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Video Analysis of Motor Events in REM Sleep Behavior Disorder

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Abstract: In REM sleep behavior disorder (RBD), several studies focused on electromyographic characterization of motor activity, whereas video analysis has remained more general. The aim of this study was to undertake a detailed and systematic video analysis. Nine polysomnographic records from 5 Parkinson patients with RBD were analyzed and compared with sex- and age-matched controls. Each motor event in the video during REM sleep was classified according to duration, type of movement, and topographical distribution. In RBD, a mean of 54 ± 23.2 events/10 minutes of REM sleep (total 1392) were identified and visually analyzed. Seventy-five percent of all motor events lasted <2 seconds. Of these events, 1,155 (83.0%) were classified as elementary, 188 (13.5%) as complex behaviors, 50 (3.6%) as violent, and 146 (10.5%) as vocalizations. In the control group, 3.6 ± 2.3 events/10 minutes (total 264) of predominantly elementary simple character (n = 240, 90.9%) were identified. Number and types of motor events differed significantly between patients and controls ($P < 0.05$). This study shows a very high number and great variety of motor events during REM sleep in symptomatic RBD. However, most motor events are minor, and violent episodes represent only a small fraction. © 2007 Movement Disorder Society

Key words: REM sleep behavior disorder (RBD); violent behavior; Parkinson disease; polysomnography; sleep.

In REM sleep behavior disorder (RBD), the muscle atonia normally prevailing during REM sleep is lost, and elaborate motor activity takes place in association with dream mentation.[1] According to the International Classification of Sleep Disorders (ICSD-2), diagnostic criteria of RBD include the presence of REM sleep without atonia plus one of the following: sleep related injurious, potentially injurious, or disruptive behaviors documented by history or during polysomnography, and the absence of EEG epileptiform activity during REM sleep.[2]

Schenck and colleagues[3] observed apparently dream enacting behaviors such as vocalizations (talking, laughing, yelling, swearing), scenic behavior (gesturing, reaching, grabbing), or violent movements (punching, kicking, jumping out of bed) in 87.5% of 96 RBD patients. Several studies focused on electromyographic characterization of motor activity.[4-6] A few studies have also incorporated video results, restricted to simple versus complex movements[7,8] or mild, moderate, or severe RBD intensity.[9] Gagnon and colleagues[10] provided a preliminary qualitative description of REM behavioral manifestations during video-polysomnography.

The aim of this pilot study was to provide for the first time a detailed analysis of number and types of motor events occurring during REM sleep in patients with RBD by video analysis.

METHODS

Patients

Five consecutive male patients with different parkinsonian syndromes (three with probable idiopathic Parkinson's disease, one with probable multiple system atrophy, and one with probable Lewy body dementia according to published criteria[11-13]) and severe RBD according to ICSD-2 criteria[2] were included in this study.

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Severe RBD was defined as the occurrence of gross movements during polysomnographic registration as previously published.[14] All parkinsonian patients were referred for routine polysomnography (PSG) for at least two consecutive nights because of clinically suspected parasomnia, insomnia, excessive daytime sleepiness, or suspected sleep-related breathing disorder. Patients in whom a clinically relevant sleep-related breathing disorder was confirmed by PSG were excluded from further analysis. In addition, a sex- and age-matched control group was established based on patients without RBD, who were examined for suspected sleep-related breathing disorder. Patients could be included in the control group if sleep-related breathing disorder was excluded by PSG and if they had no further history or polysomnographic manifestation of any sleep disorder (e.g. restless legs syndrome, periodic limb movement disorder).

Polysomnography

Polysomnography was performed with a digital polygraph (Brainlab, Schwarzer Inc., Munich, software version 2001, Germany) and consisted of vertical and horizontal electrooculography, electroencephalography (five channels C4-A1, C3-A2, C3-C4, O1-A2, O2-A1), electrocardiography, electromyography of the mental, submental, right and left tibial muscles, recording of nasal air flow, thoracal and abdominal respiratory effort, oxygen saturation, microphone, and digital videography. Video was recorded by an infrared camera (Elbex Inc., EX series, Regensburg, Germany) and processed by a data rate of 1,500,000 bits/second. The screen resolution for video analysis was 1280 $\times$ 960 pixels.

Sleep stages were scored according to the criteria of Rechtschaffen and Kales[15] with allowance to score REM sleep despite persistence of tonic or phasic muscle activity.[16] Arousals were defined according to published criteria.[17] Furthermore, the tonic and phasic components of REM sleep were scored separately from the mental and submental EMG as proposed by Lapierre and Montplaisir.[4] Data are gathered from both nights, presented for each night separately.

