (4.52)	11.2, (2.85)	<15 s	0.128
Habitual gait speed over 6 m (m/s)	1.1, (0.19)	1.1, (0.2)	0.6—1.5 m/s	0.567
Fast gait Speed over 6 m (m/s)	1.4, (0.29)	1.5, (0.29)	0.8—2.1 m/s	0.434

Cm = centimeters, m = meters, m/s = meters/second.

controls was seen on BBS (9.2 points). Standardized data suggests BBS scores for patients without balance impairment in their sixties and seventies should be above 54 and above 52 respectively [20]. On DGI, a score less than 19 out of a possible 24 is predictive of falls in the elderly. Those scoring 23 or 24 are considered safe ambulators and scores 19 to 22 correlate to mild risk of falls. Eight OT subjects were in the high risk category and only 7 were safe ambulators by this scale. Normal functioning on FGA is determined by age with a perfect score being set at 30 and normal score for a person in their 60s and 70s is estimated at 27.1 and 24.9 respectively. The control group fell within this range as would be expected for someone in their late 60s but the OT subjects had a lower than expected score which correlates to higher risk for falls.

One of the challenges of this study was not having balance scales for stance and gait separately. The current balance scales may over read the standing balance problems and under represent instability of gait. Another issue is that an individual with normal balance but a sensation of instability and associated fear of falling down may perform poorly on balance scales due to poor effort, creating false positive results. Tasks like FRT for example, that require maximum comfortable reach without falling, may be severely limited by the cautiousness driven by fear of a potential fall.

Additional points of weakness in the study include the large proportion of female cases and male controls. Other studies of OT have also found a female preponderance of close to 70% [2,3]. Our number of 88% females may be even higher due to the tendency for females to participate in prospective research studies. The controls were majority male due to using spousal controls. The normative data for the balance scales and tasks is based on age and we did have good age matching between cases and controls. Measures were in place to avoid unblinding of the examiners. The therapists were given folders that only had the participant's number, and the scale forms to be filled out. They did not have access to any other research data on the participants. Physical therapists were not told the diagnostic status of the subject. Subjects were instructed not to discuss their diagnosis with the evaluators and to avoid any language during testing that may tip off their diagnosis or their level of discomfort with the testing.

In conclusion, it is clear from our study that primary OT patients perform poorly on standard balance assessments compared to healthy controls. However, we argue that it is not clear if these abnormal results are due to true imbalance, or due only to a sensation of imbalance and associated fear of falling. The widely reported sensation of instability in primary OT patients seems out of proportion to the number of reported falls. Assessments that could differentiate the sensation of instability from true impaired balance would be extremely useful, and are needed to evaluate this complex relationship in patients with Orthostatic Tremor. We are currently performing a follow up study evaluating balance scales and tasks to determine which components may be most compromised by a fear of falling.

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