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Characterization of motor events in REM sleep behavior disorder

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Abstract We used video-polysomnography to characterize motor events (ME) in 14 Parkinson's disease (PD-RBD) and 18 idiopathic (iRBD) REM sleep behavior disorder cases. ME occurred predominantly in the upper limbs and were mostly simple, non-emotional, distal and focal. There were no significant differences in ME features between PD-RBD and iRBD groups. Our data suggests that RBD ME are mostly non violent. Similarity between PD-RBD and iRBD groups suggests that motor dysfunction does not affect ME features.

Keywords Parkinson's disease · REM sleep behavior disorder · REM sleep motor events

Introduction

Motor symptoms significantly affect daytime performance in Parkinson's disease (PD), but less in known regarding nighttime motor dysfunction. REM sleep behavior disorder (RBD) is a parasomnia in which patients retain muscle tone during REM sleep, allowing for the enactment of dream content (Schenck et al. 1986). RBD is common in PD patients (PD-RBD), but is also present in patients without PD (idiopathic RBD–iRBD), who do not suffer from relevant daytime motor dysfunction. Motor events (ME) during RBD episodes represent a window to assess motor dysfunction during sleep.

It has been suggested that parkinsonian symptoms do not affect PD patients during RBD dream enacting episodes (De Cock et al. 2007). A detailed analysis of motor events during RBD could help to better investigate the REM sleep motor restoration hypothesis and to provide alternative pathways for motor dysfunction treatment. If motor function restoration during REM sleep does occur, we would expect no significant differences in ME between PD and iRBD.

Our objectives were to:

1. Provide a detailed description of ME during RBD episodes in PD patients.
2. Compare ME between PD-RBD and iRBD patients.

Methods

Setting and participants

PD with RBD and iRBD patients were consecutively selected from the Egas Moniz Hospital outpatient clinic. PD was diagnosed according to the UK Brain Bank Diagnostic criteria (Hughes et al. 1992). Motor dysfunction was assessed with the Unified Parkinson's Disease Rating Scale Part III (UPDRS) (Fahn et al. 1987). Dopaminergic treatment was calculated as L-Dopa equivalent doses (DED) (Parkin et al. 2002).

All patients underwent one-night video-polysomnography, which was performed with a digital polygraph and included electrooculography, electroencephalography (six channels) electrocardiography, electromyography of the

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submental, right and left tibial muscles, recording of nasal air flow, thoracic and abdominal respiratory effort, oxygen saturation, microphone, and digital EEG-synchronized videography with infrared camera. Sleep stages and REM sleep muscular tone were scored according to the American Academy of Sleep Medicine recommendations (AASM) (Berry et al. 2015).

The International Classification of Sleep Disorders (American Academy of Sleep Medicine, 2014) criteria were used for RBD diagnosis:

1. repeated episodes of behavior or vocalization that are either documented by PSG to arise from REM or are presumed to arise from REM based on reports of dream enactment,
2. evidence of REM sleep without atonia on PSG.

To define excessive muscle activity, we used the criteria from the AASM scoring manual (Berry et al. 2015). Tonic excessive muscular activity was assessed in 30 s epochs and considered when sub-mental EMG activity exceeded twice that of background activity for more than 50% of the epoch. Phasic excessive muscular activity was measured in 3 s mini-epochs and defined as sub-mental EMG activity bursts lasting 0.1–5 s and exceeding four times that of the background. Phasic and tonic activities percentages were calculated by dividing the number of epochs with excessive activity by the total number of REM epochs. According to the work by Frauscher et al. (2012), the cutoff for excessive muscular activity was 18% for "any" (tonic or phasic) EMG activity in the mentalis muscle.

REM sleep motor events assessment

We defined REM sleep ME as any movement visible in video recordings during this sleep stage, regardless of type, duration or severity. We determined total number of events and duration, in seconds, of each event. ME were individually classified according to the system developed by Frauscher et al. (2007) which rates movement events according to type (myoclonic vs. simple vs. stereotypes vs. scenic vs. vocalizations vs. violent), emotional tone (negative vs. positive), complexity (elementary vs. complex), body region (trunk, neck/head vs. upper extremity vs. lower extremity), spatial distribution (focal vs. segmental vs. multifocal vs. global), laterality (symmetric vs. asymmetric left vs. asymmetric right) and proximal vs. distal.

Data analysis

The frequency of total ME and each ME subtype was expressed as the number of events per 10 min of REM sleep. Continuous variables were compared between these two groups by way of t tests (normal distribution) or Mann–Whitney tests (non-normal distribution). Variables are presented as mean (standard deviation). Significance was set at $p < 0.05$.

Ethics

Patients signed informed consent forms. The ethics committee of the institution approved the investigation protocol and the study was conducted in accordance with the Declaration of Helsinki.

Results

We recruited 14 PD-RBD patients: 5 females; age 66.14 (11.02) years; disease duration 6.64 (4.60) years; DED 642.64 (446.85); UPDRS 26.79 (12.30). The iRBD group was composed by 18 patients: 2 females; age 68.85 (9.50) years. All PD patients were under dopaminergic drugs. There were no patients on SSRI, or other anti-depressants, and no patients on rivastigmine or neuroleptics. Five iRBD and 3 PD-RBD patients were under clonazepam.

