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A Modest Celebration of Collaboration

Leslie C. Dort, DDS, Diplomate, ABDSM, Editor-in-Chief *Journal of Dental Sleep Medicine*

Calgary, Alberta, Canada

This issue of the *Journal of Dental Sleep Medicine* (JDSM) is cause for modest celebration. The October 2015 issue marks the final issue of the first full year of publication. Any new journal is faced with many challenges. A new journal has no presence in the scientific world. It takes time to be listed in the major databases such as PubMed and therefore potential contributors may not be aware of our journal if they are not AADSM members. Academic authors often need to publish their research in the most well established journals possible in order to maintain and advance their academic status. A fledgling journal is rarely the first choice. Funding sources for research are diminishing as are the funds themselves. This is particularly true in dental sleep medicine where funds are almost non-existent. The number of manuscripts produced in this field is therefore limited compared to many other clinical areas of sleep medicine. To successfully meet these challenges requires the collaboration of many individuals with a variety of roles. The managing editorial staff is a critical component of journal success. They manage to make each issue the best it can be: often stretching deadlines and working to the last moment before publication.

The journal has published some original research, a number of relevant substantial review articles, case studies, editorials and special articles. This issue has the most original articles to date as well as other manuscripts. This growth despite the challenges that exist is encouraging for the future of dental sleep medicine. Thank you to the senior researchers who have contributed reviews and in particular to the researchers who have contributed primary research manuscripts. These articles and reviews give readers information on the state of the art of dental sleep medicine. They also increase the continuing education opportunities for those readers who take advance of the continuing education credits available (see <http://www.jdsm.org/CE.aspx>). Bravo also to the many clinicians who have ventured out of their comfort zones to write a case report. Often these case reports are a clinician's first encounter with the world of academic publishing and the many tedious steps involved. Case studies are our direct contact with clinical practice.

It is notable that many of the manuscripts published in the journal are collaborative in nature.¹⁻⁴ These reflect the need for collaboration among health care providers to give the most benefit to those with sleep disorders. The JDSM will continue to publish material to inform all those who are collaborating in the research and treatment of sleep disordered breathing. This is the future of dental sleep medicine.

CITATION

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DISCLOSURE STATEMENT

Dr. Dort is Editor-in-Chief of *Journal of Dental Sleep Medicine*.

How Close Can Single-Channel EMG Data Come to PSG Scoring of Rhythmic Masticatory Muscle Activity?

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STUDY OBJECTIVES: Assessment of jaw-muscle activity during sleep is needed to establish a definite diagnosis of sleep bruxism (SB). Multichannel polysomnographic (PSG) studies are the gold standard (GS) but are unfortunately not readily available, so single-channel electromyographic (EMG) devices have been developed. This study attempted to evaluate an EMG algorithm for single-channel EMG recordings in comparison with the outcome from PSG recordings.

METHODS: PSG data from 20 participants with different frequency of jaw-muscle EMG activity were analyzed with the GS algorithm, including previously published criteria for EMG analyses and contrasted to two different algorithms: one based on a signal recognition (SR) algorithm and the other based on a moving average (MA) estimation method, which is characterized by a comparison of the EMG amplitude to the estimated background level, and applying the rules for detection of rhythmic masticatory muscle activity (RMMA).

RESULTS: The highest correlation coefficients ($r = 0.96$) were obtained between the GS and the MA algorithm; however, there were no significant differences in the absolute numbers of EMG bursts or episodes between the SR and MA algorithms and GS during sleep. However, both algorithms significantly overestimated the EMG bursts and episodes when awakenings during sleep were included in the analyses. There were no significant differences between muscles or side ($p > 0.06$).

CONCLUSIONS: This study strongly indicates that a MA algorithm may be useful for analysis of EMG activity during sleep but with recognition of the potential overestimation of EMG bursts and episodes due to transient awakenings.

KEYWORDS: sleep bruxism, polysomnography, single-channel electromyographic device, rhythmic masticatory muscle activity, electromyography

CITATION: Dreyer P, Yachida W, Huynh N, Lavigne GJ, Haugland M, Svensson P, Castrillon EE. How close can single-channel EMG data come to PSG scoring of rhythmic masticatory muscle activity? *Journal of Dental Sleep Medicine* 2015;2(4):147–156.

BruXism is a repetitive jaw-muscle activity characterized by clenching or grinding of the teeth and/or by bracing or thrusting of the mandible that could happen during awake and sleep periods.¹ Although sleep bruxism (SB) is not a life-threatening disorder, it can affect the patient's quality of life, especially because of dental problems such as tooth wear, damage or fractures of tooth structures or dental restorations, pain in the orofacial region, and tension-type headache.^{2,3} SB is often suggested as a cause of temporomandibular disorders (TMD) orofacial pain and headache, but scientific evidence does not support a simple link between SB and craniofacial pain conditions.^{4–7} A set of clinical research diagnostic criteria (RDC) for SB were proposed in 1996⁸ and has since been considered the gold standard (GS) in many clinical studies of SB. The criteria were re-validated by Rompré et al.⁹ and used for classification of a subgroup of bruxism patients with a higher risk of pain. Still, the relation between SB and many adverse clinical symptoms and signs is unclear, and the factors causing SB and the physiological mechanisms behind SB are still being discussed.³

One reason for the relative paucity of research data on SB may be that the RDC includes use of full polysomnographic (PSG) and audio-video recordings that can be done in sleep laboratory or at home (type 1 and 2 type recording system, respectively). While PSG and audio-video recordings provide highly accurate scoring of rhythmic masticatory muscle

activity (RMMA),⁸ it is also a costly and time-consuming procedure, which is difficult to do for most clinicians and even for most research groups. It also requires training of the person scoring the data, and still there is some level of discrepancy between how the data are scored by different scorers and from different laboratories.¹⁰

Due to the costs and efforts associated with PSG, there has been great interest in scoring RMMA with more simple and portable devices that can be used over long periods of time in the patients' own home.^{11–18} These recording systems can be type 3 (with 3–4 physiological variables such as muscle, cardiac, and respiration) or simplified type 4 with one EMG channel. Recently, a portable single-channel EMG device was introduced for recordings of jaw-muscle activity during sleep based on a signal recognition (SR) algorithm.^{15,16} However, single-channel EMG devices are notoriously known to record too many "true" events of RMMA,¹⁸ and the absence of audio-video also complicate the scoring, resulting in an overestimation of 25%.¹⁹

To overcome this problem, the original Grindcare SR algorithm was updated. The modified algorithm is characterized by a comparison of the EMG amplitude to the estimated background level (moving average, MA) and applying the rules for detection of RMMA activity described by Lavigne and collaborators⁸ for quantification of RMMA.

Thus, the overall aim of the present study was to determine how close a simple algorithm of a single-channel EMG could come to PSG and audio-video recording in terms of quantification of RMMA during sleep. Specifically, there were three main objectives: (1) investigate the performance of two signal analysis methods, the SR and the MA vs. GS (RMMA); (2) investigate if there was a significant difference in the quantification of RMMA from EMG activity recorded from masseter or temporalis muscles or between left/right body sides; and (3) determine the impact of awake periods before, during, or after sleep on the quantification of RMMA frequency.

