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# Comparison between an automatic and a visual scoring method of the chin muscle tone during rapid eye movement sleep



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#### ABSTRACT

*Objective:* To compare two different methods, one visual and the other automatic, for the quantification of rapid eye movement (REM) sleep without atonia (RSWA) in the diagnosis of REM sleep behavior disorder (RBD).

*Methods:* Seventy-four RBD patients (mean age,  $62.14 \pm 9.67$  years) and 75 normal controls (mean age,  $61.04 \pm 12.13$  years) underwent one night video-polysomnographic recording. The chin electromyogram (EMG) during REM sleep was analyzed by means of a previously published visual method quantifying the percentage of 30 s epochs scored as tonic (abnormal,  $\ge 30\%$ ) and that of 2 s mini-epochs containing phasic EMG events (abnormal,  $\ge 15\%$ ). For the computer quantitative analysis we used the automatic scoring algorithm known as the atonia index (abnormal, < 0.8). The percentage correct classification, sensitivity, specificity, and Cohen kappa were calculated.

*Results:* The atonia index correctly classified 82.6% of subjects, similar to the percentage of correct classifications with individual components of the visual analysis (83.2% each for tonic and phasic), and the combined visual parameters (85.9%). The sensitivity and specificity of automatic analysis (84% and 81%) was similar to the combined visual analysis (89% and 83%). The correlation coefficient between the automatic atonia index and the percentage of visual tonic EMG was high (r = -0.886, P < 0.00001), with moderately high correlation with the percentage of phasic EMG (r = -0.690, P < 0.00001). The agreement between atonia index and the visual parameters (individual or combined) was approximately 85% with Cohen's kappa, ranging from 0.638 to 0.693.

*Conclusion:* Sensitivity, specificity, and correct classifications were high with both methods. Moreover, there was general agreement between methods, with Cohen's kappa values in the 'good' range. Given the considerable practical advantages of automatic quantification of REM atonia, automatic quantification may be a useful alternative to visual scoring methods in otherwise uncomplicated polysomnograms. © 2014 Elsevier B.V. All rights reserved.

## 1. Introduction

Rapid eye movement (REM) sleep behavior disorder (RBD) is a parasomnia characterized by absent or greatly diminished physiological atonia during REM sleep and by dream-enacting behavior [1]. REM sleep without atonia (RSWA) is the required polysomnographic (PSG) hallmark for the diagnosis of RBD; however, the current International Sleep Disorders criteria do not specify cut-offs for atonia. In order to overcome the lack of clear cut-off values for RSWA, during the last 20 years several methods have been proposed for its scoring and quantification, with both visual and automatic computer-based approaches [2]. Among them, only very few have been validated in more than one study, and so reliability and replicability remain unclear. Moreover, most methods to quantitate REM atonia rely on manual visual scoring – this requires specialized expertise (not available in all centers) and involves considerable demands of time and cost, limiting broad applicability.

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The aim of this study was to analyze the results obtained with two different methods, one visual [3,4] and the other automatic [5,6], in order to assess their reciprocal agreement on the same dataset.

## 2. Methods

#### 2.1. Subjects

Eighty patients (62 males and 18 females; mean age,  $62.7 \pm 9.5$  years) who were diagnosed with RBD between 1993 and 2006 at the Sleep Disorders Clinic at the Hôpital du Sacré-Coeur de Montréal were recruited, as well as 80 normal controls (62 males and 18 females; mean age,  $61.3 \pm 12.0$  years). The detailed description of their clinical features and of the recruiting process can be found in an earlier paper [4]. The protocol was approved by the University Hospital ethics committee and patients signed a consent form before their participation.

#### 2.2. Polysomnographic recordings

All participants underwent one night of video-PSG monitoring in the sleep laboratory including a full electroencephalogram (EEG) montage to rule out epilepsy. For both RBD patients and normal controls, sleep was recorded according to the Rechtschaffen and Kales' criteria [7] with standard EEG leads (C3–A2, O2–A1), bilateral electro-oculogram (EOG), and chin electromyogram (EMG) recordings. Respiration was monitored using an oro-nasal thermistor or a nasal cannula and with thoracic and abdominal strain gauges, whereas blood oxygen saturation (SaO<sub>2</sub>) was continuously recorded by cutaneous finger pulse oxymeter. None of the patients or controls showed an apnea index (number of apneas per hour of sleep) >10 or an index of respiratory events (apneas + hypopneas) >15.

As RBD patients usually have RSWA, REM sleep cannot be scored on the basis of chin EMG as in the method of Rechtschaffen and Kales [8]. Therefore, the occurrence of the first REM was used to determine the onset of REM sleep. Termination of REM sleep period was identified either by the occurrence of a specific EEG feature of another sleep stage (K complex, sleep spindle) or EEG sign of arousal, or by the absence of rapid eye movements for three consecutive minutes.

