Chapter 5
Mandible behaviour interpretation during wakefulness, sleep and sleep-disordered breathing

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SUMMARY

Mandible movement (MM) signal provides information on the mandible activity. It can be read visually to assess sleep/wake state and respiratory events. The study aimed to assess (1) the training of independent scorers to recognize the signal specificities; (2) the intra-scorer reproducibility, and (3) the inter-scorer variability.

MM was collected in the mid-sagittal plane of the face of 40 patients. The typical MM was extracted and classified into 7 distinct pattern classes: active wakefulness (AW), quiet wakefulness or quiet sleep (QW/S), sleep snoring (SS), sleep obstructive events (OAH), sleep mixed apnoea (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 inter-scorer agreements were assessed using the Cohen kappa (κ).

The intra-scorer reproducibility for the two sessions ranged between 0.68 (95% CI : 0.59-0.77) and 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 inter-scorer mean agreement was 90.7% (κ : 0.83; 95% CI [0.79-0.88]). The discrimination

of OAH, CAH, MA characteristics was good with an inter-scorer agreement of 80.8 % (κ : 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)[1-3] and is different than mandibular posture during sleep in healthy adults.[4] 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.

Globally, during obstructive sleep apnoea, a passive gradual lowering of the mandible occurs, characterized by repeated downward mandible excursions linked to respiratory effort (which is absent in case of central apnoea) and ending by a closing movement of the mandible. This salient mandible movement, as a marker of arousal, contributed to hypopnoea detection in an automated analysis combined with SpO_2 and nasal airflow (see chapter 2).[5] 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 (see chapter 3)[6], 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).

The hypothetical mechanism explaining the mandible behaviour during sleep has been described in chapter 1.

MATERIAL AND METHODS

1. 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 Centre of the University Hospital of Liège, between February 2007 and October 2010. All PSGs were analysed visually.

A polygraph N7000 (EMBLA Medcare, Denver, 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, oxygen saturation (SpO_2), body position (POS) and electrocardiogram (ECG). To these 9 signals were added sleep snoring (SS) recorded by a microphone fixed in the left side of the neck, oesophageal pressure (Pes) and mandible movements (Jawsens®) recordings.

2. Definitions

Wake and sleep stages were scored according to the AASM rules[7]. Sleep was scored in 4 stages (N1, N2, N3 – the non-REM stages (NREM) - and REM).

Definitions used for respiratory events and single snoring are given in Annex 1. 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 in between these two extremes was not admitted in the qualification procedure.

Respiratory related arousals (RERA) were defined by the presence of gradually increasing negative Pes, for at least 10 seconds, culminating in a moment of arousal, without any apnoea or hypopnoea.[8]

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

3. 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 7 distinct classes of interest: active wakefulness (AW), quiet wakefulness or quiet sleep (QW/S), sleep snoring (SS), sleep obstructive events (OAH), sleep mixed apnoea (MA), respiratory related arousal (RERA) and sleep central events (CAH). Signals without artefacts (NAF, SpO_2, belts and Pes) were randomly selected. The Jawac tracings from these epochs were isolated, extracted and presented into charts of the same duration and amplitude (100 seconds x 10 mm). Each of them was captured according to the presence or absence of breathing event, the nature of breathing events, the time of the night, 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 7 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 7 different classes and used for training

sessions. A formal description of the main characteristics was also given for each class (Figure 1).
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.