MATERIALS AND METHODS

Subjects

Data from a total of 20 individuals (12 women/8 men) were selected for this retrospective analysis (mean age: 26.8 ± 1.41 years). The data used in the present study had all been recorded previously, as part of a standard PSG examination at University of Montreal and this accordingly to ethical standards of Sacre Coeur Hospital research center. Six datasets were selected from healthy control subjects with no history or physical signs of bruxism. The other 14 sets were patients with a positive history and physical signs of bruxism but otherwise no neurological or sleep disorders.^{9,19,20} This combination of individuals with and without history and physical signs of bruxism was chosen to reflect the continuum of RMMA during sleep, i.e., to avoid ceiling or floor effects in the detection of RMMA with the different EMG algorithms.

Polysomnographic Data

The PSG data included electroencephalography (EEG: 7 channels), electrooculography (EOG: 2 channels), EMG from the masticatory muscles (right anterior temporalis muscle TAR, left anterior temporalis muscle TAL, right masseter muscle MAR, and left masseter muscle MAL), EMG from the leg muscles (anterior tibialis muscle), electrocardiogram (ECG: 3 channels), audio and video recordings. EMG data was low-pass filtered at 70 Hz and sampled at a 256 samples/second rate.

The PSG data from all 20 individuals had been scored for sleep stage and for RMMA activity according to standard and published routines at University of Montreal.^{8,9} This scoring was done manually, based on all the available information described above, as well as audio and video recordings. The compiled information from the PSG scoring (“RMMA burst” and “RMMA episodes”) was considered to be the GS in the present study and was provided in addition to the raw EMG data. Data was translated from Steallate (Natus) format and saved in the European Data Format for Biosignals (EDF). This enabled us to read the signals into Matlab (Mathworks Inc) for analysis. The GS information on RMMA was then compared to the outcome from 2 different EMG algorithms (SR; MA), which were applied to the 4 masticatory EMG channels. The PSG data from all the 20 individuals were scored and an “RMMA index” was calculated, which is the total number of RMMA episodes activity divided by sleep duration.

Due to the fact that it is normal to have transient awakenings during sleep the data was also classified as EMG

activity during sleep (“sleep period”) and EMG activity during sleep plus awakenings (“sleep + awakenings period”). The GS criteria to score RMMA excluded the transient awaken periods using the sleep staging.

EMG Algorithms

Signal Recognition Algorithm

The first EMG algorithm was an approximation of the SR algorithm used in the original Grindcare device (Medotech A/S, Herlev, Denmark).¹⁵ This algorithm was developed to work along with contingent electrical stimulation, which imposes some constraints in terms of EMG measurement and stimulation through the same electrode. Very briefly, this EMG algorithm compares the amplitude of the EMG to a threshold level, which is set to 20% of the maximum EMG during a clench to about 60% of the maximum voluntary contraction (MVC). Setup of the threshold level is done every time the device is mounted before sleep, during which the user is required to produce a bite force to approximately 60% MVC. An “SR grind” is detected, counted in the log-file and registered when the amplitude of the EMG signal has been above the threshold for more than 0.1 s.

For the purpose of the contingent electrical stimulation, it was thought to be important that the electrical stimulation was delivered as soon as possible after detection of the EMG activity. This means that it is not possible for the device to wait and see if an EMG burst is indeed part of an episode of rhythmic EMG activity, a long (tonic) EMG burst, or merely a single brief EMG event. The electrical stimulation is delivered as soon as any EMG activity is detected, and for the next 1 second, the stimulation interferes with the EMG recording. This means it is not possible to measure the duration of bursts of EMG activity and counting of bursts is difficult, due to the 1-second “blind period.” When 1 second has passed, the EMG signal is monitored again, and if above threshold, a new EMG event can be detected. This has the implication that if a long burst of EMG appears in the signal (i.e., > 1 second), several events may be detected and counted.

It was, however, not possible to do a full simulation of the Grindcare SR algorithm due to differences in filtering and sample rate between standard PSG equipment and Grindcare. Grindcare samples at a much higher rate (2,000 samples/s compared to 256 samples/sec) and removes the low frequency content in the signal, in order to reduce possible interference of low-frequency noise (e.g., 50/60 Hz noise). Moreover, Grindcare uses a fast Fourier transform-based, proprietary, method for further reducing the influence of noise and detection of pure EMG signals. This part of the EMG analysis was not possible to simulate with the available PSG data. However, in the sleep laboratory measures have been taken to reduce interfering noise, and operators continuously ensure that there is good electrode contact and that the signals look good, reducing the need for the noise reduction techniques used with Grindcare. Unfortunately, the datasets provided from the sleep laboratory did not include EMG data, wherein the subject was instructed to clench to 60% MVC. There was, however, a part of the standard “biocalibration,” where individuals were instructed to “clench teeth strongly together 3 times.”

Moving Average Algorithm

This MA algorithm for detection of EMG bursts used a dynamic method for estimation of background EMG noise. Bursts of EMG that exceeded the background noise with more than 3 times the background amplitude were detected. Furthermore, the rules for classification of EMG as RMMA as described in Lavigne et al.⁸ were applied: An EMG burst must be ≥ 0.25 sec in duration, an EMG burst can be phasic (< 2 sec) or tonic (> 2 sec), and an EMG burst must be part of an EMG episode to be counted; an EMG episode consists of either ≥ 3 phasic EMG bursts and/or one or more tonic EMG bursts; an EMG episode consists of EMG bursts < 3 sec apart. The EMG bursts and episodes detected using this MA algorithm will be called “MA burst” and “MA episode.”

Both the SR and MA algorithms were applied to the EMG recordings from MAL, MAR, TAL, TAR in all 20 individuals. Analyses were done by Morten Haugland in Denmark blind to SB diagnostic or RMMA episodes frequency of data collected in Montreal.

Statistics

Kolmogorov-Smirnov tests indicated the majority of parameters were not normally distributed, and therefore nonparametric descriptive statistics (median and interquartile ranges) were applied except for the RMMA index. Friedman repeated measures analysis of variance on ranks (ANOVA) was used to test the EMG data. We compared the number of SR grinds, MA bursts, and RMMA bursts per hour of sleep. Moreover, MA episodes and RMMA episodes (GS) were also compared. PSG data were also compared between algorithms (3 levels: SR, MA GS), masticatory muscles (4 levels: MAL, MAR, TAL, TAR) and between PSG sleep EMG or sleep + awakening EMG (2 levels). Tukey post hoc tests were used to compensate for multiple comparisons and the Dunnett method when appropriate. The RMMA-index was compared between bruxers and non-bruxers with the use of an unpaired t-test. Pearson product moment correlation tests were used to test for associations between the outcomes from the 2 algorithms versus the GS criteria. This was done for all EMG channels (MAL, MAR, TAL, TAR) and for PSG data containing both sleep and sleep including awakening. $P < 0.05$ was considered statistically significant. Moreover, levels of agreement between outcomes from the 2 algorithms versus GS criteria were tested using Bland-Altman test.

