Mandible behaviour interpretation during wakefulness, sleep and sleep-disordered breathing

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Keywords
breathing effort, mandible study, sleep study, visual analysis

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Accepted in revised form 15 May 2014; received 6 November 2013

DOI: 10.1111/jsr.12180

SUMMARY
The mandible movement (MM) signal provides information on mandible activity. It can be read visually to assess sleep–wake state and respiratory events. This study aimed to assess (1) the training of independent scorers to recognize the signal specificities; (2) intrascorer reproducibility and (3) interscorer variability. MM was collected in the mid-sagittal plane of the face of 40 patients. The typical MM was extracted and classified into seven distinct pattern classes: active wakefulness (AW), quiet wakefulness or quiet sleep (QW/S), sleep snoring (SS), sleep obstructive events (OAH), sleep mixed apnea (MA), respiratory related arousal (RERA) and sleep central events (CAH). Four scorers were trained; their diagnostic capacities were assessed on two reading sessions. The intra- and interscorer agreements were assessed using Cohen’s $\kappa$. Intrascorer reproducibility for the two sessions ranged from 0.68 [95% confidence interval (CI): 0.59–0.77] to 0.88 (95% CI: 0.82–0.94), while the between-scorer agreement amounted to 0.68 (95% CI: 0.65–0.71) and 0.74 (95% CI: 0.72–0.77), respectively. The overall accuracy of the scorers was 75.2% (range: 72.4–80.7%). CAH MMs were the most difficult to discern (overall accuracy 65.6%). For the two sessions, the recognition rate of abnormal respiratory events (OAH, CAH, MA and RERA) was excellent: the interscorer mean agreement was 90.7% (Cohen’s $\kappa$: 0.83; 95% CI: 0.79–0.88). The discrimination of OAH, CAH, MA characteristics was good, with an interscorer agreement of 80.8% (Cohen’s $\kappa$: 0.65; 95% CI: 0.62–0.68). Visual analysis of isolated MMs can successfully diagnose sleep–wake state, normal and abnormal respiration and recognize the presence of respiratory effort.

INTRODUCTION
Mouth opening is a common observation during sleep in patients suffering from sleep-disordered breathing (SDB) (Miyamoto et al., 1999). The conjunction of sleep (and sleep stage) and neuro-anatomical factors of the upper respiratory airway will contribute in varying proportions to the occurrence of the obstructive event. Many factors influence the upper airway (UA) resistance and the occurrence of SDB. Inability to prevent UA obstruction during sleep is a feature of obstructive sleep apnea patients, inducing a cyclical phenomenon with persistent breathing effort. In the complex mechanisms leading to UA obstruction, the inferior jaw position (and linked structures including pharyngeal dilator muscles) is both influenced by and participates in pharyngeal patency. Mandible lowering during sleep is thought to be related to UA patency, as it is associated with both reduced cross-sectional area of the lumen and increased collapsibility of the UA (secondary to an inferior–posterior movement of the mandible) (Isono et al., 2004; Kuna and Remmers, 1985), and may contribute to sleep-related breathing abnormalities.

The hypothetical mechanism explaining the mandible behaviour during sleep is postulated as follows.

In normal conditions, the central respiratory drive commands the phasic inspiratory contraction of the pharyngeal dilator muscles [mainly the genioglossus muscle (GG) but
also the geniohyoid and mylohyoid) about 50–100 ms before diaphragmatic contraction (Van Lunteren and Dick, 1992). The electrical phasic activity (inspiratory bursts induced by the respiratory drive) of the GG and its nerve (the XIIth cranial nerve, called the hypoglossal nerve) occurs in a tonic permanent state, and both activities are reduced during sleep (Dobe et al., 1985; Sauerland and Harper, 1976).

One of the pathophysiological elements contributing to initiate a respiratory obstructive event during sleep is a reduction or a suppression of the activity of the dilator muscle of the oropharynx. Indeed, the GG activity decreases prior to the respiratory obstructive event. Conversely, during an UA obstructive event, the GG electromyographic activity increases (simultaneously with repeated breathing effort), reaching the highest amplitude before the termination of the obstructive event (Sauerland et al., 1981). During an obstructive apnea, the progressively increasing inspiratory contractions of the dilator muscle could induce a tongue protrusion which could cause a small inspiratory lowering of the mandible.