Eleven out of 160 recordings (six RBD patients and five controls) were excluded because of technical reasons that did not allow a correct transformation of the signal into European Data Format [9] for the subsequent automatic computer analysis. Thus the following procedures were carried out on 74 recordings of RBD patients (56 males and 18 females; mean age,  $62.1 \pm 9.67$  years) and 75 controls (57 males and 18 females; mean age,  $61.0 \pm 12.13$  years).

#### 2.3. Visual quantitative analysis of the chin EMG during REM sleep

This analysis was carried out following previously published criteria, adapted to 30 s epochs [3,4]. Epochs were scored as tonic or atonic depending on whether chin EMG activity was present for more or less than 50% of the epoch duration. EMG activity was defined by amplitude of the chin EMG signal of at least twice that of the background or >10  $\mu$ V. Phasic chin EMG density was scored from the submental EMG activity and represented the percentage of 2 s mini-epochs containing EMG events lasting 0.1–10 s, with an amplitude exceeding four times the amplitude of the background EMG activity. The phasic EMG activity can be scored on an epoch, which is scored either as atonic or as tonic. Based on previous findings, REM sleep chin EMG levels were considered to be

abnormal when tonic chin EMG density was  $\ge 30\%$  and/or phasic chin EMG density was  $\ge 15\%$  [4].

#### 2.4. Automatic quantitative analysis of the chin EMG during REM sleep

For the computer quantitative analysis of the submentalis muscle EMG activity we used an established automatic scoring algorithm [5,6,10]. The submentalis muscle EMG signal was digitally band-pass filtered at 10–100 Hz, with a notch filter at 60 Hz and rectified. Subsequently, each sleep epoch included in the analysis was divided into thirty 1 s mini-epochs. The average amplitude of the rectified submentalis muscle EMG signal was then obtained for each mini-epoch. After a noise reduction procedure [6], the values of the submentalis muscle EMG signal amplitude in each 1 s mini-epoch were used to compute the percentage of values in the following 20 amplitude (amp) classes (expressed in  $\mu$ V):  $amp \leq 1, 1 < amp \leq 2, \dots, 18 < amp \leq 19, amp > 19$ . Muscle atonia is expected to be reflected by high values of the first class  $(amp \leq 1)$  whereas phasic and tonic activations are expected to increase the value of the other classes [5,6]. As proposed in previous studies, an index summarizing in a single value the degree of preponderance of the first class was used in REM sleep:

Atonia index =  $amp \le 1/(100 - 1 < amp \le 2)$ .

Mathematically, this index can vary from 0 (absence of miniepochs with amp  $\leq 1$ ), i.e. complete absence of EMG atonia, to 1 (all mini-epochs with amp  $\leq 1$ ) or stable EMG atonia in the epoch. The REM sleep atonia index correlates significantly with the percentage of epochs of REM sleep without atonia detected by the method by Lapierre and Montplaisir [3,5].

The algorithm was run blinded to the results of the manual scoring procedure. According to previous findings, REM sleep chin EMG levels were considered to be abnormal when atonia index was <0.8 or <0.9 [6]; however, other cut-off values were also used in this study in order to find the optimum level, from 0.7 to 0.9, with 0.05 steps.

## 2.5. Statistical analysis

Specificity, sensitivity, positive predictive value (PPV), negative predictive values (NPV), and correct classifications of RBD were computed for the atonia index <0.8 and <0.9, tonic chin EMG density  $\ge$  30%, phasic chin EMG density  $\ge$  15%, and chin EMG density  $\ge$  30% or phasic chin EMG density  $\ge$  15%. The accuracy of the different parameters to discriminate RBD cases from normal cases was also evaluated using receiver operating characteristic (ROC) curve analysis and the calculation of the area under the curve (AUC). The linear correlation coefficients between the different pairs of visual and automatic parameters were computed, and the relative data scatterplots were drawn, as well as the corresponding least squares lines. Additionally, the extent of agreement between the different pairs of visual and automatic parameters was guantified by means of Cohen's kappa coefficient [11], which is a standard measure of inter-rater agreement for categorical data which can be interpreted as a measure of agreement that exists beyond the amount expected by chance alone [11]. A value of one indicates perfect agreement; a value of zero indicates that agreement is no better than chance. Although there is no universal concordance on the meaning of the magnitude of the Cohen's kappa value, the strength of agreement can be interpreted as follows [12]: <0.20 'poor', 0.21-0.40 'fair', 0.41-0.60 'moderate', 0.61-0.80 'good', and 0.81-1.00 'very good.'

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