Mean percentage of phasic REM sleep without atonia (RWA) was significantly higher in the iRBD compared to the PD-RBD group. Mean percentage of tonic RWA was not significantly different between groups (Table 1).

Both in PD-RBD and iRBD, ME were mostly elementary, emotionally neutral or negative, affected predominantly the upper limbs and were most frequently simple, distal, focal and as frequently symmetric as asymmetric. Scenic and violent ME were rare, as well as ME involving axial regions or more than one corporal segment at a time. There were no significant differences in ME characteristics between the two groups (Table 1).

Discussion

We found that ME during REM sleep in PD-RBD and iRBD patients were mainly constituted by simple, distal and focal movements affecting the upper limbs. Movements were short lasting, and more usually neutral or negatively toned and movements with discernible scenic features, indicating dream enactment, were rare. Both Frauscher et al. (2007) and De Cock et al. (2007) reported a higher percentage of simple movements and a predominance of negatively toned and upper extremity movements. Manni et al. (2009) referred a low percentage of "acting out" movements in iRBD patients and Cygan et al. (2010) a percentage lower than 1% of scenic events. Ours and previous results show that the types of ME that clinically define RBD represent, in fact, a small fraction of the totality of motor activity in this disorder and that most

Table 1 REM sleep motor events classification in PD and iRBD patients

	PD-RBD ($n = 14$)	iRBD ($n = 18$)	p
RWA percentages—phasic activity	27.57 (24.40)	34.60 (17.49)	0.04*
RWA percentages—tonic activity	20.61 (21.35)	36.64 (21.09)	0.35
Mean duration of motor events (scs)	4.21 (3.01)	5.82 (5.47)	0.87
Number of motor events per 10 min of REM sleep			
Total	15.17 (17.32)	12.61 (10.80)	0.85
Elementary	11.62 (14.43)	9.94 (8.06)	0.65
Complex	2.71 (3.49)	3.76 (4.72)	0.46
Emotion			
Positive	0.00	1.61 (2.43)	0.12
Negative	0.31 (1.03)	2.00 (2.66)	0.68
Type			
Myoclonic	3.21 (6.03)	1.41 (1.61)	0.89
simple	6.06 (6.81)	4.90 (3.96)	0.89
Stereotypes	2.31 (3.15)	2.10 (2.23)	0.71
Scenic	1.57 (2.21)	2.48 (3.78)	0.65
Vocalizations	2.00 (3.68)	2.33 (3.96)	0.49
Violent	1.60 (2.44)	1.53 (2.35)	0.87
Body region			
Trunk	2.46 (5.43)	2.27 (2.47)	0.19
Neck/head	4.88 (5.17)	3.58 (5.29)	0.32
Upper extremity	9.96 (13.59)	8.45 (8.28)	0.62
Lower extremity	5.41 (9.56)	3.78 (3.95)	0.82
SPATIAL distribution			
Focal	7.57 (10.56)	3.48 (3.28)	0.66
Segmental	3.04 (4.03)	4.24 (4.07)	0.16
Multifocal	2.59 (4.00)	1.91 (3.02)	0.97
Global	1.64 (3.27)	1.39 (1.58)	0.48
Laterality			
Symmetric	7.46 (10.17)	4.44 (4.21)	0.57
Asymmetric	7.87 (9.52)	6.89 (5.80)	0.91
Proximal	6.34 (7.49)	4.93 (4.54)	0.62
Distal	12.14 (16.97)	9.38 (7.67)	0.91

Values are mean (standard deviation)

* $p < 0.05$

movements lack the complexity to suggest a dream enactment behavior. The rate of total events was smaller than in Frauscher et al. (2007) study, which is probably explained by methodological differences: these authors included only patients with severe RBD, while there was not such selection in our study.

PD-RBD ME characteristics were similar to those of iRBD patients, who do not suffer from daytime motor dysfunction. Oudiette et al. (2012), in a study using a different methodology for ME characterization, also did not find significant differences in the movements among three subtypes of RBD (PD, idiopathic and narcolepsy related) except for "pseudohallucinatory behaviors" (more common in PD patients). Our results thus suggest that motor dysfunction, which is the hallmark of PD and is absent or very mild in iRBD patients, does not affect the performance of ME during dream enactment episodes. It has been suggested (De Cock et al. 2007) that this phenomenon could be caused by a disjunction between pyramidal and extrapyramidal systems specific to REM sleep, during which movement control would not be affected by PD-related basal ganglia dysfunction.

In conclusion, our study, in which we provide a detailed analysis of ME during REM sleep in idiopathic and PD-related RBD cases, suggests that most episodes lack the more distinctive feature of this parasomnia (violent and dream enactment movements), while also confirming that RBD patients with and without daytime motor dysfunction

present with similar ME, which is in favor of the REM sleep motor restoration hypothesis proposed by previous studies. Further analysis of ME during REM sleep in RBD patients, including more precise and quantitative measurements of nighttime motor function and correlation with daytime deficits, would be valuable to better understand the pathophysiology of motor symptoms in PD and represent an opportunity to find alternative treatments.

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