As the upper airway is occluded or suboccluded, an increased negative pressure into the thoracic cavity will induce, through the thoracic inlet, a longitudinal traction on it. This is called the thoracic tug (Van de Graaff, 1988), which is thought to generate a passive lowering of the mandible and added cycling respiratory movements during obstructive apnea or hypopnea (Lugaresi et al., 1979; Van de Graaff, 1991), an oscillating pattern that is lacking in central respiratory events.

At the end of the obstructive respiratory event, contraction of the GG is necessary to restore upper airway permeability. The contraction of masticatory muscles, among them the masseter, after apnea has been assumed to stabilize the mandible (Hollowel and Suratt, 1991; Miyamoto et al., 1998; Yoshida, 1998). Without this stabilization, the GG contraction could generate a posterior movement of the mandible. The sequence of muscle activity at the end of the respiratory event promotes upper airway patency. The resultant closing movement of the mandible at the end of the apnea has been described (Hollowel and Suratt, 1991; Miyamoto et al., 1998) and previously named ‘salient mandible movement’ (SMM) by our team (Maury et al., 2013).

Globally, during obstructive sleep apnea, passive gradual lowering of the mandible occurs, characterized by oscillating movement linked to respiratory effort (which is absent in case of central apnea) and ending by a closing movement of the mandible. This salient mandible movement, as a marker of arousal, contributed to hypopnea detection in an automated analysis combined with oxygen saturation (SaO₂) and nasal airflow (Maury et al., 2013). Moreover, during active wakefulness, this regular pattern is lacking. As the anarchic jaw movements have been used in an automated analysis to discriminate sleep–wake periods (Senny et al., 2012), we postulated that all these typical mandible patterns could be used to visually recognize active wakefulness, quiet wakefulness and quiet sleep, snoring, respiratory events and the presence of an associated effort (obstructive and mixed events).

**MATERIAL AND METHODS**

**Recordings**

Overnight nocturnal polysomnography (PSG) was performed in 40 consecutive patients who accepted that oesophageal pressure and mandibular movement (JAWAC) recordings were added to the classical procedure used with the recommended setting of sensors for sleep-disordered breathing measurements. They were admitted to the Sleep/Wake Center of the University Hospital of Liége, between February 2007 and October 2010. All PSGs were analysed visually.

A polygraph N7000 (EMBLA Medcare, Denver, CO, USA) processed 12 parameters: electroencephalogram (EEG) derivations F4-M1, C4-M1, 02-M1, with backup electrodes placed at F3, C3, O1 and M2; electro-oculogram (EOG) derivations E1-M2 and E2-M2, electromyogram (EMG) for the chin and two electromyograms for the legs, airflow signal, recorded by nasal cannula-pressure transducer (NC-PT), respiratory effort signal by two inductive plethysmography belts, SaO₂, body position (POS) and electrocardiogram (ECG). To these nine signals were added sleep snoring (SS) recorded by a microphone fixed in the left side of the neck, oesophageal pressure (Pes) and mandible movements (JAWAC) recordings.

For Pes, the zero, plus and minus 10 hectopascals, on a U-shaped tube, half-full of water was checked for a GaelTec EP sensor EL1 (GaelTec Utilities, Isle of Skye, Scotland, UK). This sensor was then passed through the nose and swallowed up to the middle third of the patient’s oesophagus and connected to a free DC entry of the polygraph.

JAWAC recordings were performed by a distance-meter based on the principle of magnetometry (Jawsens®; NOM-ICS, Liége, Belgium). The sensors were composed of two coils and capacitors, each embedded in a small cylinder (7 mm diameter; 25 mm main axis). They were disposed parallel to each other, perpendicular to the midline of the face (Fig. 1). They were connected to an electronic circuit by two cables. The electronic circuit converted distance into voltage. The signal was digitalized with a sampling frequency of 10 Hz, transmitted by cable to the polygraph. Physical calibration (maximal closing and opening of the mouth) was made by displaying the mandibular movement tracing on the same screen as the full PSG.

