# ALEXITHYMIA AND NIGHTMARE DISTRESS IN REM BEHAVIOR DISORDER

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# Alexithymia Associated with Nightmare Distress in Idiopathic REM Sleep Behavior Disorder

Isabelle Godin, BA<sup>1,2</sup>; Jaques Montplaisir, MD, PhD<sup>1,4</sup>; Jean-François Gagnon, PhD<sup>1,3</sup>; Tore Nielsen, PhD<sup>1,4</sup>

<sup>1</sup>Center for Advanced Research in Sleep Medicine, Hôpital du Sacré-Coeur de Montréal, Québec, Canada; <sup>2</sup>Department of Psychology, Université de Montréal, Québec, Canada; <sup>3</sup>Department of Psychology, Université du Québec à Montréal, Québec, Canada; <sup>4</sup>Department of Psychiatry, Université de Montréal, Québec, Canada

**Study Objectives:** Idiopathic REM sleep behavior disorder (iRBD) is characterized by atypical REM sleep motor activity, vivid dreams and nightmares, and dream-enacting behaviors that can result in injuries to the patient and bed partner. It is also a known predictor of Parkinson disease (PD). Alexithymia has been associated with disturbances in sleep and dreaming (e.g., nightmares) and is a non-motor symptom of PD. We assessed alexithymia and disturbed dreaming in iRBD patients with the aim of determining if these two factors are elevated and interrelated among this population.

Design: Questionnaire study of clinically diagnosed patients.

Setting: Clinical sleep disorders center.

Patients or participants: Thirty-two iRBD patients and 30 healthy age- and sex-matched control participants.

**Measurements and Results:** Participants completed the 20-item Toronto Alexithymia Scale (TAS-20), the Dream Questionnaire, and the Beck Depression Inventory. iRBD patients obtained higher TAS-20 total scores (62.16  $\pm$  13.90) than did controls (52.84  $\pm$  7.62;  $F_{1,59}$  = 10.44, P < 0.01), even when controlling for depressive symptoms, and more frequently attained the suggested cutoff for alexithymia than did controls (P < 0.01). iRBD patients obtained higher scores on the Difficulty Identifying Feelings alexithymia subscale. For both iRBD and control groups, the Difficulty Indentifying Feelings subscale correlated positively with the Nightmare Distress scale of the Dream Questionnaire.

**Conclusions:** Elevated alexithymia scores among idiopathic rapid eye movement sleep behavior disorder patients, and especially a difficulty in identifying feelings, parallels evidence of dysautonomia in this population. The higher incidence of distressing nightmares and the association of nightmares with alexithymia further extend similar findings for both clinical and non-clinical samples and suggest that an affect regulation disturbance may be common to the two sets of symptoms.

Keywords: REM sleep behavior disorder, alexithymia, nightmares, dysautonomia

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#### INTRODUCTION

Idiopathic REM sleep behavior disorder (iRBD) is characterized by potentially injurious sleep motor activity associated with dream mentation<sup>1,2</sup> and an intermittent or complete loss of normal REM sleep atonia.<sup>1,3</sup> Dream content is vivid.<sup>2,5,6</sup> and thought to be more violent in RBD patients than in healthy adults, although the latter finding has been challenged.<sup>4</sup> RBD is a known risk factor for neurodegenerative syndromes,<sup>7</sup> especially Parkinson disease (PD), multiple-system atrophy, and dementia with Lewy bodies. It often precedes the onset of these syndromes by as much as 10 to 15 years.<sup>7</sup>

The emotional symptoms of iRBD patients are not yet well understood. Elevated levels of daytime passivity have been noted anecdotally<sup>5</sup> and documented as lower scores on aggressiveness questionnaires.<sup>2,4</sup> Higher anxiety levels have been reported among children with RBD<sup>9</sup> and higher anxiety and depression levels among adults with PD.<sup>10</sup> PD patients show higher levels of alexithymia features.<sup>11,12</sup> especially patients

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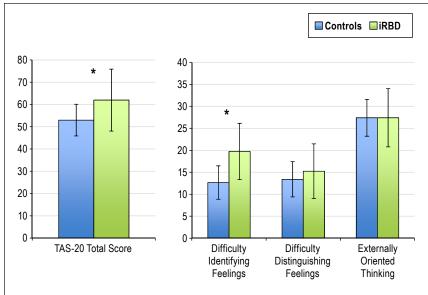
Address correspondence to: Tore Nielsen, Center for Advanced Research in Sleep Medicine, Hopital du Sacré-Coeur de Montréal, 5400 boul. Gouin Ouest, Montréal, Québec, Canada (H4J 1C5); Tel: (514) 338-2222 ext. 3350; Fax: (514) 338-2431; E-mail: tore.nielsen@umontreal.ca

with postural instability/gait symptoms.<sup>13</sup> But alexithymia has not yet been evaluated in iRBD patients.