Definition of Motor Events

The video classification system used for this analysis was developed at our sleep laboratory (BH, EB, BF). The videos of all REM episodes were carefully looked through by one of the authors (BF). In order not to overlook small movements in different body parts, the videos were looked through several times. To check reproducability of the findings, the analysis of 10% of motor events (n = 139) was cross-checked by a second rater (VG). A list with the findings of scorer 1 was given to scorer 2, and agreement or disagreement for every single motor event and its classification was assessed by independent video viewing. The mean interscorer agreement was 0.91 ($P < 0.001$). In case of divergent scoring or any uncertainties during scoring, video event classification was supervised by one senior neurologist (BH).

All visible movements regardless of type, amplitude, and duration were described as motor events. The exact onset and offset of motor events was observed in the video, and the *duration* determined with a hand-held chronograph. Motor events shorter than 2 seconds were presented together because a reliable measurement of duration in the video was not possible for a time span shorter than 2 seconds.

Type of Motor Event.

Based on videographic analysis, the following classification of motor events was used: Myoclonic events were defined as sudden, brief, jerky, involuntary movements involving extremities, face, and trunk.[18] Simple motor events were defined as small excursions that usually would not be noticed by a sleeping bed partner. For example, a myoclonic event can be elementary simple in case of isolated finger twitches, or major in case of a whole body jerk. Stereotypes refer to automatism-like events (e.g. smacking or fumbling). The term complex/scenic is used to describe apparent "acting out" of dream contents or movements different from elementary simple events in term of complexity of action. Events were further subdivided into vocalizations (e.g. talking, crying) and orofacial events (e.g. clenching or grimacing). Vocalizations were subcategorized in those with and without comprehensible speech and those not associated with any visible motor event. Moreover, the apparent emotional state (apparently positive emotions, e.g. when the patient is laughing, or apparently negative emotions, e.g. when the patient is screaming or crying, no change in emotional state) was assessed for complex/scenic behaviors and vocalizations. Violent refers to forceful and vehement motor events that could potentially injure a bed partner (e.g. kicking or punching). Motor events were dichotomized into elementary motor events (myoclonic motor events, simple motor events, stereotypes) and complex behaviors (complex/scenic motor events, violent motor events).

Topographical Distribution.

In this section, involved body parts were listed, subdivided in head/neck, trunk, upper extremity, and lower extremity. *Spatial distribution* of motor events was subdivided into categories as has been described previously.[19] Focal motor events are localized to one

TABLE 1. *Clinical characteristics of the patient sample*

	Pat. 1	Pat. 2	Pat. 3	Pat. 4	Pat. 5
Gender	M	M	M	M	M
Age (yr)	65	52	73	59	71
Clinical diagnosis	MSA	LBD	PD	PD	PD
Hoehn and Yahr stage in "on"	4	3	3	3	3
Age at onset of motor symptoms	55	47	62	45	65
Levodopa (mg/day)	800	–	1,060	1,000	800
Pergolide (mg/day)	–	–	1.5	–	–
Pramipexole (mg/day)	–	1.05	–	2.8	–
Entacapone (mg/day)	400	–	500	–	400
Amantadine (mg/day)	–	–	300	–	–
Choline-esterase inhibitor use	–	+	–	–	+

MSA, multiple system atrophy; LBD, Lewy body dementia; PD, Parkinson's disease.

body part. Segmental ones involve one or more contiguous body parts. Multifocal indicates involvement of noncontiguous body parts and generalized motor events involve multiple or all body parts. Additionally, motor events are classified as proximal, distal, and axial. *Laterality* was described as unilateral with motor events involving only one side of the body or symmetric when being identical on both sides. Additionally, a subdivision into proximal and distal motor events was performed.

Statistics

SPSS 12.0 for Windows (SPSS, Inc., Chicago, IL) was used for all statistical analyses. Descriptive statistics are given as means ± standard deviations as well as frequencies (percentages of motor events per 10 minutes of REM sleep), as applicable. Motor events were examined for associations using χ^2-tests. Correlations were calculated using Spearman's ρ if data were not normally distributed. Differences in number and types of motor events between patients and controls were examined using Mann–Whitney-*U* test. In case of multiple comparisons, Bonferroni correction was applied. *P*-values below 0.05 were considered significant.

RESULTS

Demographics

Mean age of the patients was 64.0 ± 8.7 years (range: 52–73 years). Further characteristics of the patients are given in Table 1. Controls were sex- and age-matched (5 men; mean age, 64.0 ± 8.7 years). None of the controls had or has a history of central nervous system active medication.