RESULTS

Polysomnographic Data

Figure 1 shows an example of a full-night PSG recording with focus on the right masseter muscle. The awaken periods have been excluded with the use of the sleep staging and the analyses of the EMG activity shown according to the GS criteria, SR and MA algorithms. From this example it is evident that the SR algorithm due to its inherent nature overestimates the number of SR grinds compared to the number of MA episodes detected by the MA algorithm and the RMMA episodes of the GS.

The GS scoring showed that the RMMA index for the included data ranged from 0.3 episodes/h of sleep to 13.5

episodes/h of sleep (Table 1). The mean RMMA index in the designated sleep bruxers was 4.9 ± 3.5 episodes per hour of sleep compared to 1.4 ± 0.8 episodes per hour of sleep in non-bruxers (unpaired t-test: $p = 0.03$).

Comparison between Algorithms

The ANOVA indicated no significant difference between the number of SR grinds, MA bursts and RMMA bursts detected with the SR, MA algorithms, and the GS during sleep period ($p > 0.40$, Figure 2A). However, both algorithm SR and MA detected significantly more SR grinds/MA bursts than the RMMA bursts of GS during sleep + awakening period ($p < 0.02$, Tukey: $p < 0.05$, Figure 2B).

The ANOVA for episodes did not include the number of SR algorithm analyses results but only contrasted the MA episodes and the RMMA episodes of GS: There were no significant differences on the episodes between these 2 algorithms ($p = 0.19$, Figure 3A) during sleep, but significantly higher MA episodes during sleep + awakenings period ($p < 0.001$, Tukey: $p < 0.05$, Figure 3B).

Comparison between Muscles

There were no significant differences on the number of SR grinds/MA bursts and on the number of MA episodes detected between the 4 masticatory muscles ($p > 0.06$) (Figure 2 and 3).

Comparison between Sleep and Sleep Including Awakening

The quantitative analyses clearly indicated that the number of SR grinds, MA bursts (Figure 2), and MA episodes (Figure 3) during the sleep period were significantly lower than the during sleep + awakening period ($p < 0.001$).

Correlations and Levels of Agreement between Different Algorithms

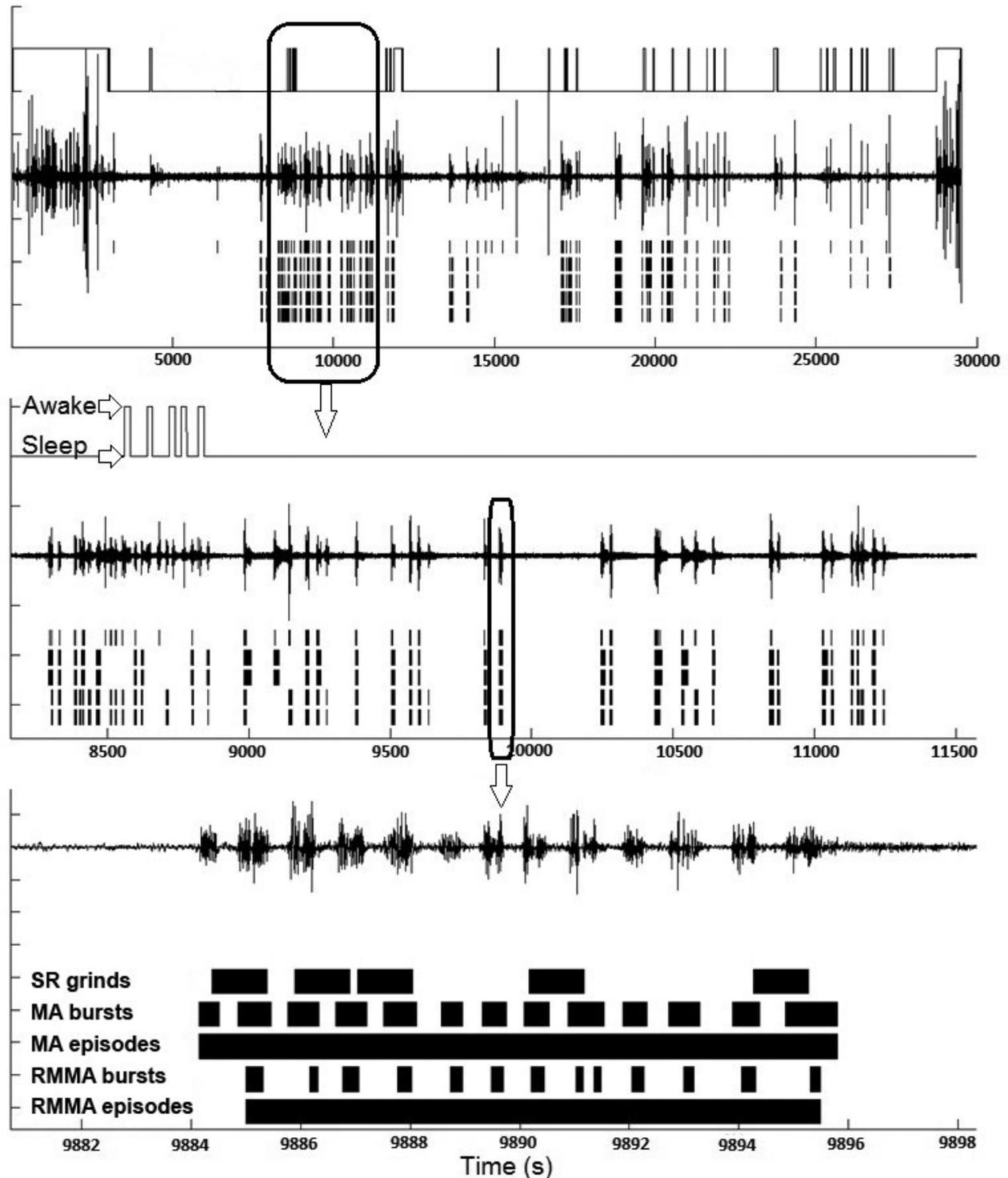
Figure 4 shows the correlation plots between the RMMA (GS) bursts and SR grinds using the GS criteria and SR algorithm for the 4 masticatory muscles recorded only during sleep. In a similar way, Figure 5 and 6 shows the plots of the data of RMMA and MA algorithm for the four different muscles recorded only during sleep (bursts and episodes, respectively; Table 2, 3).

There were significant positive correlations between the GS and MA algorithm in the different muscles both during sleep periods and during sleep + awakening periods with exception of the right side of anterior temporalis (TAR) during sleep + awakening periods ($r > 0.46$, $p < 0.05$). However, there were no significant correlations ($r \leq 0.13$) between the GS and SR algorithm in any muscles ($p > 0.60$) (Table 2, 3).

Moreover, levels of agreement between outcomes from the 2 algorithms versus GS criteria are shown in Table 4.

DISCUSSION

The Grindcare device has, until now, scored muscle activity using SR algorithm, as defined above.¹⁵ The proposed MA algorithm uses a novel adaptive threshold to determine the occurrence of MA bursts and the rules for detection of bursts and episodes activity were applied in a similar way

Figure 1—Illustration of PSG data from a single subject (#4).

