A dissociation between subjective and objective unsteadiness in primary orthostatic tremor

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Summary
Patients with primary orthostatic tremor (OT) experience a disabling sense of unsteadiness but rarely fall. In order to study the relationship between the development of subjective unsteadiness, objective unsteadiness and tremor, we recorded standing under four conditions (eyes open or closed, feet together or apart) in six patients with OT. Subjective unsteadiness was indicated by the patients on a four-point scale using a hand-held slider. Objective unsteadiness was assessed by measuring the path lengths of the centre of foot pressure and body motion at the level of the cervical spine. Tremor was measured by surface electromyography from leg and paraspinal muscles. OT patients were objectively more unsteady than controls. Objective unsteadiness also increased disproportionately in patients when standing with eyes closed. These findings suggest that balance control in OT is abnormal and shows increased visual dependence. Subjective unsteadiness increased from mild to severe over seconds to minutes. The increase was faster when standing with eyes closed or feet together. However, although escalating subjective unsteadiness was paralleled by an increase in leg tremor, there were no comparable changes in either paraspinal tremor or objective unsteadiness during the course of a stand. We conclude that there is a dissociation between subjective and objective unsteadiness. This implies that subjective unsteadiness does not arise simply from an awareness of increased body sway. We postulate that the sensation of unsteadiness arises from a tremulous proprioceptive afferent activity from the legs. This disturbance gives rise to increased co-contraction drive to the leg muscles in order to stiffen the joints and increase stability. Since muscle activity remains tremor-disproportionately in patients when standing with eyes locked, the tremulous proprioceptive feedback is increased, which then further increases the sensation of unsteadiness, and so on in a vicious circle of escalating activity.

Keywords: primary orthostatic tremor; balance; posture; proprioception; co-contraction

Abbreviations: ANOVA = analysis of variance; CoP = centre of foot pressure; OT = primary orthostatic tremor

Introduction
Primary orthostatic tremor (OT) is a distinct clinical syndrome characterized by lower limb tremor on standing accompanied by a profound sense of unsteadiness (Pazzaglia et al., 1970; Heilman, 1984; Thompson et al., 1986). EMG recording from the lower limbs reveals high-frequency (13–18 Hz) bursting that is virtually pathognomonic of this condition (Thompson et al., 1986; Veilleux et al., 1987; Britton et al., 1992; McManis and Sharbrough, 1993). The patient’s sense of unsteadiness can be intense even if the person has never fallen, and this symptom may overshadow that of the leg tremor. The sensation increases in intensity when the patient is forced to remain standing (Heilman, 1984), and invariably patients are compelled to seek relief by sitting or walking (Heilman, 1984; Walker et al., 1990; Britton et al., 1992). This urge is so strong that most patients eventually avoid situations where they may be forced to stand still, such as shopping or queuing; alternatively they may resort to carrying aids, such as shooting sticks, which can be easily converted to a seat (Britton and Thompson, 1995). Despite the sense of unsteadiness being the most disabling symptom of orthostatic tremor, its pathogenesis remains unknown (Thompson, 1999).

The apparent strong link between OT and standing suggests that its pathophysiology might be specifically related to the control of balance. However, there is increasing evidence that

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the tremor is related more to isometric muscle contraction than to balance. Thus it can be variably present in the muscles of the face, arms and trunk as well as the legs (Walker et al., 1990; Britton et al., 1992; Boroojerdi et al., 1999; Koster et al., 1999).

It is not evident, therefore, why the subjective disturbance is so specific to the act of standing still. In addition, it seems paradoxical that patients with OT rarely fall despite their severe sense of unsteadiness. In the present study, we charted the time course of OT patients’ sensation of unsteadiness (subjective unsteadiness) while at the same time measuring their standing performance (objective unsteadiness) and tremor. With this approach we have attempted to answer two questions. (i) Do OT patients have a frank disruption of the normal balance mechanisms controlling upright stance? (ii) What is the relationship between objective unsteadiness, subjective unsteadiness and the development of tremor?