Polysomnographic Results

The patient group showed a severely disturbed sleep efficiency (68.7% ± 22.6% of sleep period time (SPT, 475.1 ± 20.4 minutes)). Sleep latency was 68.7 ± 22.6 minutes. The percentages of sleep stages were as follows: stage 1 sleep, 29.1% ± 23.6% of SPT; stage 2 sleep, 29.6% ± 20.0% of SPT; slow wave sleep, 1.8% ± 3.8% of SPT; REM sleep, 9.0% ± 5.9% of SPT. REM sleep was registered in 9 of 10 nights. In the control group, sleep efficiency was 82.1% ± 12.7% of SPT (451.8 ± 31.9) with slightly increased S1 sleep (12.3% ± 2.4%), whereas SWS (5.4 ± 4.5) and REM sleep (18.5 ± 5.6) were slightly reduced. Detailed data concerning REM sleep in the patient and the control group are given in Table 2.

TABLE 2. *REM sleep data: Polysomnography, electromyography, and video analysis*

	Pat. 1		Pat. 2		Pat. 3		Pat. 4		Pat. 5	Controls,	Patients,	
	1*	2	1	2	1	2	1	2	2	mean ± SD	mean ± SD	*P*-value
REM episodes (n)	2	3	2	6	1	3	2	1	2	4.3 ± 1.2	2.4 ± 1.5	0.014
Total REM duration (min)	41	34	13.5	58.5	26	38.5	11	8	49	105.3 ± 57.8	31.1 ± 17.7	<0.001
Phasic EMG activity (%)	29.7	25.6	26.4	40.3	45.3	45.3	34.2	41.3	46.7	8.8 ± 2.7	33.5 ± 14.2	<0.001
REM atonia (%)	17.1	48.5	1	1.6	0	1.3	18.2	31.3	46.9	97.3 ± 1.4	16.6 ± 19.5	<0.001
Motor events in video (n)	175	80	47	225	204	344	76	58	183	29.3 ± 14.9	154.7 ± 98.2	<0.001
Motor events/10 min REM	43	24	35	38	78	89	69	73	37	3.6 ± 2.3	54.0 ± 23.2	<0.001

*1 and 2 represent nights.

TABLE 3. *Motor events in video PSG recordings: Types of movements*

	Pat.1		Pat.2		Pat.3		Pat. 4		Pat. 5	Controls, mean ± SD	Patients, mean ± SD	*P*-value
	1*	2	1	2	1	2	1	2	2			
Total events	43	24	35	38	78	89	69	73	37	3.6 ± 2.3	54 ± 23.2	<0.001
Elementary	37	21	27	35	72	64	56	66	29	3.4 ± 2.4	45.3 ± 19.3	<0.001
Myoclonic	8	5	2	18	35	29	35	33	9	1.7 ± 1.4	19.3 ± 13.7	<0.001
Simple	32	17	22	24	56	51	31	50	21	4.7 ± 5.2	33.8 ± 14.9	<0.001
Stereotypes	8	3	8	1	12	10	5	4	2	0.1 ± 0.1	5.9 ± 3.8	<0.001
Complex	3	3	4	5	11	16	7	4	6	0.3 ± 0.4	6.6 ± 4.3	<0.001
Complex/scenic	3	3	4	5	11	15	6	3	6	0.3 ± 0.4	6.5 ± 4.3	<0.001
Violent	1	1	0	3	0	6	1	1	1	0	1.5 ± 1.7	<0.001
Vocalization	7	3	0	3	1	4	6	9	11	0.2 ± 0.3	5.0 ± 3.7	0.001

Values are given in numbers of motor events per 10 min REM sleep. More than one category was applicable to specify motor events.
*1 and 2 represent nights.

Descriptive Analysis of Motor Events

In the patient group, 1,392 motor events were analyzed (54 ± 23.2 events/10 minutes of REM sleep). Seventy-five percent of the events were shorter than 2 seconds. The mean duration of events longer than 2 seconds was 10.6 ± 9.2 seconds. One-thousand-three-hundred-and-sixty-five motor events (98.1% of total motor events) did not go along with arousals. In the controls, a total of 264 motor events (3.6 ± 2.3 events/10 minutes of REM sleep) were registered. The frequency of motor events between patients and controls differed significantly ($P < 0.001$)

Type of Movement.