**Definitions**

Wake and sleep stages were scored according to the American Academy of Sleep Medicine (AASM) rules (Iber et al., 2007). Sleep was scored in four stages [N1, N2, N3—the non-rapid eye movement (REM) stages (NREM)—and REM].

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Apneas were defined by a drop of nasal airflow of 80% during at least 10 s.

Hypopneas were defined by a drop of nasal airflow of 30% during at least 10 s, with either a drop of SaO2 equal to or higher than 3% or an arousal (Berry et al., 2012). Obstructive hypopneas were noted when these events were associated with an increase in negative Pes. Central hypopneas were characterized by a parallel drop in negative Pes. Any situation between these two extremes was not admitted in the qualification procedure.

Respiratory-related arousals were defined by the presence of gradually increasing negative Pes for at least 10 s, culminating in a moment of microarousal, without any apnea or hypopnea (AASM, 2005).

Single sleep snoring was associated with a regular respiratory noise (SS), emitted in the upper airway, and modulated by uninterrupted breathing cycles.

**PSG patterns database**

A sleep specialist scored the different stages of sleep and wakefulness on the polysomnography. Wakefulness was subdivided into active wakefulness and quiet wakefulness. All SDB events were then identified and marked on the polysomnography as references. A technician selected 338 epochs corresponding to seven distinct classes of interest: active wakefulness (AW), quiet wakefulness or quiet sleep (QW/S), sleep snoring (SS), sleep obstructive events (OAH), sleep mixed apnea (MA), respiratory-related arousal (RERA) and sleep central events (CAH). Signals without artefacts (NAF, SaO2, belts and Pes) were selected randomly. The JAWAC tracings from these epochs were isolated, extracted and entered into charts of the same duration and amplitude (100 s × 10 mm). Each of them was captured according to the presence or absence of breathing event, the nature of breathing events, the time of night and the sleep or wake stages in which they occurred. Due to the specificities of the diagnoses, it was not possible to reach a balanced sampling of the seven classes for each recording. When present, all charts of JAWAC were centred on SDB. A training database was created of 199 charts representative of the seven different classes and used for training sessions. A formal description of the main characteristics was also given for each class (Fig. 2).

The behaviour of the mandible recorded in the sagittal plane during the various SDB events was relatively stereotyped and contrasted with what was found when patients were awake and active (speaking, chewing, swallowing...). It also contrasted with the silent tracing observed when subjects were awake, inactive or asleep without SDB.

**Scoring sessions**

The remaining set of 139 charts was stored in another database and devoid of any diagnostic indication. Charts could then be selected randomly from this database for recognition sessions by trained scorers.

None of the staff members who selected the events and formatted the charts were subsequently involved in the recognition procedure, and none of the four scorers were involved in the analysis of data.

Four scorers—a physiotherapist, a pulmonologist and two psychologists—volunteered to participate in the recognition sessions. There were two reading sessions. Scorers had been instructed previously during two training sessions and received a copy of the atlas. Then, each JAWAC pattern was provided without contextual pattern or patient information.

The 199 charts were presented in a random order to each scorer for identification. Twenty charts were related to active wakefulness, 20 to quiet wakefulness or to a sleep stage without SDB, 20 to single snoring, 19 to RERA, 20 to obstructive events, 20 to mixed events and 20 to central events. There was an equal proportion of patterns issued from NREM sleep and REM sleep. Therefore, five obstructive apneas occurred in REM and five in NREM sleep. The same criterion was applied for the selection of obstructive hypopnea, central apnea and central hypopnea. The number of events in each class was unknown to the scorers.

During the reading sessions, each scorer had to be alone. He/she could refer to the atlas and disposed of unlimited time, provided each session be completed during the same day. The JAWAC tracings were presented randomly on a computer screen and each scorer had to assign them one of the seven pattern classes, using the numeric keypad of the computer. Changes during the session were allowed. The two readings sessions were at least 2 weeks apart.