Methods

Subjects

We studied six patients with OT (mean age 63.5 years, range 51–81 years, five female) and six control subjects (mean age 64.5 years, range 50–78 years, five female). Control subjects had no history of neurological disease and no family history of tremor or other movement disorder. Characteristics of the OT patients are shown in Table 1. All OT patients experienced symptoms typical of OT and had 14–18 Hz tremor in the legs during standing confirmed by surface EMG recordings. It is noteworthy that five of the patients had never fallen as a result of OT and the remaining patient had not fallen for some years. Patients were asked to withhold medications used to treat OT overnight prior to the study. The patients and control subjects gave informed consent according to the Declaration of Helsinki and the study was approved by the combined ethics committee of the National Hospital for Neurology and Neurosurgery and the Institute of Neurology, London.

Experimental procedure

Subjects stood barefoot on a force platform (type 9281B; Kistler Instrumente AG, Winterthur, Switzerland) facing and 1 m away from a wall that was draped with a black curtain. In order to investigate the effect of varying degrees of balance difficulty, subjects were requested to stand with two stance widths (16 or 0 cm distance between the medial borders of the feet) and two visual conditions (eyes open or closed). Thus, we recorded four separate standing conditions in the following order: (i) feet apart, eyes open; (ii) feet apart, eyes closed; (iii) feet together, eyes open; (iv) feet together, eyes closed. This cycle was repeated three times for each subject.

At the beginning of every trial, the subject sat on a 50-cm high stool (without backrest or arm support) with the feet placed together or apart on the ground according to the experimental condition. In response to an auditory cue the subject stood up and attempted to stand as still as possible (with eyes closed when required) while indicating their subjective state of unsteadiness with a hand-held indicator (see below). This continued until either they were no longer able to stand or 90 s had elapsed, whichever was the shorter. In practice, only one OT patient was able to remain standing for >90 s, by which time his sense of unsteadiness was graded as severe (see below). Control subjects were able to stand for 90 s during all trials. During each trial, OT patients were asked to remain standing as long as possible, even if their sense of unsteadiness had reached the severe level. However, subjects were observed closely to prevent undue distress and to prevent falling. In addition, all subjects wore a safety harness which was attached via a steel rope and pulley system to an anchor point on the wall. Subjects were asked to sit and rest between separate trials.

In order to investigate the ability of OT patients to activate their muscles voluntarily during tremor, we recorded patients during voluntary ‘rocking’ (alternating dorsiflexion/plantar flexion ankle movements) while standing. We also recorded patients as they used the strategy of their choice (e.g. voluntary movement of the legs, stepping on the spot) to maintain standing once their subjective sense of unsteadiness had reached the severe grade.

Table 1 Patient characteristics

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Duration of symptoms (years)</th>
<th>Self-assessment of approximate time able to stand unsupported</th>
<th>Falls</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>E.H.</td>
<td>65</td>
<td>F</td>
<td>8</td>
<td>30 s</td>
<td>Never</td>
<td>None</td>
</tr>
<tr>
<td>T.A.</td>
<td>68</td>
<td>F</td>
<td>17</td>
<td>Seconds</td>
<td>Never</td>
<td>Clonazepam</td>
</tr>
<tr>
<td>W.B.</td>
<td>81</td>
<td>F</td>
<td>25</td>
<td>Never</td>
<td>Occasional</td>
<td>None</td>
</tr>
<tr>
<td>W.V.</td>
<td>55</td>
<td>M</td>
<td>9</td>
<td>5 min</td>
<td>Never</td>
<td>Primidone, acetazolamide, levodopa</td>
</tr>
<tr>
<td>P.J.</td>
<td>51</td>
<td>F</td>
<td>7</td>
<td>Seconds</td>
<td>Never</td>
<td>None</td>
</tr>
<tr>
<td>M.H.</td>
<td>61</td>
<td>F</td>
<td>10</td>
<td>1–2 min</td>
<td>Never</td>
<td>Primidone</td>
</tr>
</tbody>
</table>

F = female; M = male.