Alexithymia most commonly refers to a difficulty in identifying and describing feelings, a difficulty in distinguishing feelings from bodily sensations of emotional arousal, a lack of fantasies, and an overly concrete style of thinking. <sup>14-18</sup> It is known to be a risk factor for depression, <sup>19</sup> anxiety, dissociative symptoms in PTSD, <sup>20-22</sup> somatic symptoms, <sup>23</sup> eating disorders, <sup>24</sup> and poor response to psychotherapy. <sup>25</sup> Depressive patients with alexithymia show more severe symptoms than their non-alexithymic counterparts. <sup>25,26</sup> Alexithymia is also associated with reduced life satisfaction <sup>27</sup> and is prospectively associated with all-cause mortality, even among healthy individuals. <sup>28</sup>

Alexithymia has been associated with general alterations in dreaming, <sup>18</sup> e.g., individuals high in alexithymia have poorer recall of their dreams, have simpler and shorter dreams, and show a lack of interest in their dreams. <sup>29-32</sup> It has also been associated with many sleep problems, including insomnia, excessive sleepiness, and sleepwalking. In particular, it has been reliably linked to the occurrence of nightmares. <sup>33-36</sup>

The frequent occurrence of nightmares among RBD patients, and RBD's association with PD, suggest that iRBD patients, too, may suffer from an alexithymia deficit. The aim of this study was therefore to assess alexithymia and nightmare distress in iRBD patients using standardized measures and to compare this group to a matched group of healthy participants. We expected to find iRBD patients to have a higher prevalence and higher



**Figure 1**—Comparisons between iRBD and Controls on the Toronto Alexithymia Scale-20 total score and the three TAS-20 subscales: iRBD patients scored higher on TAS-20 total score (left panel) and the Difficulty Identifying Feelings subscale (right panel). \*P < 0.05.

levels of alexithymia and greater nightmare distress. We also expected to find a correlation between alexithymia levels and nightmares in both controls and iRBD participants.

#### **METHODS**

#### **Participants**

Thirty-four iRBD patients (25 men; mean age:  $62.0 \pm 10.8$ years) and 31 healthy age and sex matched control participants (19 men; mean age,  $56.9 \pm 16.61$  years) were studied. Control participants were recruited through a newspaper advertisement or by word of mouth. All were screened for mental, neurologic or medical conditions with a medical history, a neuropsychological battery, polysomnographic (PSG) recording, and the BDI-II. Patients showed no signs of any other neurodegenerative syndrome or medical condition that might have an impact on sleep (sleep apnea syndrome, insomnia, etc). Two patients and one control had missing responses on their questionnaires and were thus removed from the analyses. Thus, the final sample consisted of 32 iRBD patients (23 men; mean age:  $61.5 \pm 11.12$  years; education:  $12.97 \pm 3.94$  years) and 30 controls (19 men; mean age,  $57.2 \pm 14.47$  years; education:  $13.59 \pm 3.19$  years). Six of the iRBD patients had a history of mood disorder, and 3 reported having previously suffered an anxiety disorder. At the time of the PSG, 4 RBD patients took an anticonvulsant medication, and 6 took both an anxiolytic and an antidepressant. Of the control participants, none had a history of psychiatric trouble, and none was taking a psychoactive medication.

RBD was diagnosed by a physician according to the International Classification of Sleep Disorders, second edition (ICSD-II) criteria, <sup>37</sup> that is, the presence of excessive muscle activity during REM sleep (> 30% of REM sleep with tonic EMG activity or > 15% of REM sleep with phasic EMG activity) and  $\geq 1$  of the following: history of disruptive sleep behaviors, abnormal REM sleep behaviors confirmed by PSG monitoring with synchronized videotape.

A university-affiliated hospital ethics review board approved the protocol, and written informed consent to participate was obtained.