The Institutional Review Board of the University Hospital and Hospital of Liège approved the study protocol and all participants provided informed consent. Data were kept anonymous and confidential.
Statistics

Accuracy and reliability in the recognition of SDB events through visual scoring of JAWAC patterns were evaluated. Accuracy was measured by the percentages of correct answers performed by each scorer during each of the two reading sessions. To compare the accuracy of two different scorers, McNemar’s test was used.

Reliability was defined as the ability of each scorer to reproduce his/her diagnosis (whether correct or not) in the two reading sessions. It was assessed by the percentage of equal assessments and by Cohen’s $\kappa$ coefficient of agreement with associated 95% confidence interval (95% CI).

To assess the legibility of each diagnostic class, namely its difficulty to be identified by the scorers, the mean accuracy (%) of the four scorers over the two reading sessions was computed for each class. Diagnostic classes were then determined by increasing legibility.

To calculate the recognition rates of abnormal respiratory events, the seven patterns were grouped into two categories: (1) AW, QW/S, SS and (2) O, C, M and RERA. Further, to assess the discrimination of O/C/M characteristics, another grouping was used: (1) AW, QW/S, SS and RERA and (2) O, C and M events. Statistical analyses were performed for both sessions separately.

Results were considered significant at the 5% critical level ($P < 0.05$). All calculations were performed with SAS (version 9.3 for Windows) and S-PLUS (version 8.1) statistical packages.

RESULTS

Patient characteristics

Of the forty patients who participated in the study, 25 suffered from obstructive sleep apnea syndrome, six patients from insomnia and anxiety or depression, six from central sleep apnea syndrome, two from restless legs syndrome and one from enuresis. Table 1 summarizes the main anthropometric and sleep data of the patients.

Global analysis

Diagnostic accuracy

The accuracy of the scorers, i.e. the percentage of correctly diagnosed events during the two sessions, varied between
Central events were poorly recognized and mainly mistaken with QW/S (13% were classified erroneously as QW/S) and M (11% were classified as M) events and less with O (9% were classified as O).

**Within- and between-scorer agreement**

As seen in Table 4, the intrascorer agreement was good, with Cohen’s $\kappa$ coefficients ranging from 0.68 to 0.88. Diagnostic decision remained fairly stable. The between-scorer agreement amounted to 0.71 (95% CI: 0.69–0.73) for both recognition sessions.

**Qualitative discrimination**

For the identification of abnormal respiratory events (O/C/M and RERA versus AW, QWS, S), the diagnostic accuracy of the four scorers was equal to 90.1 and 91.2%, with an average of 90.7%. The interscorer agreement was 0.83 (95% CI: 0.79–0.88).

Further, regarding the discrimination of O, C, M characteristics, the diagnostic accuracy of both sessions amounted, respectively, to 78.4 and 83.1%, with an average of 80.8%. The interscorer agreement was 0.65 (95% CI: 0.62–0.68).

**DISCUSSION**

We inventoried the different mandible behavior patterns associated with active wakefulness, quiet wakefulness and quiet sleep, sleep snoring and disordered breathing for visual analysis procedure by four scorers.

The study showed a diagnostic accuracy of about 75.2%, a within-scorer agreement ranging from 0.68 to 0.88 and a between-scorer agreement of 0.71 for both recognition sessions. RERA was the easiest JAWAC tracing to recognize (with an accuracy of 87.5%) while central apnea was the most difficult (65.6% correct answers).