Subjective unsteadiness

Subjects indicated their sense of unsteadiness by moving a slider linked to a linear potentiometer enclosed in a
lightweight box (dimensions 7.5×5×2.5 cm), which they held with both hands just in front of the abdomen. The position of the slider was converted to a proportional voltage and was sampled continuously (200 Hz). However, it contained four discrete positions to give a scale from 0 to 3, positions 0 and 3 being at the two ends and positions 1 and 2 being indicated by easily palpable clicks in between.

OT patients were instructed to (i) keep the slider in position 0 if they felt steady; (ii) move it to position 1 when they first felt a sense of unsteadiness (mild); (iii) move it to position 2 when their unsteadiness began to feel uncomfortable but not to the extent that they felt the necessity to sit or walk (moderate); and (iv) move it to position 3 if their sense of unsteadiness had reached the point at which they felt the need to sit down or commence walking (severe). To ensure that control subjects were carrying out a comparable mental task, they were instructed to use the slider to indicate their perceived degree of body sway, position 0 indicating no perceived sway and position 3 equivalent to the body sway experienced when standing on one leg with the eyes closed (although they never stood in this condition during testing). All subjects received a brief period of training to ensure that they understood the scoring task and to ensure familiarity with manipulation of the slider.

Data acquisition
All data were sampled at 200 Hz. Ground reaction forces transmitted via the feet were recorded in three dimensions from each of four transducers located at the corners of the force plate. From these data, the position of the point of application of the resultant force in two dimensions on the plate [referred to as the centre of foot pressure (CoP)] was calculated. Motion of the body was recorded in three dimensions by a non-contact, optoelectronic motion analysis system (Selspot II; Selcom AB, Pantille, Sweden), which measured the position of an infrared-emitting diode fixed over the C7 spinous process. This site was chosen as representative of global body motion without contamination from head movements. Surface EMG activity was recorded using 9-mm Ag–AgCl electrodes placed 5 cm apart over the tibialis anterior, gastrocnemius, quadriceps femoris, biceps femoris and tensor fasciae latae on the right and the lumbar paraspinals bilaterally. EMG activity was low-pass filtered at 100 Hz with a time constant of 3 ms.

Data analysis
An interactive cursor program was used. Each trial was viewed on a computer screen and various time events were determined. The start time was defined as the moment at which the marker over C7 reached its maximum height as the subject stood. From the unsteadiness indicator trace, the times to reach mild, moderate and severe subjective states were measured relative to the start time.

Objective unsteadiness was assessed both from motion of the CoP on the force plate surface and from the motion of the top of the trunk. The more unsteady the subject, the greater should be the total displacement of one or both of these variables. Therefore, we measured the length of the path described both by the CoP in two dimensions and by the top of the trunk in three dimensions over the time interval of interest. In order to reduce the effect of noise on the path length measures, the raw data were smoothed by averaging every 20 consecutive data points, which effectively reduced the sampling rate from 200 to 10 Hz. The distance moved between each consecutive pair of data points was then measured and cumulated over the total measurement interval. The resulting path length was normalized over time by dividing by the time interval of interest to give a parameter of unsteadiness with units of mm/s.

Each EMG signal was full-wave rectified and then integrated over the time interval of interest. This value was divided by the time interval to give a mean rectified EMG level with units of µV for each muscle during the measurement period. The values obtained from each of the five lower limb muscles were averaged to give a single parameter of leg EMG. Similarly, the values obtained from the two paraspinal muscles were averaged to give a single parameter of trunk EMG.

Comparison of objective unsteadiness between OT and control subjects
The objective unsteadiness of OT patients was compared with that of controls by calculating path length (CoP and body) over a fixed time interval from 2 to 25 s, time 0 being the start time as defined above. The first 2 s of data after this event were omitted in order to allow any disturbance associated with standing up to settle. In some trials, OT patients were unable to remain standing for 25 s, in which case the duration up to when the patient indicated a severe subjective state of unsteadiness was used (note that all measures were normalized with respect to time). For each parameter (CoP and body), a three-factor analysis of variance (ANOVA) with repeated measures was performed with group (OT versus controls), vision (eyes open versus eyes closed) and stance (feet apart versus feet together) as the factors. A level of $P < 0.05$ was regarded as statistically significant.