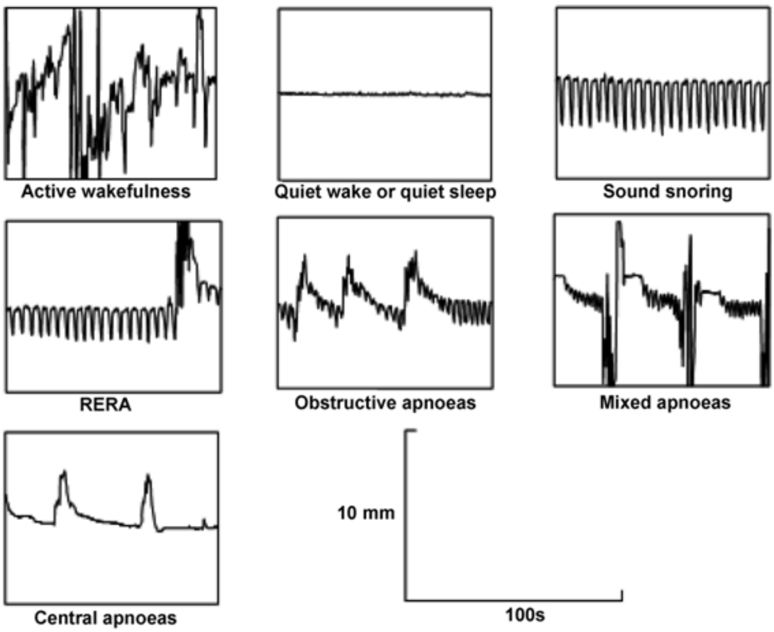

Figure 1 : Examples of the different patterns submitted to identification by the four raters.

All charts were presented with the same duration and the same amplitude. Active wakefulness corresponded to an irregular tracing with erratic, ample moves of the mandible. Quiet wakefulness or quiet sleep, without SDB was associated with a flat tracing with small artefacts due to electronic noise, which was less than 0.5 mm. 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. This abruption was associated on the polysomnography with an arousal. After the abruption, regularly oscillating moves reappeared. Obstructive respiratory events are characterized by mandible downward excursions (in an oscillating pattern) which progressed in two successive segments: firstly, a series of openings and closings of the mouth superimposed on a lowering of the mandible – this segment corresponded to the apnoea itself, and secondly, a salient movement of the mandible. Mixed apneoas presented as an identical sequence, except that during apneoas, the oscillating moves were preceded by a short segment of a few seconds without any discernible oscillations, even if the mandible could change its position. Central respiratory events corresponded to a non-oscillating pattern of the mandible, regularly interrupted by an abruption, which could be more or less oscillating.

4. Scoring sessions

The remaining set of 139 charts was stored in another database and devoid of any diagnostic indication. Charts could then be randomly selected 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 4 scorers were involved in the analysis of data.

Four scorers – a physiotherapist, a pulmonologist and 2 psychologists – volunteered for participating to the recognition sessions. There were two reading sessions. Scorers had been previously instructed 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. There were 20 charts 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, 20 to central events. There was an equal proportion of patterns issued from NREM-sleep and REM sleep. Therefore, five obstructive apnoeas occurred in REM and 5 in NREM-sleep. The same criterion was applied for the selection of obstructive hypopnoea, central apnoea and central hypopnoea.

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 7 pattern classes, using the numeric keypad of the computer. Changes during the session were allowed. The two readings sessions were at least two weeks apart.

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

5. 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 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 sorted out by increasing legibility.

To calculate the recognition rates of abnormal respiratory events, the 7 patterns were grouped in 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 done for both sessions separately.

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

RESULTS

1. Patient's characteristics

Of the forty patients who participated in the study, 25 suffered from obstructive sleep apnoea syndrome, 6 patients from insomnia and anxiety or depression, 6 from central sleep apnoea syndrome, 2 from restless legs syndrome, and 1 from enuresis.