At both sessions, misclassification appeared for central apnea and mixed apneas, on one hand, and central apnea...
and QS/W on the other hand (these events include a same stable signal). This was probably linked to the difficulty to define the limits of the event. The MM in central apnea was partially similar to QW/S, as showed in Fig. 2. Moreover, mixed apneas were partly central and partly obstructive. Breathing effort, pointed as the oscillatory movement, was present in obstructive apnea or hypopnea. A similar pattern occurred in sleep snoring and RERA. The breathing effort was an important parameter to detect. Commonly in sleep studies, it is far from easy to measure. We considered that the effort increased progressively, as a continuum, from sleep snoring to RERA, and finally obstructive hypopnea and apnea. For this study, the threshold of abnormal breathing effort was placed at oscillatory movement of 1 mm based on the analysis of previous sleep studies performed before 2007. This method allowed us to assess satisfactorily the presence or absence of obstructive events. Sleep snoring was characterized by regularly oscillating moves of the mandible, higher than 0.5 mm. RERA was similar to snoring, except that oscillations were interrupted by some salient moves of the mandible. Indeed, for the two sessions, interscorer mean agreement for the recognition of abnormal respiratory events was very good (O/C/M and RERA; accuracy was 90.7%), as well as the discrimination of O/C/M characteristics (global diagnostic accuracy was 80.8%). There was little discrepancy in the interpretation, particularly for the recognition of abnormal respiratory events (Cohen’s κ coefficient 0.83; 95% CI: 0.79–0.88). The possibility of teaching independent scorers to recognize signal specificities is confirmed. Despite some confusion, the results of the two scoring sessions were not significantly different. The scorers remained quite stable in their interpretation from one session to the other.

Nevertheless, mandibular oscillations could be influenced by the sleep position, the sleep stage, associated respiratory comorbidities or other patient characteristics. This is probably the reason for some confusion in the identification of the seven patterns when the criterion of the definition is borderline (for example, quiet sleep and sleep snoring around 0.5 mm).

The sensors for measuring mandibular movements are easy to set up (one sensor on the forehead and the other under the chin) and non-invasive. They provide a unique signal to address three key features in the context of a screening device for SDB; the first is the ability to detect abnormal breathing effort during sleep. It is postulated here that a threshold for abnormal effort is reached once mandibular oscillations appear in the frequency domain of breathing. A relationship between the amplitude of the oscillations and the amplitude of the inspiratory efforts can also be appreciated. Oesophageal pressure measurement is the reference method to measure respiratory effort (Baydur et al., 1982). This is an invasive method which requires the place of an oesophageal catheter, and contraindications exist. Practically, this method is limited to physiological research. In our study, oesophageal pressure was used in the selection procedure of SDB to constitute the database reliably.

The second key feature is the ability to separate active wakefulness from quiet wakefulness or sleep without SDB. It can be used as an actimeter, and may exclude from the analysis any blatant period of wakefulness. This was studied in an automated manner with very interesting results (Senny et al., 2012). This is an important contribution to the index

### Table 3 Table of score by class (percentage) for the four scorers and the two sessions combined

<table>
<thead>
<tr>
<th>Polysomnography</th>
<th>AW</th>
<th>QW/S</th>
<th>SS</th>
<th>OAH</th>
<th>M</th>
<th>CAH</th>
<th>RERA</th>
</tr>
</thead>
<tbody>
<tr>
<td>Scoring</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>AW</td>
<td>81.3</td>
<td>14.4</td>
<td>0.0</td>
<td>3.8</td>
<td>0.0</td>
<td>0.63</td>
<td>0.0</td>
</tr>
<tr>
<td>QW/S</td>
<td>0.0</td>
<td>67.5</td>
<td>15.6</td>
<td>0.0</td>
<td>0.0</td>
<td>13.1</td>
<td>0.0</td>
</tr>
<tr>
<td>SS</td>
<td>1.3</td>
<td>3.1</td>
<td>81.9</td>
<td>11.3</td>
<td>0.0</td>
<td>0.0</td>
<td>1.3</td>
</tr>
<tr>
<td>OAH</td>
<td>10.6</td>
<td>3.1</td>
<td>1.9</td>
<td>70.0</td>
<td>8.8</td>
<td>9.4</td>
<td>9.2</td>
</tr>
<tr>
<td>M</td>
<td>0.0</td>
<td>5.0</td>
<td>0.6</td>
<td>6.9</td>
<td>73.1</td>
<td>10.6</td>
<td>1.3</td>
</tr>
<tr>
<td>CAH</td>
<td>6.9</td>
<td>3.8</td>
<td>0.0</td>
<td>5.0</td>
<td>13.8</td>
<td>65.6</td>
<td>0.7</td>
</tr>
<tr>
<td>RERA</td>
<td>0.0</td>
<td>3.1</td>
<td>0.0</td>
<td>3.1</td>
<td>4.4</td>
<td>0.6</td>
<td>87.5</td>
</tr>
</tbody>
</table>

AW, active wakefulness; QW/S, quiet wakefulness + normal sleep; SS, sleep snoring; OAH, obstructive apnea + hypopnea; MA, mixed apnea; RERA, respiratory event-related arousal; CAH, central apnea + hypopnea.