### **Procedures**

Alexithymia was assessed with the 20-item Toronto Alexithymia Scale (TAS-20), an extensively validated self-report questionnaire.<sup>38-41</sup> The TAS-20 uses 1-5 Likert-type scales where 1 = strongly disagree, 2 = moderately disagree, 3 = neither agree nor disagree, 4 = moderately agree, and 5 = strongly agree. It consists of 3 subscales assessing difficulty identifying feelings (DIF), difficulty describing feelings (DDF), and externally oriented thinking (EOT). The TAS-20 total score has been used<sup>38,39</sup> to categorize participants as either non-alexithymic (scores from 20-51), borderline alexithymic (52-60), or alexithymic ( $\geq$  61). The Beck Depression Inventory-II<sup>42</sup> was also given to assess comorbid depressive symptoms.

Dreams were assessed with a previously validated Dream Questionnaire<sup>35</sup> on which

items were scored with the same scales used for the TAS-20. The Dream Questionnaire consists of 3 subscales: Nightmare Distress, Dream Recall, and belief in Dream Meaning.

iRBD patients were referred for at least one night of PSG recording in the hospital sleep clinic. Questionnaires were administered on the evening of the first night of recording. Patients and control participants reported themselves to be free of any medications known to influence sleep, vigilance, or motor activity for ≥ 1 week before the PSG evaluation. The recording montage included 2 EEG leads (C3/A2 and O2/A1), left and right electroculogram (EOG), and a chin electromyogram (EMG). All recordings were conducted by trained PSG technicians. Sleep stages 1-4 were scored by a highly proficient PSG technician according to a standard method,<sup>43</sup> and REM sleep was scored on the basis of EEG and EOG recordings using a method developed in our center.<sup>44,45</sup> Healthy controls and iRBD patients did not differ on any PSG variables other than lower REM sleep atonia in iRBD patients.

Between-groups comparisons on the TAS-20 total score, on age, and on years of education were investigated with 2  $\times$  2 ANOVA designs. A  $\chi^2$  test was used to compare groups on their alexithymia classification (not alexithymic, borderline, or alexithymic). MANOVAs were used to investigate differences on the 3 scales of the Dream Questionnaire and the 3 scales of the TAS-20 and interactions between group and sex. A 2  $\times$  2 ANCOVA with BDI-II score as covariate was used to control the effect of depressive symptoms on the TAS-20 total score. Pearson correlations were used to assess relationships between scales of the Dream Questionnaire and the TAS-20 variables separately for the 2 groups.

## **RESULTS**

iRBD patients obtained higher TAS-20 total scores (62.16  $\pm$  13.90) than did controls (52.84  $\pm$  7.62;  $F_{1,59}$  = 10.44, P < 0.001; see Figure 1). Men and women did not differ on TAS-20 total score (P > 0.43), and there was no interaction

between Group and Sex (P > 0.33). There were no significant between-group differences for years of education (P > 0.82) and age (P > 0.15). Twenty-one iRBD patients (65%) but only 6 controls (20%) attained the cutoff for alexithymia; 5 iRBD (16%) and 10 controls (33%) were borderline alexithymic; and 6 iRBD (18%) and 14 controls (47%) were not alexithymic ( $\chi^2 = 11.99$ , df = 2, P < 0.01). These 3 categories of iRBD patients did not differ on age, education, BDI-II score, psychiatric history, or intake of medication (P > 0.18).

When controlling for depressive symptoms, iRBD patients still had higher TAS-20 total scores (67.3  $\pm$  3.82) than did controls (49.84  $\pm$  3.82,  $F_{1,20}$  = 5.49, P < 0.05), and there was no difference between men and women (P > 0.80).

iRBD patients (19.74  $\pm$  6.51) scored higher than controls (12.67  $\pm$  3.15) on the DIF subscale of the TAS-20 ( $F_{1,56}$  = 12.79; P < 0.001, Figure 1), but did not differ on the DDF (P > 0.08) or EOT subscales (P > 0.99). iRBD patients scored higher (15.05  $\pm$  4.80) than controls (9.00  $\pm$  3.90) on the Nightmare Distress subscale of the Dream Questionnaire ( $F_{1,60}$  = 22.28; P < 0.001, Figure 2), but not on the Dream Recall (P > 0.97) or Dream Meaning (P > 0.62) subscales. There were again no Sex main effects or Group × Sex interactions for any of the 3 Dream Questionnaire subscales (all P > 0.32).