Table1 summarizes the main anthropometric and sleep data of the patients

Table 1 : Anthropometric and polysomnographic data of the 40 patients			
	Mean ±SD	**Minimum**	**Maximum**
Age, *y*	49 ± 11.6	25	77
Women, *n(%)*	5 (12.5)		
Body mass index, kg/m^2	29.8 ± 5.9	19	43
TDT, *minutes*	572 ± 80.7	392	733
TST, *minutes*	444 ± 94.1	257	664
Apnoea-hypopnoea index, *n/1h*	41.4 ± 33.4	2	136
RERA index, *n/1h*	2.3 ± 2.8	0	109
Obstructive events, *n*	131 ± 221	0	944
Central events, *n*	36.0 ± 87.5	0	370
Mixed events, *n*	41.1 ± 136.6	0	713
Hypopnoea, *n*	107± 81.3	1	291

2. Global analysis

2.1. Diagnostic accuracy

The accuracy of the scorers, that is the percentage of correctly diagnosed events during the two sessions, varied between 72.4% and 80.7%, with a mean of 75.2%.

There was no significant improvement between the two reading sessions.

Table 2 displays the percentage of correct recognitions (in comparison with PSG) for the different classes of events, obtained by the four scorers, during the two sessions (S1 and S2) and the global mean.

Table 2. Diagnostic accuracy of each scorer for each class of event and scoring session (S1 and S2)

	Scorer 1			Scorer 2			Scorer 3			Scorer 4			Global Mean SM
Class	**S1**	**S2**	**Mean**	**S1**	**S2**	**Mean**	**S1**	**S2**	**Mean**	**S1**	**S2**	**Mean**	
AW	80.0	85.0	82.5	85.0	85.0	85.0	90.0	80.0	85.0	70.0	75.0	72.5	**81.3**
QW/S	70.0	70.0	70.0	65.0	75.0	70.0	60.0	65.0	62.5	65.0	70.0	67.5	**67.5**
SS	80.0	85.0	82.5	80.0	85.0	82.5	85.0	90.0	87.5	70.0	80.0	75.0	**81.9**
OAH	70.0	75.0	72.5	70.0	80.0	75.0	65.0	70.0	67.5	55.0	75.0	65.0	**70.0**
M A	70.0	75.0	72.5	85.0	85.0	85.0	55.0	70.0	62.5	60.0	85.0	72.5	**73.1**
CAH	70.0	70.0	70.0	70.0	75.0	72.5	50.0	60.0	55.0	65.0	65.0	65.0	**65.6**
RERA	94.7	89.5	92.1	100	89.5	94.8	84.2	89.5	86.9	73.7	78.9	76.3	**87.5**
Mean	76.3	78.4	**77.4**	79.1	82.0	**80.7**	69.8	74.8	**72.4**	65.5	75.5	**70.5**	**75.2**

SM is the global mean for both scoring sessions of the four scorers. Active Wakefulness (AW); Quiet Wakefulness + Normal sleep (QW/S); Sleep Snoring (SS); Obstructive Apnoea + Hypopnoea (OAH), Mixed Apnoea (MA); Respiratory Event-Related Arousal (RERA); Central Apnoea + Hypopnoea (CAH).

2.2. *Legibility of diagnostic classes*

As seen in Table 2, the legibility of each diagnostic class (based on the two reading sessions), namely its ability to be correctly identified by the scorers varied greatly among classes. By increasing order of magnitude, the legibility of the classes ranked as follows: central apnoea (65.6%), quiet wakefulness or quiet sleep (67.5%), obstructive apnoea (70.0%), mixed apnoea (73.5%), active wakefulness (81.3%), sleep snoring (81.9%) and RERA (87.5%). The overall percentages of correct recognitions for each class (scorers and sessions combined) are given in Table 3.

Central events were poorly recognized and mainly mistaken with QW/S (13% were erroneously classified as QW/S) and M (11 % were classified as M) events and less with O (9 % were classified as O).