For each class, correct diagnoses (%) are in bold.

### Table 4 Global analysis. Within- and between-scorer agreement (n = 139 patterns)

<table>
<thead>
<tr>
<th>Polysomnography</th>
<th>Percentage concordance</th>
<th>Kappa</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Within-scorer agreement</td>
<td>Scorer 1</td>
<td>82.7</td>
<td>0.80</td>
</tr>
<tr>
<td></td>
<td>Scorer 2</td>
<td>89.9</td>
<td>0.88</td>
</tr>
<tr>
<td></td>
<td>Scorer 3</td>
<td>79.1</td>
<td>0.76</td>
</tr>
<tr>
<td></td>
<td>Scorer 4</td>
<td>72.7</td>
<td>0.68</td>
</tr>
<tr>
<td>Between-scorer agreement</td>
<td>Session 1</td>
<td>72.7</td>
<td>0.68</td>
</tr>
<tr>
<td></td>
<td>Session 2</td>
<td>77.2</td>
<td>0.74</td>
</tr>
</tbody>
</table>

K, confidence interval.
accuracy provided by portable devices. Indeed, the denominator used in the calculation of the AHI is the total sleep time.

The third key feature is that the movements of the mandible are particular for each event. A specific pattern is present in case of respiratory arousal, called the SMM. When compared to the acceleration and augmentation provided in the EEG and chin EMG, the salient movements associated with the arousals represent a simple signal which could probably reflect a more specific activation of brain stem centres.

By reading a PSG the scorer has to integrate the characteristics of the signals, and can find much information in other concomitant registered signals. In the study conditions, 139 windows were extracted from PSG of 40 patients and isolated, presented in a randomized database. Artefacts were not considered. The decision of the scorer was based on this unique signal, without contextual or clinical information. Moreover, the aim of this study was not to assess the relationship between mandibular oscillations and breathing effort: the presence of breathing effort was qualitatively noted.

Our study has a number of limitations: the number of charts was limited to 338, of which 139 were used for scoring, and there were only four scorers. The charts used for this study were selected randomly, and this selection was based on the quality of the signals. Therefore, due to this selection and the diagnostic specificities, to have a balanced sampling of respiratory events was not possible. The extracts used for training sessions were different from those used in the scoring sessions, but issued from the same 40 patients. We did not examine the effects of sleep stages, of positions adopted during sleep or gender effects on the recognition of SDB. We have not studied either abnormal conditions such as bruxism or sleep-talking, which must necessarily interfere with the JAWAC signal, or activations of other origins than breathing disorders (movement time, periodic limb movements). By the approach used here, we studied event by event and provided a separate detailed analysis of each pattern in the extracted windows. This could reduce the global information (for example, the trend of mouth-opening during the night). Finally, the measurement of swings in oesophageal pressure by an oesophageal catheter is the reference method of respiratory effort measurement (Baydur et al., 1982). None the less, the catheter presence in the upper airways could influence their properties and collapsibility (Pépin et al., 1995).

In summary, the diagnosis of events, as well as the intrasession reliability, provided very good results. The authors concluded that the visual analysis of the isolated MM was useful to assess the sleep–wake state and the occurrence of abnormal respiration and to recognize the presence of respiratory effort.

ACKNOWLEDGEMENTS

Gisèle Maury was supported by a PhD grant from the Mont Godinne Foundation.

CONFLICT OF INTEREST

No conflicts of interest declared.

AUTHOR CONTRIBUTIONS

The clinical study was conducted by GM, FS, LC and RP. AA and LS performed the statistical analysis. All the authors contributed to the writing and the revision of the manuscript.

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