As shown in Table 1, the TAS-20 total and DIF scores correlated positively with Nightmare Distress score for both iRBD patients (r = 0.45, P < 0.05 and r = 0.54, P < 0.01, respectively) and controls (r = 0.39, P < 0.05; r = 0.48, P < 0.01).

## **DISCUSSION**

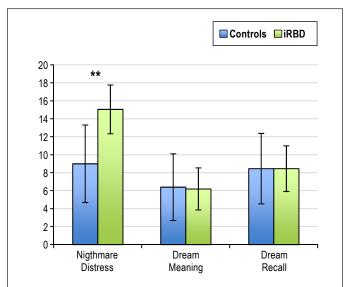
The results demonstrate substantially higher alexithymia scores and a higher proportion of individuals falling in the alexithymic category in a population of iRBD patients relative to matched healthy controls. This finding extends a previous finding that alexithymia scores are elevated in PD patients, <sup>11</sup> a population for whom RBD is frequently comorbid or a significant precursor condition. The difference in alexithymia scores was obtained even after controlling for a parallel difference in depressive symptoms, for which RBD patients may also be at risk. Alexithymia and depression are overlapping constructs, but the present

results support the idea that they are substantially independent.<sup>46</sup> Nonetheless, similar to other patients with comorbid alexithymia and depression, iRBD patients in our sample may have an elevated risk of developing psychiatric and health problems. <sup>19,26,47</sup> Further, the most severely alexithymic patients may have a greater incidence of all-cause mortality.<sup>28</sup>

Our sample of iRBD patients was particularly deficient on the TAS-20 subscale that assesses difficulties in identifying feelings (DIF), a subscale that previous work has shown is elevated for patients possessing a variety of sleep disorder symptoms. 33,35,48 Our patients, however, did not differ from controls on any objective measure of NREM

sleep. Thus, the alexithymia deficit in iRBD patients may not be a unique comorbid symptom but rather a more general symptom reflective of alterations in REM sleep, such as reduced and variable muscle atonia, strong intermittent muscle twitching, reduced heart rate variability, vivid, repetitive, and stereotypical dreams and nightmares, and overt dream-enactments. This is narrowly consistent with claims that alexithymia is secondary to sleep problems to the extent that such problems affect REM sleep. 49-51

The additional findings that our iRBD patients suffer from more nightmare distress than control participants and that their levels of nightmare distress are correlated with severity on the alexithymia total and DIF scales suggest that the alexithymia deficit for these patients may be comorbid even more specifically with the occurrence of nightmares. The higher frequency of nightmare distress reported by our iRBD patients extends results from previous studies in which dream content has been found to be more violent in RBD, <sup>2,5,6</sup> although the latter finding has been contested for a sample of medicated patients.<sup>4</sup>



**Figure 2**—Comparisons between iRBD patients and matched control participants on the Dream Questionnaire subscales. \*\*P < 0.01.

**Table 1**—Correlations between TAS-20 total score, TAS-20 subscales (DIF, DFF, EOT) and Dream Questionnaire subscales (ND, DM, DR)

		iRBD group						
		TAS-20	DIF	DDF	EOT	ND	DM	DR
Control group	TAS-20	-	0.94**	0.88**	0.76**	0.45*	0.15	0.24
	DIF	0.68**	-	0.85**	0.53**	0.54**	0.05	0.34
	DDF	0.75**	0.33	-	0.48**	0.33	0.32	0.34
	EOT	0.75**	0.19	0.35*	-	0.29	-0.01	-0.08
	ND	0.39*	0.48**	0.28	0.13	-	-0.11	0.14
	DM	-0.04	0.16	-0.14	-0.09	0.39*	-	0.30
	DR	0.13	0.27	0.06	-0.05	0.34	0.39*	_

Coefficients for iRBD patients are shown in the upper right half and values for controls in the lower left half of the table. iRBD, idiopathic REM sleep behaviour disorder; DIF, difficulty identifying feelings; DFF, difficulty distinguishing feelings; EOT, externally oriented thinking; ND, nightmare distress; DM, dream meaning; DR, dream recall.  $^*P < 0.05$ ;  $^{**}P < 0.01$ .