Top panel shows data from a full night, including the standard calibration exercises that were performed in the beginning and end of the recording session. Middle and lower panels show selection of the same data at different timescales. Top trace indicates whether the subject was asleep or not, based on the results from the off-line sleep staging. Middle trace is the raw EMG signal (in this case the right masseter). The bars below the EMG trace show the SR grinds using the SR algorithm, MA bursts and MA episodes using the MA algorithm, and finally RMMA bursts and RMMA episodes scored manually based on the full PSG data set and RMMA (gold standard). PSG, polysomnography; SR, signal recognition; MA, moving average; RMMA, rhythmic masticatory muscle activity.

as when RMMA is scored manually following GS criteria. The algorithms were compared with correlation coefficients and differences between the total amounts of EMG activity detected with the different methods.

Bursts and episodes detected with the MA algorithm in general correlated well with the manual scoring based on the full PSG data. The datasets scored in PSG analysis as having "high" RMMA frequency, in most cases also came out as being

Table 1—Characteristics of PSG data from 20 individuals.

| Individuals | Total Duration (h) | Staged Sleep Duration (h) | Staged Awake Duration (h) | RMMA Bursts | RMMA Bursts/h | RMMA Episodes | RMMA Episodes/h (Total Duration) | RMMA Episodes/h (Sleep Duration) | RMMA Index | Category |
|-------------|--------------------|---------------------------|---------------------------|-------------|---------------|---------------|----------------------------------|----------------------------------|------------|----------|
| 1 | 7.6 | 6.8 | 0.8 | 179 | 23.7 | 20 | 2.6 | 2.9 | 3.0 | Mild |
| 2 | 8.0 | 7.0 | 1.0 | 288 | 36.1 | 40 | 5.0 | 5.7 | 5.8 | High |
| 3 | 7.0 | 6.6 | 0.4 | 383 | 54.6 | 64 | 9.1 | 9.6 | 9.4 | High |
| 4 | 7.1 | 6.9 | 0.3 | 625 | 87.7 | 93 | 13.1 | 13.5 | 13.5 | High |
| 5 | 8.6 | 8.5 | 0.1 | 40 | 4.6 | 12 | 1.4 | 1.4 | 1.3 | Normal |
| 6 | 8.1 | 6.5 | 1.6 | 169 | 20.9 | 31 | 3.8 | 4.8 | 4.7 | High |
| 7 | 8.8 | 8.2 | 0.5 | 44 | 5.0 | 11 | 1.3 | 1.3 | 1.3 | Normal |
| 8 | 8.3 | 7.4 | 0.9 | 51 | 6.2 | 10 | 1.2 | 1.4 | 1.4 | Normal |
| 9 | 4.0 | 3.8 | 0.2 | 272 | 68.6 | 28 | 7.1 | 7.3 | 7.4 | High |
| 10 | 8.4 | 8.1 | 0.3 | 301 | 36.0 | 44 | 5.3 | 5.4 | 5.3 | High |
| 11 | 7.4 | 7.4 | 0.0 | 258 | 34.9 | 38 | 5.1 | 5.1 | 4.8 | High |
| 12 | 7.7 | 7.7 | 0.1 | 226 | 29.2 | 32 | 4.1 | 4.2 | 4.2 | High |
| 13 | 7.2 | 6.7 | 0.4 | 89 | 12.4 | 22 | 3.1 | 3.3 | 3.3 | Mild |
| 14 | 6.6 | 5.8 | 0.8 | 52 | 7.9 | 12 | 1.8 | 2.1 | 2.1 | Mild |
| 15 | 7.7 | 7.3 | 0.4 | 41 | 5.4 | 10 | 1.3 | 1.4 | < 2 | Control |
| 16 | 7.9 | 7.8 | 0.1 | 81 | 10.3 | 18 | 2.3 | 2.3 | < 2 | Control |
| 17 | 8.3 | 7.0 | 1.3 | 40 | 4.8 | 10 | 1.2 | 1.4 | < 2 | Control |
| 18 | 6.5 | 6.0 | 0.5 | 14 | 2.1 | 4 | 0.6 | 0.7 | < 2 | Control |
| 19 | 7.7 | 6.2 | 1.5 | 6 | 0.8 | 2 | 0.3 | 0.3 | < 2 | Control |
| 20 | 8.3 | 8.0 | 0.4 | 76 | 9.1 | 19 | 2.3 | 2.4 | < 2 | Control |

PSG, polysomnography; RMMA, rhythmic masticatory muscle activity.

Table 2—Correlation of SR and MA outcomes with the RMMA.

| | SR | | | | MA | | | |
|-------------------|----------------------|----------------------|-----------------------|-----------------------|----------------------|----------------------|----------------------|----------------------|
| | MAL | MAR | TAL | TAR | MAL | MAR | TAL | TAR |
| Sleep, GS | r = 0.13 p = 0.60 | r = 0.11 p = 0.64 | r = -0.05 p = 0.85 | r = 0.004 p = 0.99 | r = 0.94 p < 0.01 | r = 0.95 p < 0.01 | r = 0.96 p < 0.01 | r = 0.55 p = 0.02 |
| Sleep + Awake, GS | r = 0.12 p = 0.63 | r = 0.09 p = 0.73 | r = -0.07 p = 0.77 | r = -0.04 p = 0.86 | r = 0.87 p < 0.01 | r = 0.84 p < 0.01 | r = 0.91 p < 0.01 | r = 0.32 p = 0.18 |

Correlation between the number of SR grinds detected with SR algorithm, MA bursts with the MA algorithm and RMMA bursts with the gold standard criteria during sleep period only (upper panels) or during sleep + awake periods (lower panels) in 20 individuals. Correlation coefficient (r) is from Pearson product moment correlation tests. SR, signal recognition; MA, moving average; RMMA, rhythmic masticatory muscle activity; GS, gold standard; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right.

Table 3—Correlation of MA episodes with RMMA episodes.

| GS | MA, Sleep | | | | MA, Sleep + Awake | | | |
|----|----------------------|----------------------|----------------------|----------------------|------------------------|----------------------|----------------------|----------------------|
| | MAL | MAR | TAL | TAR | MAL | MAR | TAL | TAR |
| | r = 0.88 p < 0.01 | r = 0.94 p < 0.01 | r = 0.90 p < 0.01 | r = 0.46 p < 0.05 | r = 0.6795 p < 0.01 | r = 0.77 p < 0.01 | r = 0.81 p < 0.01 | r = 0.15 p = 0.53 |

Correlation between the number of MA episodes detected with MA algorithm and RMMA episodes with the gold standard criteria during sleep periods only (left panels) or during sleep + awake periods (right panels) in 20 individuals. Correlation coefficient (r) is from Pearson product moment correlation tests. MA, moving average; RMMA, rhythmic masticatory muscle activity; GS, gold standard; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right.

highest when scored on a single channel of EMG. However, it was more difficult to distinguish between datasets with “mild” RMMA from the control subjects and the patients with normal RMMA frequency.