In the RBD group, 1,155 motor events (83.0%) were classified as elementary, 188 (13.5%) as complex behaviors, and 146 (10.5%) as vocalizations. Myoclonic events were comparably frequent (n = 469, 33.7%). Complex/scenic events were quite rare (n = 169, 12.1%). Of the complex/scenic events, 67 (39.6%) were associated with no apparent change in emotional state, 40 (23.7%) with apparently negative emotions, and 8 (4.7%) with apparently positive emotions. Only a small fraction were so-called violent motor events (n = 50, 3.6%). Periodic leg movements were rare (n = 8, 0.6%). For detailed information see Table 3. Controls had mostly minor motor events, whereas complex/scenic behaviors were rare and violent behaviors were not registered. Types of motor events differed significantly between patients and controls ($P \leq 0.001$).

Topographical Distribution.

Table 4 presents the distribution of motor events per patient and night and the control group according to involved *body parts*. In this section, also more than one category was applicable. Only one body part was involved in most motor events (69.2% ± 11.5%/per patient and night), and the remaining events involved two to four body parts. According to *spatial distribution*, 30.4% ± 11.6% of motor events per patient and night were focal, 30.3% ± 4.6% segmental, 22.8% ± 8.7% multifocal, 13.8% ± 8.4% generalized, and 2.8% ± 2.6% classified as others. Overall, involvement of distal and axial body parts was more frequent than that of proximal body parts (Table 5).

Laterality.

Unilateral motor events (n = 575) were frequent (41.3% of total motor events) and involved in 83.0% distal, in 45.9% proximal, and in 26.4% axial body parts,

TABLE 4. *Motor events in the video PSG recordings: Involved body parts*

	Pat. 1		Pat. 2		Pat. 3		Pat. 4		Pat. 5	Controls, mean ± SD	Patients, mean ± SD	*P*-value
	1*	2	1	2	1	2	1	2	2			
Total events	43	24	35	38	78	89	69	73	37	3.6 ± 2.3	54 ± 23.2	<0.001
Head	27	15	15	31	38	34	21	35	27	1 ± 0.9	26.9 ± 8.6	<0.001
Trunk	2	2	1	10	8	10	26	15	6	1.4 ± 1.6	8.9 ± 7.8	0.003
Upper extremity	22	12	18	20	52	72	52	50	21	1.6 ± 2.2	35.3 ± 21.1	<0.001
Lower extremity	4	3	7	13	7	10	31	23	6	1.1 ± 0.9	11.4 ± 9.4	<0.001

Values are given in numbers of motor events per 10 min REM sleep. More than one category was applicable to specify motor events.
*1 and 2 represent nights.

TABLE 5. *Motor events in video PSG recordings: Spatial distribution*

	Pat. 1		Pat. 2		Pat. 3		Pat. 4		Pat. 5	Controls,	Patients,	
	1*	2	1	2	1	2	1	2	2	mean ± SD	mean ± SD	*P*-value
Total events	43	24	35	38	78	89	69	73	37	3.6 ± 2.3	54 ± 23.2	<0.001
Distal	11	7	0	17	31	49	40	33	13	1.8 ± 2.2	22.2 ± 16.5	0.003
Proximal	19	12	22	20	39	68	49	48	21	0.7 ± 0.7	32.9 ± 18.9	<0.001
Axial	27	14	4	31	40	36	33	38	27	1.8 ± 1.8	27.9 ± 11.8	<0.001

Values are given in numbers of motor events per 10 min REM sleep. More than one category was applicable to specify motor events.
*1 and 2 represent nights.

mostly by head and neck movements. Symmetric motor events were quite rare (n = 129, 9.3% of total motor events).

Vocalizations were present in 146 events. In a subtype analysis, 64 (43.8%) vocalizations were registered together with complex/scenic behavior, 37 (25.3%) isolated, 11 (7.5%) with orofacial motor events, and 7 (4.8%) in the context of violent behavior. Ninety-four (64.4%) consisted of vocalizations that were not associated with any visible motor event, 49 (33.6%) contained incomprehensible speech, and 3 comprehensible speech (2.1%). Besides, of the 146 vocalizations, 26 (17.8%) were associated with apparently negative emotions, 11 (7.5%) with apparently positive, and 109 (74.7%) with no apparent change in emotional state.

Proximal movements were significantly stronger associated with movements of the trunk than distal events ($P < 0.001$). Violent motor events were strongly associated with arousals ($P < 0.001$). There was no night-to-night correlation between type of motor events, their topographical, and spatial distribution per individual patient.

DISCUSSION

This is the first study to systematically examine motor events in patients with REM sleep behavior disorder (RBD) by video analysis and to compare them with healthy controls to better characterize this disorder.