Table 3 : Table of score by class (percentage) for the four scorers and the 2 sessions								
	Correct diagnosis (%) for the four scorers and the 2 sessions combined							
	Polysomnography							
Scoring	Classes	AW	QW/S	SS	OAH	M	CAH	RERA
	AW	**81.3**	14.4	0.0	3.8	0.0	0.63	0.0
	QW/S	0.0	**67.5**	15.6	0.0	0.0	13.1	0.0
	SS	1.3	3.1	**81.9**	11.3	0.0	0.0	1.3
	OAH	10.6	3.1	1.9	**70.0**	8.8	9.4	9.2
	M	0.0	5.0	0.6	6.9	**73.1**	10.6	1.3
	CAH	6.9	3.8	0.0	5.0	13.8	**65.6**	0.7
	RERA	0.0	3.1	0.0	3.1	4.4	0.6	**87.5**

Correct diagnosis (%) is in bold

2.3. Within- and between-scorer agreement

As seen in Table 4, the intra-scorer agreement was good, Cohen 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.

Table 4 : Global analysis. Within and between-scorer agreement (n =139 patterns)			
	Percent concordance	**Kappa**	**95% CI**
Within-scorer agreement			
Scorer 1	82.7	0.80	0.73 - 0.87
Scorer 2	89.9	0.88	0.82 - 0.94
Scorer 3	79.1	0.76	0.68 - 0.83
Scorer 4	72.7	0.68	0.59 - 0.77
Between-scorer agreement			
Session 1	72.7	0.68	0.65 - 0.71
Session 2	77.7	0.74	0.72- 0.77

3. Qualitative discrimination

For the identification of abnormal respiratory events (O/C/M and RERA versus AW, QW/S, S), the diagnostic accuracy of the 4 scorers was equal to 90.1% and 91.2 %, with an average of 90.7%. The inter-scorer agreement was 0.83 (95% CI : 0.79-0.88).
Further, as far as the discrimination of O, C, M characteristics was concerned, the diagnostic accuracy of both sessions amounted respectively to 78.4% and 83.1%, with an average of 80.8%. The inter-scorer agreement was 0.65 (95% CI : 0.62-0.68).

DISCUSSION

We inventoried the different mandible behaviour 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 apnoea was the most difficult (65.6% correct answers).

At both sessions, misclassification appeared for central apnoea and mixed apnoeas on one hand and central apnoea 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 apnoea was partially similar to QW/S, as showed in Figure 1. Moreover, mixed apnoeas were partly central and partly obstructive. Breathing effort, pointed as the oscillating pattern movement was present in obstructive apnoea or hypopnoea. 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 apnoea. For this study, the threshold of abnormal breathing effort was placed at a difference between minimum and maximum level of amplitude of the movement (oscillating pattern) 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 event. 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 2 sessions, inter-scorer 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 kappa coefficient 0.83; 95% CI: 0.79-0.88). The possibility to teach independent scorers to recognize the signal specificities is confirmed. Despite some confusion, the results of the 2 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 co-morbidities or other patient characteristics. This is probably the reason of some confusion in the identification of the seven patterns when the criterion of the definition is border-line (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 one is the ability to detect abnormal breathing effort during sleep. It is posited 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 (see chapter 6). Oesophageal pressure measurement is the reference method to measure respiratory effort.[9] This is an invasive method which requires the place of an oesophageal catheter and contraindications exists. Practically, this method is limited to physiological research. In our study, oesophageal pressure was used in the selection procedure of the sleep disordered breathing to constitute reliably the database.

The second one 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 way with very interesting results (see chapter 3).[6] This is an important contribution to the indexes accuracy provided by portable device. 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 brainstem 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 appreciated.

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 4 scorers. The charts used for this study were randomly selected 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 body position adopted during sleep neither the 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 (i.e., periodic limb movements). By the approach used here, we studied event by event and provided a separate detailed analysis of each patterns in 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.[9] Nonetheless, the catheter presence in the upper airway could influence their properties and collapsibility.[10]

CONCLUSION

The diagnosis of events, as well as the intra and inter-scorer reliability provided very good results. We concluded that the visual analysis of the isolated MM was useful to assess sleep/wake state, the occurrence of abnormal respiration and to recognize the presence of respiratory effort.

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