Influence of balance conditions on subjective unsteadiness in OT
In order to determine the effects of vision and stance width on subjective unsteadiness, we compared the mean duration of time OT patients spent at each subjective unsteadiness level, i.e. start to mild (grade 0), mild to moderate (grade 1) and moderate to severe (grade 2). The duration of time spent in the severe state was not analysed, as some patients were unable to remain standing immediately on reaching this grade. A three-way repeated measures ANOVA was performed with severity
was regarded as statistically significant.

**Relationship between subjective unsteadiness, objective unsteadiness and EMG in OT**

We compared path length of CoP motion, path length of body motion, leg EMG and trunk EMG during grades 1 (mild to moderate) and 2 (moderate to severe) of subjective unsteadiness in the feet-apart standing conditions. We chose not to include the grade 0 (start to mild) state or the feet-together conditions, as some of the measurement epochs were extremely short (<1 s) in the more disabled patients, giving rise to unreliable estimates of objective unsteadiness. For each variable, a two-way repeated measures ANOVA was performed with severity (grade 1 or 2) and vision (eyes open versus eyes closed) as the factors. A level of \( P < 0.05 \) was regarded as statistically significant.

**Results**

**General characteristics**

Figure 1 shows a representative example from one OT patient of the time course of change in severity of subjective unsteadiness, measures of standing performance (motion of the body and of the CoP) and EMG activity in leg and trunk muscles. Although this patient was able to continue standing for a number of seconds after the severe state was reached, this was not true of all OT patients. The general pattern, however, was quite typical in that as subjective unsteadiness increased in severity, EMG activity in the legs increased but with little obvious change in motion of the body or the CoP.

We found tremor to be present either immediately or within a few seconds of standing, consistent with the findings of others (McManis and Sharbrough, 1993). Inspection of EMG activity late in the course of single trials of standing showed that it always remained tremulous (Fig. 2). In other words, muscle activity was almost exclusively tremor-locked, implying that increased EMG activity during standing was largely due to changes in the tremor burst amplitude.

When asked to rock backwards and forwards, both severely and mildly affected patients were able to modulate leg muscle activity. Although the low-frequency modulation showed a reciprocal pattern in antagonist muscles, the underlying EMG activity remained tremor-locked (Fig. 3). This was also true of the modulation of muscle activity that occurred when patients voluntarily moved their legs as part of a strategy to prolong standing (data not shown).

**Objective unsteadiness in OT versus control subjects**

Objective unsteadiness in OT patients and control subjects during standing is shown in Fig. 4 and the statistical comparisons are given in Table 2.

The strong main effect of group, when considering both body and CoP motion, indicated that patients with OT were more unsteady than control subjects. Making the standing task more difficult, either by narrowing stance width or by closing the eyes, increased the OT patients’ sway disproportionately, as evidenced by the significant group interactions. The more consistent of these effects was related to the vision factor. This suggests that OT patients were more dependent than normal upon vision to maintain stability. This was particularly so when the feet were together (significant three-way interaction for CoP motion). The vision effect was reflected in a significantly larger Romberg quotient (eyes closed/eyes open) of trunk motion for the patient group than the controls (mean ± standard deviation Romberg quotient averaged across the feet apart and feet together conditions: controls, 1.26 ± 0.17; patients, 1.64 ± 0.32; \( t \)-test, \( P < 0.05 \)).

**Influence of balance conditions on subjective unsteadiness in OT**

Patients with OT reported that their sense of unsteadiness increased over seconds to minutes. However, the time-course of subjective unsteadiness was not fixed but escalated faster when standing was made more difficult (Fig. 5). Thus, the duration of each grade of subjective unsteadiness was significantly shortened either by closing the eyes (vision, \( P < 0.05 \)) or by narrowing the stance width (stance, \( P < 0.05 \)). There were no consistent differences in the mean duration of each subjective severity grade (severity, \( P > 0.05 \)) and none of the interaction terms was significant. This suggests that the durations of the three subjective unsteadiness grades were approximately the same and were affected equally by standing conditions.