The major finding of this study is the high number of motor events (n = 1392, 5.4 motor events/minute) during REM sleep in RBD compared with sex- and age-matched controls. The high interindividual variability in number and types of motor events indicates that RBD features a broad clinical spectrum, which might be related to different degrees or localizations of neurodegeneration.[20-22] Even in severe RBD, as in our patients, only a small fraction of motor events were so-called violent movements (3.6%). Most events were elementary movements such as jerks and twitches (83.0%), whereas complex behaviors were less frequent (13.5%).

In sleep, video analysis has been applied to patients with nocturnal frontal lobe epilepsy,[23-28] demonstrating that video analysis is a feasible way of characterizing motor activity during sleep and that it could help in subtyping different disease features. In RBD, a few studies have also incorporated video results.[7-9] Sforza and collegues[7] examined REM sleep of RBD patients subdividing movements according to the EMG duration into myoclonic or simple events and according to the video into simple versus complex events. The same classification was used by Fantini and colleagues.[8] A more detailed qualitative video analysis of number and types of motor events, however, was not performed in those studies. Iranzo and Santamaria[9] have recently provided a classification of abnormal behavior in RBD according to its overall intensity (mild, e.g. murmuring; moderate, e.g. talking; severe, e.g. loud talking, shouting). In their study, only 7.7% of multiple system atrophy patients and 15.6% of Parkinson's disease patients suffered from severe RBD.

We were able to show that even in severe RBD, violent movements represent only a small fraction of motor events. Although our study sample was small, we think that the very high number of motor events as well as the use of a sex- and age-matched control group corroborates our findings. As a particular strength, our study does not attribute qualitative scores to particular RBD patients, but analyzes phenomenology on an event-to-event basis, using qualitative as well as quantitative descriptors. Therefore, PSG reviewers should scrutinize both the PSG and video in detail to adequately assess anyone with possible RBD, and not simply rely on florid and violent complex behaviors to be exhibited during REM sleep on video-PSG to confirm or negate the diagnosis of RBD. Because we included patients with severe RBD, all had violent motor events. However, mild RBD as a possible continuum from REM sleep without atonia[29] could be missed when not carefully analyzing the video.

Fantini and colleagues[30] reported periodic limb movements in sleep (PLMS) indices higher than 10 hr^{-1} in 70% (28/40) of RBD patients. In that study only patients with idiopathic RBD were included. In our study, PLMS represented only a small fraction of motor events (0.6%). This might be related to intake of dopaminergic agents in our patient group, which consisted of parkinsonian patients with symptomatic RBD. Moreover, the classification of motor events was made exclusively by video in the present study, and small toe movements concealed by the blanket may have been missed. We included patients with various parkinsonian syndromes, because these are frequently associated with RBD.[10] In line with previous studies in patients with Parkinson's disease or parkinsonian syndromes, our polysomnographic data indicate a reduction in total sleep time, frequent awakenings, a reduced amount of deep sleep, as well as REM sleep[31-35] compared with published values for normals.[36] Frequent awakenings and subsequent sleep fragmentation as well as drug induced suppression of REM sleep represent probable reasons for the reduced amount of REM sleep of 9% found in our patient group.

There are several potential limitations of our study. First of all, all patients analyzed in this study were suffering from parkinsonian syndromes, which might have influenced the nature and expression of the motor manifestations observed during REM sleep. On the other hand, there is increasing evidence that even idiopathic RBD is associated with subtle motor impairments[37] and that idiopathic RBD not only preceeds parkinsonian syndromes but may de facto be associated with very early or subclinical forms.[38,39] Additionally, patients were on dopaminergic therapy, which might have influenced RBD severity as described in small case series or single case reports.[8,40,41] Including patients with severe RBD might have introduced a selection bias. However, we aimed to investigate the whole spectrum of motor events, which may be assumed to be the broadest in severe RBD. Another potential drawback is that only movements visible in the video could be classified. Detection of very small movements was often difficult because of the infrared lighting and the limited resolution of the recordings. To minimize this bias, all videos were looked through several times in regard to different body parts. Furthermore, most of the patients did not tolerate sleeping without a blanket, which may have resulted in an underestimation especially of minor movements of the lower extremities.

In summary, this pilot study shows that patients with RBD associated with different parkinsonian syndromes exhibit a very high number of motor events during REM sleep. The great variety of events ranges from vocalizations and automatisms to violent behavior, and may involve all body parts. However, most of the movements are minor, and violent movements represent only a small fraction of motor events even in severe RBD.

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