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Original Article

REM sleep behaviour disorder (RBD) and its associations in young patients

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ABSTRACT

Study objectives: To retrospectively examine the characteristics of a population of patients <50 years of age with clinical and polysomnographic features diagnostic for RBD.
Methods: Review of our sleep centre's database for patients with RBD diagnosed over the last 7 years. Ninety-one patients were separated into two groups according to their age at the time of diagnosis (<50 y and ⩾50 y). Clinical and polysomnographic data were reviewed.
Results: Sixty-two were male; mean age was 52 ± 19 y. Thirty-nine were <50 y. In the group <50 y there was a male predominance but in a smaller proportion (M:F = 1.4:1) compared with the group ⩾50 (M:F = 3:1). Seventy-six patients complained of abnormal behaviour (AB) during sleep, 12 with narcolepsy complained of excessive daytime sleepiness (EDS) with the AB being elicited only during consultation, and three complained of both EDS and AB. All patients, except one in the group ⩾50, described AB related to vivid dreams with violent content. The majority of the patients had the idiopathic form of RBD in both groups (51.2% group <50, 63.4% group ⩾50). The secondary form was associated with narcolepsy in 38.4% of patients in the group <50 y and with a synucleinopathy in 28.8% of patients in the group ⩾50. A strong association was noted between RBD and non-REM parasomnias.
Conclusions: In a population of patients with RBD presenting to a regional sleep laboratory, more than one-third of patients were <50 y at time of diagnosis. The commonest associated disorder was narcolepsy in patients <50 y, and synucleinopathy in those ⩾50 y. The coexistence of RBD with a NREM parasomnia was not uncommon in cases of idiopathic RBD affecting patients <50 y.

1. Introduction

Rapid eye movement (REM) sleep behaviour disorder (RBD) is a parasomnia characterized by a history of excessive nocturnal motor activity during REM sleep. RBD usually manifests with the violent enactment of dreams; its characteristic polysomnographic finding is the absence of the normal muscle atonia during the stage of REM sleep [1,2]. The disorder can be categorized as acute or chronic in onset, and idiopathic or secondary according to associations with other functional or structural disorders of the nervous system [1]. While transient (acute) RBD can be seen after ingestion of certain drugs or during drug and alcohol withdrawal, the chronic type is usually idiopathic or associated with an underlying neurological disorder. Acute and secondary cases have contributed to the understanding of possible pathogenic mechanisms underlying RBD [3–6]. The increasingly recognized association between secondary RBD and synucleinopathies [6,7] in the older population has guided current research efforts in an attempt to clarify the relationship between RBD and the underlying neurodegenerative process in the brainstem, especially in patients with Parkinson's disease. In contrast to RBD in the elderly, there is a paucity of knowledge concerning the natural history of idiopathic and secondary RBD in younger patients with regard to its development, associations and progression. In an effort to gather further information regarding RBD in a young population we reviewed all cases who received a diagnosis of RBD in our sleep unit over the last seven years.

2. Materials and methods

2.1. Subjects

The anonymised database of all patients seen in our Regional Sleep Centre between 1999 and 2006 was reviewed retrospectively. The study cohort included patients who received the diagnosis of RBD over this period. All patients with a history of abnormal behaviour during sleep with purposeful movements related to dreaming and dream recollection, and at least one full-night video-polysomnography which confirmed the diagnosis of RBD were selected. All clinical and polysomnographic data were reviewed.

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2.2. Video-polysomnography (VPSG)

A full-night diagnostic VPSG had been performed in each subject. To determine the stages of sleep an electroencephalogram (using two central and two occipital scalp electrodes connected to the contralateral electrode to form the following EEG channels: C4–A1, C3–A2, O2–A1, O1–A2), electro-oculogram, and electromyogram of the submentalis muscle were obtained. Blood oxygen saturation was recorded with the use of a finger pulse oximeter. Thoracoabdominal excursions were measured qualitatively by respiratory effort sensors placed over the rib cage and abdomen. Snoring was detected with a vibration snore sensor and body posture with a body position sensor. Airflow was monitored using a nasal cannula pressure transducer. Muscle activity was recorded from the submentalis muscle, as well as from the anterior tibialis muscles bilaterally. All variables were recorded with a digital acquisition system. Sleep stage was scored manually in 30-s epochs [8]. Respiratory events were scored by an experienced technician using standard criteria [9]. For the estimation of REM sleep without atonia we used the criteria described in the AASM manual for scoring of sleep and associated events [10]. Tonic and phasic increases concurrent with respiratory arousals and snoring signal artefacts were excluded from analysis. For comparison purposes we quantified the percentage of 30-s REM-sleep epochs with tonic activity (at least 50% of the duration of the epoch having chin EMG amplitude greater than the minimum amplitude in NREM) in the submental channel and phasic activity (at least 50% of the duration of the epoch containing a burst of muscle activity lasting for 0.1–0.5 s and exceeding the background activity by at least four times) in the submentalis, right and left anterior tibialis. Any abnormal behaviour during REM periods as detected by the video camera or reported by the technician was reviewed.