**Relationship between standing time and objective unsteadiness**

A linear regression analysis, in which each patient contributed a pair of values for each of the four standing conditions, showed an association between objective unsteadiness and the time-course of subjective unsteadiness. The inverse of the time taken to reach a subjective state of severe unsteadiness (grade 3) was correlated \( R^2 = 0.82; P < 0.001 \) with the amount of body motion measured over a fixed time interval (Fig. 6). In other words, greater average body sway during a stand was associated with a faster escalation of subjective unsteadiness.

**Relationship between subjective and objective unsteadiness during a stand**

Measurements of objective unsteadiness (body sway and CoP) and tremor (legs and trunk) during different levels of subjective unsteadiness and vision are compared in Fig. 7, and the statistical analysis is summarized in Table 3.

There were four main findings. First, objective unsteadiness (body and CoP path rate) did not show any systematic change
with increasing subjective unsteadiness. Secondly, for a given level of subjective unsteadiness (mild to moderate or moderate to severe), objective unsteadiness was greater with the eyes closed. These two findings provide evidence of a dissociation between subjective and objective unsteadiness, and suggest that the former does not simply arise from the patient’s awareness of the latter. Thirdly, the level of leg (but not trunk) EMG increased with increasing subjective unsteadiness. This may indicate that the intensity of tremor in the legs is an important determinant of subjective unsteadiness. Fourthly, there was no difference in leg or trunk EMG between the eyes-open and eyes-closed conditions, despite the significant effect of vision on objective unsteadiness. These results suggest that changes in objective unsteadiness could occur independently of tremor amplitude and vice versa.

Discussion
The present study has shown for the first time that OT is associated with disruption of the normal balance mechanisms...
that control upright stance. OT patients consistently swayed more than normal when attempting to stand still under different standing conditions. However, the relationship between objective unsteadiness and subjective unsteadiness was not straightforward. On the one hand, there was some correspondence between the two in that there was a tendency for patients to reach a severe sense of unsteadiness sooner when their objective unsteadiness was greater. On the other hand, during the course of a single episode of standing, as subjective unsteadiness escalated from a mild to a severe state there was little change in the extent of body sway. Also, the same subjective state of unsteadiness was associated with different levels of body sway depending upon whether the eyes were open or closed. Therefore, there was also evidence of dissociation between subjective and objective unsteadiness.

### Table 2

<table>
<thead>
<tr>
<th>Effect</th>
<th>Body</th>
<th>CoP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group</td>
<td>12.19</td>
<td>33.76</td>
</tr>
<tr>
<td>Vision</td>
<td>27.81</td>
<td>21.86</td>
</tr>
<tr>
<td>Stance</td>
<td>28.33</td>
<td>7.86</td>
</tr>
<tr>
<td>Group × vision</td>
<td>14.80</td>
<td>9.41</td>
</tr>
<tr>
<td>Group × stance</td>
<td>8.63</td>
<td>0.47</td>
</tr>
<tr>
<td>Stance × vision</td>
<td>4.28</td>
<td>14.27</td>
</tr>
<tr>
<td>Group × stance × vision</td>
<td>1.53</td>
<td>5.65</td>
</tr>
</tbody>
</table>

Mean values are shown in Fig. 4. *P < 0.05; †P < 0.01; ‡P < 0.001.

This implies that the severity of a patient's sensation of unsteadiness is not simply due to awareness of the degree of body sway, and suggests that subjective and objective...
Fig. 5 Mean duration of each sensation state for the different standing conditions in OT patients. The sensation states were taken from the start of stand-up to the onset of mild unsteadiness (white), from onset of mild to onset of moderate unsteadiness (stippled) and from onset of moderate to onset of severe unsteadiness (black). Standing conditions were feet apart (16 cm), feet together (0 cm), eyes open (EO) and eyes closed (EC).