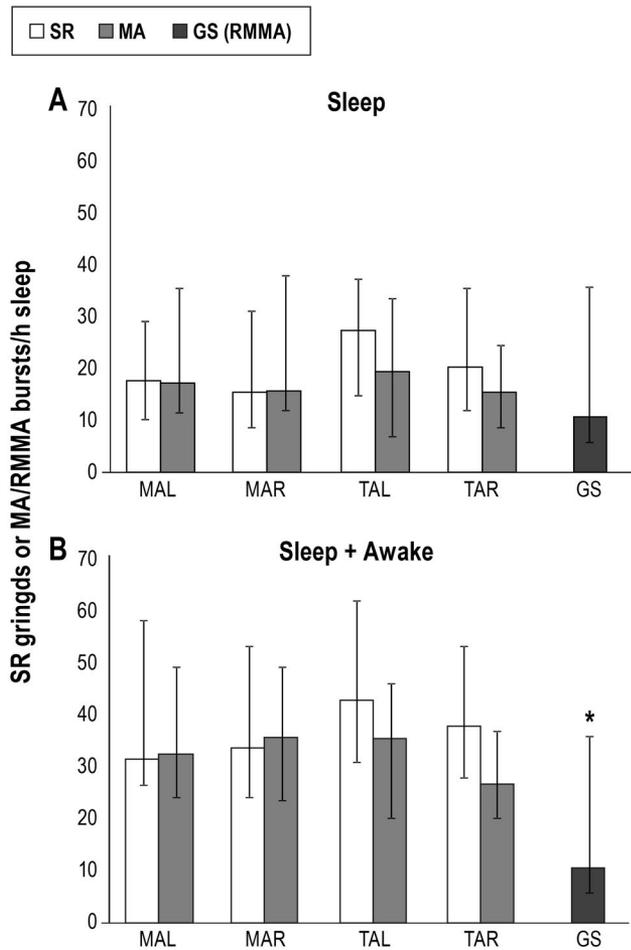
For comparison, the algorithm of another commercial device, Bitestrip, is described in brief below. The Bitestrip is a single-use device, to be placed on the masseter muscle. It measures and quantifies activity in a way that is rather

similar to the SR algorithm.¹³ The threshold is set at 30% of maximal voluntary clenches. Thirty minutes following activation (to allow time for falling asleep), the device begins counting continuously throughout the recording period those EMG masseter events that are at or above threshold for more than 0.25 s. A single count is limited to 1 s, thus an event > 1 s is counted as an additional event, as long as the additional time following the event already counted

exceeds 0.25 s. The Bitestrip device classifies the results into 4 classes: L = Very low sleep bruxism (0–30 events),

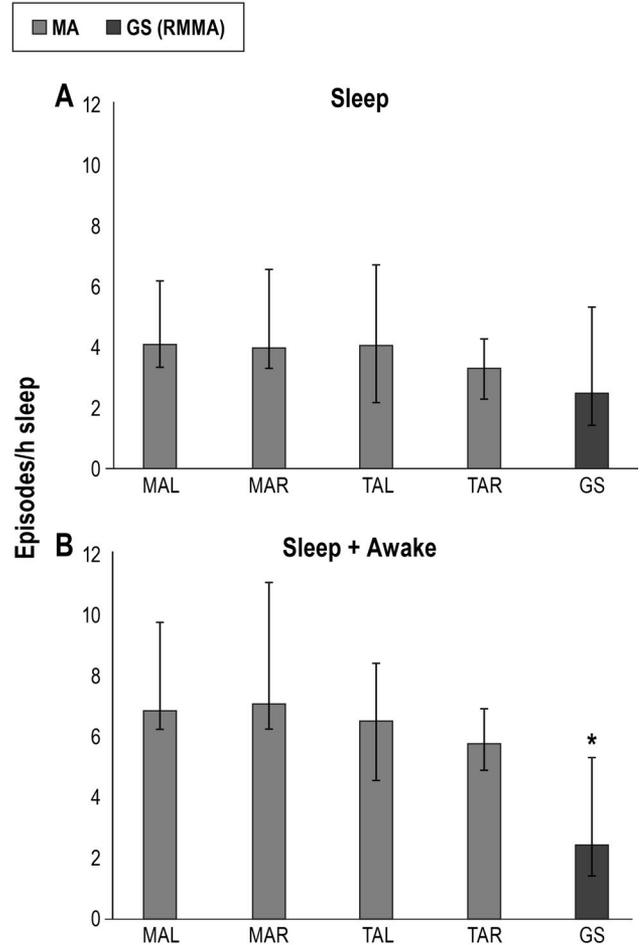
1 = Mild sleep bruxism (31–60 events), 2 = Moderate sleep bruxism (61–100 events), 3 = Severe sleep bruxism (> 101 events). Only the class is given as output to the user. The SR

Figure 2—Comparison between algorithms.



Median and interquartile ranges (n = 20) of SR grinds, MA bursts and RMMA bursts detected with the use of a signal recognition, moving average algorithm and the gold standard criteria in masseter left, masseter right, anterior temporalis left and anterior temporalis right during sleep periods (A) or during sleep + awakening periods (B). *Significant difference between the GS, and SR algorithm and MA algorithm (p < 0.05). SR, signal recognition; MA, moving average; GS, gold standard; RMMA, rhythmic masticatory muscle activity; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right.

Figure 3—Comparison between algorithms.