For the diagnosis of RBD and any other concomitant sleep disorder the clinical and polysomnographic criteria of ICSD-2 were used [11]. RBD was classified as secondary in cases of a comorbid neurological disorder/diagnosis (e.g., narcolepsy, neurodegenerative disease), or when it was associated with drug ingestion or other condition (alcohol withdrawal etc.). All other cases were termed idiopathic.

Patients were divided into two groups according to age of diagnosis. The first group included all patients under the age of 50 and the second patients who were 50 years or older at the time of testing. We did not use the age of RBD onset as a parameter of categorization as many patients, especially patients in the younger group, could not specify the age of symptom onset.

3. Results

Data are presented as mean values (±standard deviation, range). Demographic and clinical data are summarized in Table 1. One hundred and two patients were diagnosed with RBD between 1999 and 2006. Eleven patients were excluded despite having the characteristic clinical picture, because they did not fulfill PSG criteria. Four of them had very poor sleep efficiency with no REM sleep recorded during the night of PSG, three had very disruptive REM due to severe sleep apnoea and four had a normal PSG study.

Ninety-one patients fulfilled the clinical and PSG criteria. Sixty-two were male (68%) and 29 female. The mean age at the time of diagnosis was 52 years (±19, 15–88). In 15 patients the time of RBD presentation could not be established. In the remaining cohort the mean duration of the disorder was 6.8 years (±6, 0.5–26). Seventy-six patients complained of abnormal behaviour during sleep, 12 complained of excessive daytime sleepiness (EDS) with the abnormal behaviour being elicited during the consultation and three complained of both EDS and abnormal behaviour. The mean Epworth Sleepiness Scale Score was 11 (±6, 1–24). Mean time of follow-up was 2 years (±2, 0.5–8). Fifty-four cases (59%) were categorized as idiopathic and 37 as secondary. Thirty-nine patients (43%) were under 50 years, and 52 were aged 50 or over.

3.1. Patients <50 years

This group consisted of 23 male and 16 female patients. Mean age was 32 years (±9, 15–49). All patients suffered from abnormal behaviour during sleep related to vivid dreams with violent content. Twenty patients (51%) had idiopathic RBD and 19 had secondary RBD. Among the patients with idiopathic RBD, 13 gave a history of a more complex behaviour than that classic of RBD. These patients complained of abnormal behaviour during sleep, which consisted of periods of acting out during their dreams; they could all be easily aroused by their partners during this period and had retained dream recollection. In addition they described periods of sleep talking or sleep walking – during which they could not be aroused easily – and there was no recollection of event(s) the next morning. In all cases the disorder could be exacerbated by stress and/or alcohol consumption. In seven of these patients the PSG revealed spontaneous arousals from slow wave sleep in addition to loss of atonia during REM periods. Secondary RBD was associated with narcolepsy in 15 patients, antidepressant use in three (fluoxetine, venlafaxine, paroxetine – one each) and Parkinson's disease in one.

Among the patients with narcolepsy, 12 complained of EDS, one of EDS and abnormal behaviour and only two reported abnormal behaviour during sleep subjectively, despite being very sleepy, with an Epworth Sleepiness Score (ESS) >20. All narcolepsy patients had undergone a Multiple Sleep Latency Test and had a mean sleep latency of 3 ± 0.5 over four naps and an average of 3.5 ± 0.5 sleep onset during REM sleep periods. Three patients with narcolepsy were under treatment with selective serotonin reuptake inhibitor (SSRI) antidepressants when they undertook the PSG study. Follow-up time was 2 ± 2 years. During the period of fol-

Table 1
Demographics and clinical data of study participants

Age groups	No	Male/female	Mean age (years)	Mean duration of RBD[a] (years)	Epworth sleepiness scale	Follow up time (years)	Main complain ABh[b]	Main complain EDS[c]	Main complain EDS and ABh
<50%	39	23/16	32 ± 9	11 ± 7	11.5 ± 6	2 ± 2	26	12	1
	43	53/47					67	31	2
⩾50%	52	39/13	67 ± 8	8 ± 10	10 ± 5	1.5 ± 1.6	50	0	2
	57	68/32					96	0	4
Total%	91	62/29	52.2 ± 19	6.8 ± 5.7	11 ± 6	2 ± 1.7	76	12	3
		68/32					83.5	13.2	3.3

[a] RBD, REM sleep behaviour disorder.
[b] ABh, abnormal behaviour during sleep.
[c] EDS, excessive daytime sleepiness. Data are presented as mean values ± standard deviation.