Fig. 6 Relationship between body motion and time to reach severe subjective unsteadiness in OT patients. The plot shows a linear relationship ($R^2 = 0.82$) between the reciprocal of the time taken to reach the severe state and the path rate of body motion at C7 (measured over the time interval used for Fig. 4). Standing conditions were feet apart (triangles), feet together (circles), eyes open (open symbols) and eyes closed (filled symbols).

unsteadiness may have separate pathophysiological mechanisms.

Balance disruption in OT
The disruption of balance control in OT, as evidenced by the increase in objective unsteadiness, could be due to the motor effect of abnormal tremulous activation of leg muscles. Indeed, it has been suggested previously that unsteadiness in OT arises from partially fused muscle contraction caused by the high-frequency bursting (Britton et al., 1992), implying that this might interfere with the motor activity required to maintain stability. A number of lines of evidence argue against this motor hypothesis. First, we have shown that, despite the persistence of tremor, OT patients are able to modulate their leg muscle activity appropriately in order to produce rocking movements during standing, or even to take steps. Secondly, our results suggest that OT patients are able to make fine motor adjustments during standing, as their sway was reduced when visual information was available. Thirdly, we have shown a dissociation between tremor and objective unsteadiness, since increasing intensity of leg tremor during an episode of standing was not accompanied by increase in objective unsteadiness.

We found that objective unsteadiness in OT patients was increased by closing the eyes and/or standing with the feet together, a pattern similar to that reported previously in healthy subjects (Day et al., 1993). However, OT patients showed a disproportionate increase in objective unsteadiness with more difficult standing conditions, especially eye closure.
which implies that they have abnormal dependence upon visual feedback for standing control. This, in turn, suggests that other sources of sensory information that normally contribute to balance control are in some way defective or cannot be used effectively. This could be due to disruption of either vestibular or proprioceptive feedback. It has been suggested that visually and proprioceptively mediated lower limb reflexes are the predominant mechanisms used to maintain stability during quiet standing, as under this condition vestibular reflexes have a relatively low gain (Fitzpatrick and McCloskey, 1994; Fitzpatrick et al., 1996). Therefore we feel it is less likely that a vestibular defect is the basis of increased objective unsteadiness in OT, although we cannot exclude this possibility completely. It is our suggestion that balance control is degraded in OT because of a defect in proprioceptive afferent feedback. The idea of degraded proprioceptive feedback is quite plausible. The 14–18 Hz phase-locked muscle contractions (McAuley et al., 2000) would be predicted to interfere with the normal flow of sensory information from muscle spindles and Golgi tendon organs by entraining afferent discharges. This could make the system less sensitive to the small movements that are normally detected and used to make postural adjustments. The situation could be analogous to that produced by muscle vibration, even though vibration and tremor may not produce identical patterns of proprioceptive discharge. Whole-body vibration at 18 Hz in standing subjects has been shown to increase postural sway by a factor of 2–3, although, interestingly, subjects did not report that they felt unstable (Gauthier et al., 1983). Similar results were obtained when subjects were asked to stand immediately after prolonged 18 Hz vibration of the legs while seated, an effect that did not occur if vibration was confined to the head and trunk (Gauthier et al., 1983).

**Subjective unsteadiness in OT**

The crescendo in subjective unsteadiness that occurs during standing is highly characteristic of OT and is perhaps the major cause of disability as, paradoxically, patients rarely fall. However, there have been no previous attempts to investigate the determinants of subjective unsteadiness in OT. Our patients often had difficulty when asked to describe what they meant by ‘unsteadiness’, suggesting that the sensation they experience may be unlike any other encountered under normal circumstances. Surprisingly, we found that subjective unsteadiness in OT is dissociated from objective unsteadiness, since the former increased without significant change in the latter. Thus, subjective unsteadiness cannot simply be due to awareness of abnormal body sway. In contrast, we found that increasing subjective unsteadiness was accompanied by significant increases in leg tremor. Three possibilities need to be considered. First, the increase in leg tremor may have occurred independently of subjective unsteadiness, as a consequence of increasing drive from a central tremor generator. This seems unlikely, as the amplitude of tremor in paraspinal muscles during standing stayed relatively constant. The second possibility is that the increase in leg tremor was the cause of escalating subjective unsteadiness. Thirdly, the increase in leg tremor may have been a response to escalating subjective unsteadiness.