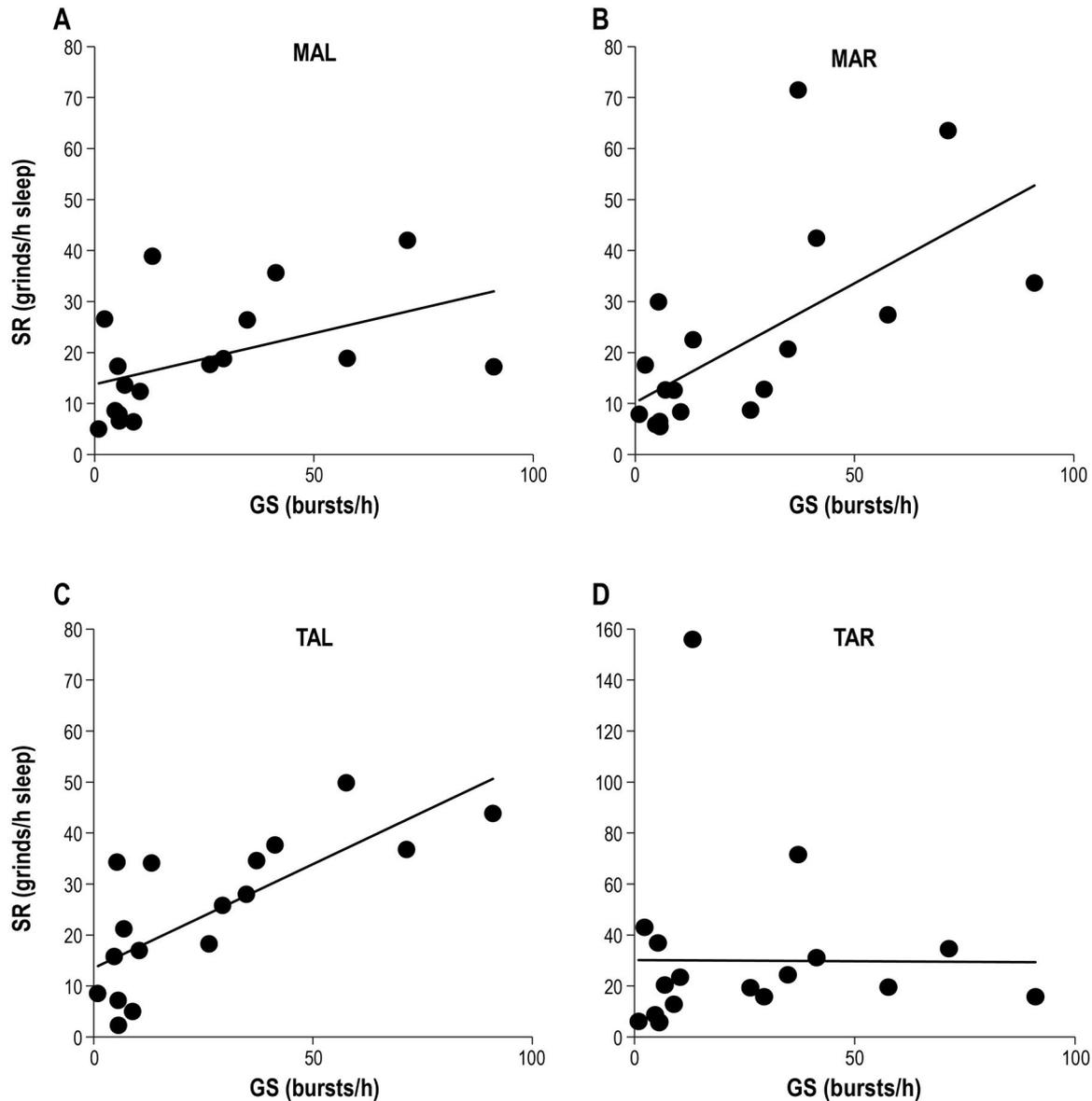


Median and interquartile ranges (n = 20) of episodes detected with the use of MA algorithm and the gold standard criteria in masseter left, masseter right, anterior temporalis right and anterior temporalis left during sleep period (A) or during sleep + awakening period (B). *Significant difference between the GS, and the MA algorithm (p < 0.05). MA, moving average; GS, gold standard; RMMA, rhythmic masticatory muscle activity; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right.

Table 4—Bland Altman test results of the agreement between SR/MA and GS assessment methods.

| | Sleep | | | | | | Sleep + Awake | | | | | |
|-----|-----------|--------------------|-----------|--------------------|-------------|--------------------|---------------|--------------------|-----------|--------------------|-------------|--------------------|
| | SR Grinds | | MA Bursts | | MA Episodes | | SR Grinds | | MA Bursts | | MA Episodes | |
| | LoA | Average Difference | LoA | Average Difference | LoA | Average Difference | LoA | Average Difference | LoA | Average Difference | LoA | Average Difference |
| MAL | 687.32 | -78.77 | 18.49 | 0.41 | 3.11 | -0.92 | 720.01 | -111.40 | 32.05 | -17.40 | 6.42 | -4.63 |
| MAR | 215.24 | -22.05 | 14.71 | -1.84 | 2.17 | -1.15 | 362.79 | -57.85 | 41.20 | -20.72 | 5.99 | -4.83 |
| TAL | 131.98 | -18.08 | 14.79 | 0.61 | 2.87 | -0.54 | 197.45 | -49.98 | 30.47 | -14.49 | 5.38 | -3.49 |
| TAR | 158.83 | -21.45 | 41.08 | 4.87 | 5.82 | 0.14 | 252.33 | -53.59 | 56.92 | -9.74 | 7.74 | -2.78 |

The numbers show that the limits of agreements for the SR method are very large. This is caused mainly by two records, where especially the MAL channel registered too many grinds. However, even after removing these outliers, the LoA's for the SR method are 3–10 times larger than for the MA (not shown), indicating that MA is a more accurate method than SR. It can also be seen that when looking at sleep only, the average difference for MA is close to 0 whereas for the SR it is negative, i.e. the SR method overestimates compared to GS and the MA does not. When including awake periods, both methods score higher than the GS, as was expected. SR, signal recognition; MA, moving average; GS, gold standard; LoA, limits of agreements; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right.

Figure 4—Correlations and levels of agreement between different algorithms.

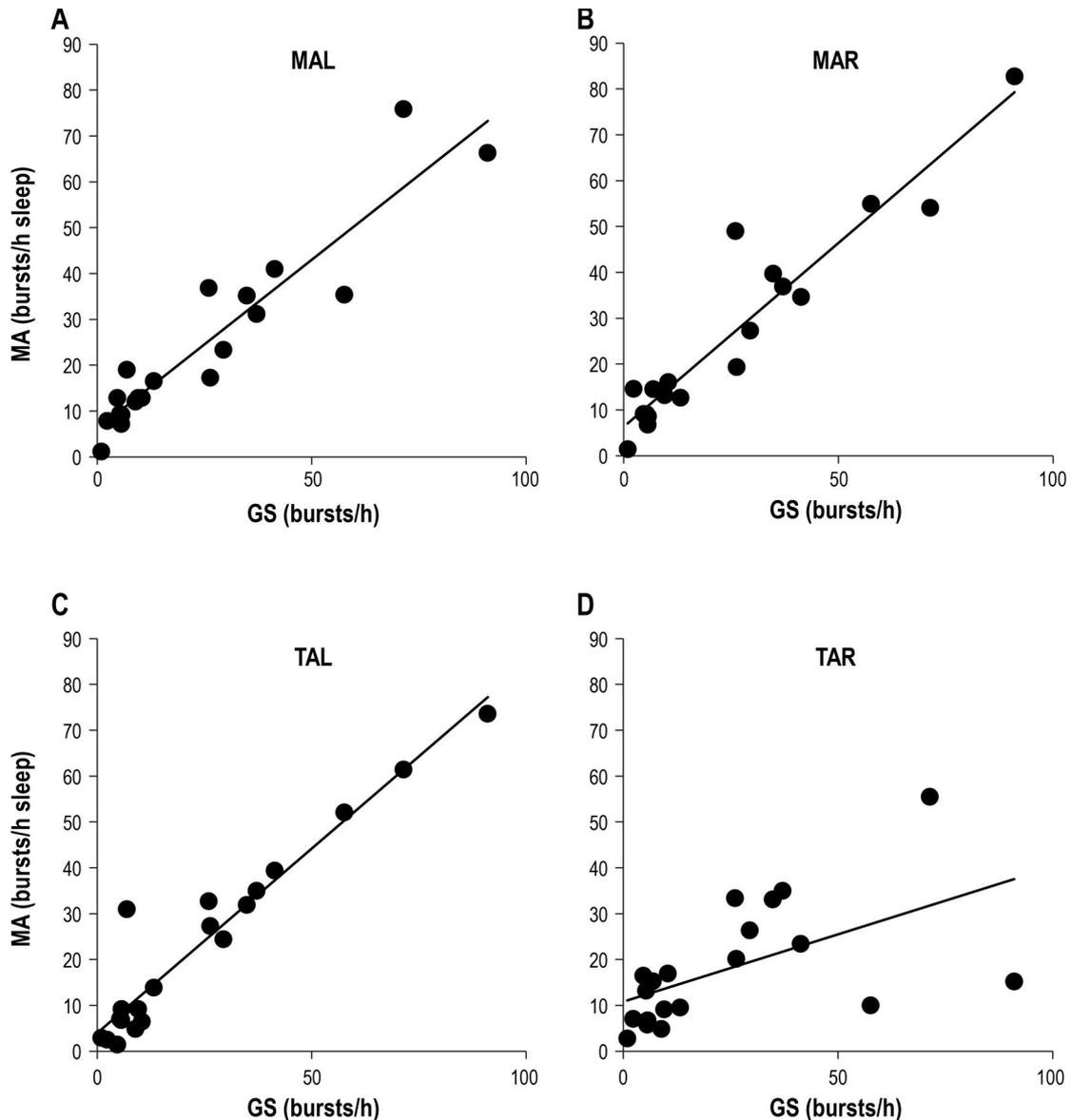
Correlation plots between the gold standard criteria for RMMA bursts detection per hour of sleep and SR algorithm for detection of number of SR grinds per hour of sleep in (A) masseter left, (B) masseter right, (C) anterior temporalis left, and (D) anterior temporalis right. $n = 20$. PSG data only scored during sleep period. SR, signal recognition; GS, gold standard; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right; RMMA, rhythmic masticatory muscle activity; PSG, polysomnography.