Confidence in our findings is bolstered by the fact that previous studies have shown high DIF scores to be associated with high nightmare distress in both clinical and non-clinical populations and for men and women considered separately.<sup>35</sup> In fact, this DIF-nightmare distress association also replicates the findings of two previous studies demonstrating that both TAS-20 total and DIF scores correlate with nightmare frequency<sup>31</sup> and other nightmare symptoms.<sup>33</sup>

We previously suggested that this relationship between DIF and nightmare distress reflects either a more generalized disposition to react negatively to events ("affect distress" personality style) or a deficit in emotion regulation processes. An affect distress explanation of the present findings seems unlikely because iRBD patients display a contrary style of passivity and non-aggressiveness.<sup>2,4,8</sup> Rather, an emotional dysregulation explanation provides a better fit with some iRBD symptoms. To illustrate, emotional dysregulation has been put forward as a factor that leads alexithymic individuals to display excessive, unexpected emotional outbursts when awake and bizarre emotional dreams and nightmares when asleep. 15 In the case of iRBD patients, these outbursts may be restricted to REM sleep, although sporadic emotional outbursts during wakefulness have not to our knowledge been assessed for this population. An emotion dysregulation explanation is also broadly consistent with evidence of higher anxiety and depression levels in child RBD patients<sup>9</sup> and in PD patients<sup>10</sup> and with accumulating evidence that dreaming and REM sleep play roles in the crossnight regulation of negative emotions. 52,53

This deficit may be related to dysfunctions in prefrontal regions that regulate emotional expression during various cognitive tasks. For example, alexithymic individuals display a disadvantageous pattern of risky decision-making on the Iowa Gambling Task that is suggestive of a failure to learn from feedback, i.e., poor consolidation of emotional experiences.54,55 Such deficits are consistent with neuroanatomic changes in emotion-relevant brain areas identified for alexithymic individuals, such as reduced gray matter volume in the amygdala, anterior insula, and anterior cingulate cortex (ACC). 56,57 A similar deficit on the Iowa gambling task was found for idiopathic RBD patients,58 although neuroanatomic studies have proved variable with respect to brain changes associated with RBD. Some report anomalies in frontal or limbic areas<sup>59-61</sup> consistent with the cognitive findings; others do not. 62,63 However, dysfunction in prefrontal regions known to down-regulate amygdala-related emotions—such as medial prefrontal cortex or ACC-may well explain the preponderance of dysphoric dreams and nightmare distress among RBD patients.64 Nonetheless, the possibility that elevated alexithymia in iRBD patients is due to an emotional deficit linked preferentially to prefrontal regulatory regions, though a viable hypothesis, requires additional neuroanatomic study.

Yet another, related, explanation for the present findings is that the emotional symptoms of iRBD patients are due to dysautonomia.<sup>65</sup> iRBD patients show autonomic denervation leading to lower heart rate variability (HRV) compared to controls during wakefulness<sup>66,67</sup> and during REM sleep.<sup>68</sup> Similarly, PD patients with RBD show reduced HRV during wakefulness.<sup>69</sup> A cardiac 123I-mIBG scintigraphy study has further demonstrated loss of cardiac sympathetic innervation in RBD.<sup>70</sup>

An impoverishment of autonomic afferent pathways leading to diminution of affect-relevant information may have an impact on how well patients are able to identify their own feelings. This would account for their low scores on aggressiveness<sup>2</sup> and their high scores on the DIF alexithymia subscale but also, by still unidentified mechanisms, for their distressing nightmares. Like iRBD, reduced HRV has also been found for idiopathic nightmares<sup>71</sup> and other affective disturbances such as anxiety, depression, and worry, and worry, for which nightmares are often also comorbid.

In sum, that iRBD patients demonstrate elevated levels of alexithymia and nightmare distress and that these symptoms are correlated may be explained by several factors. An affect distress personality style is less likely than the possibility of a dysregulation of emotions, but an underlying dysautonomia is a possibility consistent with much research and requiring additional study. Dysautonomia may reduce afferent sources of feeling and thus explain the specific difficulty in identifying feelings. However, it remains unknown how nightmares are related to the various explanations of high alexithymia in RBD. The dysautonomia explanation raises the novel possibility that degrees of autonomic afferent reduction may also be implicated in nightmares that are not related to iRBD, e.g., posttraumatic or idiopathic nightmares. The increases in dream vividness and nightmares that have been reported to precede the appearance of RBD symptoms by many years suggests that nightmares associated with alexithymia—DIF scores in particular—may be important potential markers for enhancing diagnoses of prodromal RBD and even Parkinson disease. Similar markers have been sought in disturbed olfaction, constipation, autonomic dysfunction, and other symptoms.<sup>76</sup>

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