It is possible that subjective unsteadiness in OT is caused by the same disrupted proprioceptive feedback that we hypothesize to be responsible for objective unsteadiness. Distortion of proprioceptive feedback by tendon vibration is known to produce a variety of sensations, including illusions of posture, movement and even body shape (Ecklund, 1972; Goodwin et al., 1972; Lackner and Levine, 1979; Lackner, 1988; Smetanin et al., 1993). At first glance, the dissociation between objective and subjective unsteadiness would seem to argue against the possibility that both are caused by abnormal proprioceptive feedback. However, it is possible that perceptual and balance mechanisms use proprioceptive information in different ways or involve separate but overlapping circuits. The intense sense of unsteadiness reported by OT patients is not found in Parkinson’s disease or essential tremor, even though both conditions may produce leg and trunk tremor. There are two characteristics of OT that may help to explain this difference. First, the burst frequency of OT is much higher than in other pathological tremors. Secondly, in individual subjects there is strong coherence among all the muscles that are firing in OT, but not in Parkinsonian and essential tremor (Britton et al., 1992; Koster et al., 1999). This may result in greater coherence of proprioceptive afferent activity arising from the tremor. These two factors might enhance the tremor’s capacity to interfere with perceptual mechanisms, resulting in a sensation of unsteadiness. The problem with this model is that it does not explain why the leg tremor, and hence subjective unsteadiness, grows over time.

The third possibility is that increasing leg tremor in OT is a response to rather than the cause of subjective unsteadiness. Thus, increasing subjective unsteadiness might give rise to an increased co-contracting drive to the leg extensor and flexor muscles in an attempt to stiffen and stabilize the joints, through either voluntary or reflex mechanisms (Fitzpatrick et al., 1992; Carpenter et al., 1999). However, this model fails to explain the origin of subjective unsteadiness in OT and why the sensation should escalate.

**Vicious circle hypothesis**

Patients with OT inevitably feel compelled to commence walking or to sit after a variable period of standing, corresponding to severe subjective unsteadiness as defined in the present study. Whether increased leg tremor is regarded as being primary or secondary, each model in itself fails to explain the relentless crescendo that occurs in subjective unsteadiness during standing. We feel that the most parsimonious explanation for our observations on the relationship between leg tremor and subjective unsteadiness in OT is that both mechanisms are operating. In other words,
the abnormal proprioceptive feedback that develops from the moment a patient with OT stands gives rise to an increased co-contracting drive to the leg muscles in order to stiffen the joints and increase stability. Because muscle activity remains tremor-locked, this results in increased tremulous proprioceptive feedback, which then further increases the sensation of unsteadiness, and so on in a vicious circle of escalating activity. The point at which patients decide they need to sit down or walk may be the point at which they no longer feel able to increase or sustain their co-contracting drive to the muscles.

Finally, why should patients with OT find the sense of unsteadiness less of a problem during walking, despite the persistence of tremor in the muscles of the stance leg (Britton et al., 1992; McManis and Sharbrough, 1993)? Indeed, patients will often walk to relieve their symptoms. There are a number of possible explanations based on our hypothesis. One is that, even in the presence of tremor-induced proprioceptive noise, the substantial joint motions associated with walking produce movement-related proprioceptive signals that are large enough to be extracted by the CNS. Another possibility is that, during walking, in comparison with standing, vestibular inputs contribute more and proprioceptive inputs contribute less to the perception of body state. A third possibility is that the disappearance of tremor in the swing leg, although brief, may be sufficient to break the vicious circle postulated above.

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