and MA algorithms provide a numerical output report facilitating the interpretation of the assessment of continuous multiple nights EMG recordings. This technique allows the study of other aspects such as variability, tendency, long-term averages, etc. Even though the SR algorithm did not show significant correlations with GS (Figure 4, Table 2), the ANOVA indicated no significant difference between the number of SR grinds, MA bursts and RMMA bursts detected with the SR and MA algorithms and the GS during sleep ($p > 0.40$, Figure 2A).

Although the manual scoring of RMMA is based on the complete set of data from the PSG recording, the EMG from the right masseter is used to determine the precise timing of bursts of activity. The rest of the information is used to determine whether the activity is true RMMA or other orofacial

activity/noise. For long-term home-use of a single-channel EMG device, the anterior temporalis muscle is a more convenient choice in most patients, as it is more practical to have an electrode placed on the temple rather than on the cheek, especially for people with a beard.²¹ Moreover, there seems to be no practical difference in using EMG from either the masseter or the temporalis muscle, and there was no practical difference in using either side of the head. This is in accordance with the bilateral although not symmetrical motor control of the jaw-closing muscles.²²

The new MA algorithm had a good correlation with the PSG scoring, when looking at periods of sleep only. However, only 77% of the RMMA episodes found by PSG analysis coincided with the MA episodes found by the MA algorithm, and only 64% of the bursts. If considering only the overall number of

Figure 5—Correlations and levels of agreement between different algorithms.

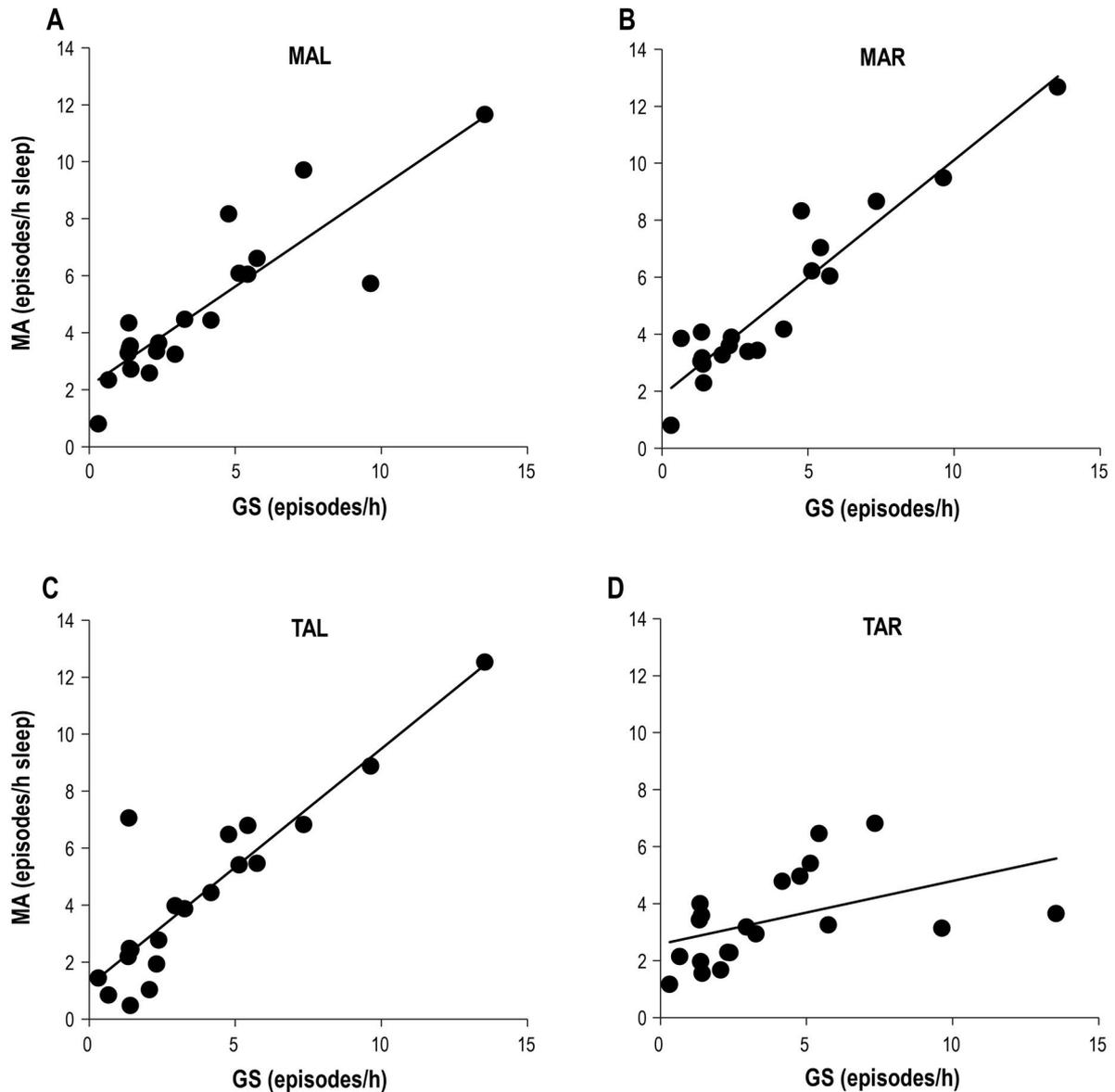
Correlation plots between the gold standard criteria for RMMA bursts detection per hour of sleep and the MA algorithm for detection of number of MA bursts per hour of sleep in (A) masseter left, (B) masseter right, (C) anterior temporalis left, and (D) anterior temporalis right. $n = 20$. PSG data only scored during sleep periods. MA, moving average; GS, gold standard; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right; RMMA, rhythmic masticatory muscle activity; PSG, polysomnography.

bursts and episodes, there seems to be no practical difference in using the number of bursts or number of episodes of RMMA for classification purposes. However, the better match between individual episodes scored implies that the number of episodes is a more robust measure.

Inclusion of awake periods in the analysis reduced the correlation between the single-channel EMG methods and the PSG scoring and generally increased the number of bursts/hour. The correlation was still fair, and the ordering of RMMA frequency was still reasonable. It is needed to highlight that the GS criteria to score RMMA excludes the transient awoken periods using the sleep staging. This exclusion can be made because the full PSG recordings including audio and video recordings make it possible to discriminate these transient awoken periods.

The present results shall be seen as a preliminary study, using previously recorded data only, to indicate to which extent a single EMG-channel can be used for scoring RMMA. Further, the knowledge gained in the study will be used for guiding the implementation of RMMA detection algorithms in a single-channel ambulatory device.

A single-channel EMG device is, however, limited in terms of diagnosis, because it is considered a type 4 device according to AASM, i.e., a screening tool under clinical use. Even so, the advantages of being able to collect large amounts of data, from many subjects, over long periods of time, in their own homes, may outweigh at least some of the limitations, especially if the device is commercially available, easy to use, and provides detailed information that is related to that provided by PSG, while the limitations are known and well described.

Figure 6—Correlations and levels of agreement between different algorithms.

Correlation plots between the gold standard criteria for RMMA episode detection per hour of sleep and MA algorithm for detection of number of MA episode per hour of sleep in (A) masseter left, (B) masseter right, (C) anterior temporalis left, and (D) anterior temporalis right. $n = 20$. PSG data only scored during sleep periods. MA, moving average; GS, gold standard; MAL, masseter left; MAR, masseter right; TAL, anterior temporalis left; TAR, anterior temporalis right; RMMA, rhythmic masticatory muscle activity; PSG, polysomnography.

In light of the present results we can conclude: (1) No significant difference was observed between the number of SR grinds, MA bursts, and RMMA bursts detected with the SR and MA algorithms and the GS during sleep. Therefore SR grinds and MA bursts may be useful for analyses of EMG activity during sleep and comparable to the GS. Nevertheless, we have to take into account that both the SR and MA algorithms detected significantly more grinds/bursts than the GS during sleep including awakening. (2) There was no significant difference in the quantification of RMMA based on either the EMG activity in the masseter or temporalis muscles and between sides. (3) The number of SR grinds, MA bursts and RMMA bursts, and MA episodes and RMMA episodes during sleep periods were significantly lower compared with during sleep + awakening periods.

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DISCLOSURE STATEMENT

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Class III Bimaxillary Orthognathic Surgery and Sleep Disordered Breathing Outcomes

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STUDY OBJECTIVES: To assess whether patients with class III malocclusions who underwent bimaxillary orthognathic surgery (BOS) are at an increased risk for obstructive sleep apnea (OSA) and/or a reduction in sleep-related quality of life compared to class III patients treated with orthodontics alone.

METHODS: Questionnaires were mailed to class III patients who had BOS and a matched control group of class III patients previously treated with orthodontics alone. Subjects were asked to complete the Berlin Questionnaire to assess OSA risk and the Functional Outcomes of Sleep-10 (FOSQ-10) and Epworth Sleepiness Scale (ESS) to assess sleep-related quality of life.

RESULTS: Seventy-eight subjects in the BOS group responded (29.8% response rate) and 24 subjects in the control group responded (13.7% response rate). Compared to the control group, the surgery group was significantly older, had longer follow-up times, and had more Caucasians. There was no significant difference between the surgery and orthodontic-only groups in their responses to the Berlin Questionnaire or the FOSQ-10. According to the Berlin Questionnaire, 9.0% of the surgery group were at high risk for OSA, while 16.7% of the orthodontic-only group were at high risk. The median total FOSQ-10 score for the surgery group and the orthodontic-only group was 18.3 and 18.1, respectively. The surgery group had a significantly lower ESS score of 6.3 compared to the orthodontic-only group score of 6.9. These findings compared favorably with scores for healthy individuals.

CONCLUSIONS: Patients receiving BOS for the correction of class III malocclusions are at no greater risk for OSA and/or reduction in sleep-related quality of life compared to Class III patients treated with orthodontics alone.

KEYWORDS: sleep disordered breathing, obstructive sleep apnea, quality of life, bimaxillary orthognathic surgery, 2 jaw surgery, Berlin Questionnaire, Functional Outcomes of Sleep Questionnaire-10, Epworth Sleepiness Scale

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Sleep disordered breathing (SDB) is regarded as a spectrum of diseases involving increased upper airway resistance during sleep and includes snoring, upper airway resistance syndrome, and obstructive sleep apnea (OSA).¹ Individuals with SDB can progress in severity from snoring to OSA, with increased airway collapse over time. OSA is characterized by the recurrent narrowing and obstruction of the pharyngeal airway during sleep. OSA and other forms of SDB have been reported to increase the risk of morbidity and mortality through the association with diabetes, hypertension, cardiovascular disease, and cerebrovascular disease.^{2–4} With the prevalence of SDB among adults in the United States estimated at 26%,⁵ jaw surgeries that could alter the risk for SDB should be carefully evaluated.

Mandibular setback surgery, either alone or in conjunction with maxillary advancement, is a surgical treatment option for patients with skeletal class III malocclusions. This type of malocclusion is characterized by either mandibular prognathism, maxillary deficiency, or a combination of both. Several studies have suggested that patients may develop OSA after mandibular setback surgery due to a narrowing of the posterior airway space (PAS).^{6–8} In a recent systematic review of cephalometric and cone-beam computed tomography (CBCT) studies on setback surgery and airway, the authors concluded that there is moderate evidence that isolated mandibular setback surgery leads to a decrease in oropharyngeal airway volume after surgery.⁹ Follow-up studies of a year or greater

have also shown a continued decrease in upper and middle airway dimension over time.^{10,11}

Due to concerns about airway reduction and unfavorable facial profile esthetics, many surgeons in the United States are doing fewer isolated mandibular setbacks. Less than 10% of class III surgery patients are receiving isolated setbacks, while approximately 40% undergo bimaxillary orthognathic surgery (combination of mandibular setback and maxillary advancement); the other half receive maxillary advancement surgery alone.¹² With the growing preference for bimaxillary orthognathic surgery (BOS), many recent studies have looked at its effect on the airway. In recent CBCT studies on changes in airway volume after BOS, the effect on the airway is still not clear. Some CBCT studies found an overall decrease in airway volume after BOS,^{13–15} but others found an increase,¹⁶ or even no change.^{17,18}

Although studies have reported an association between reduced airway volume and the risk for sleep disordered breathing,¹⁹ threshold limits for airway size have not been established for the development of SDB. Even if BOS leads to a decrease in airway volume, the risk for developing SDB after surgery has not been sufficiently explored. Studies are limited, and conclusions vary, regarding the extent to which BOS leads to SDB confirmed by polysomnography (PSG).^{16,20,21} Moreover, no study was identified that assessed patients' perception of sleep-related quality of life after BOS.

With the prevalence of sleep disordered breathing known to increase with age and evidence suggesting continual decreases

in airway space after setback surgery, long-term follow up studies on BOS and SDB risk are needed.^{10,11,22} The purpose of this study was to assess whether patients with skeletal class III malocclusions who underwent bimaxillary orthognathic surgery are at an increased risk for OSA and/or a reduction in perceived sleep-related quality of life compared to a group of non-surgical class III patients treated with orthodontics alone.

METHODS

This study was approved by the Biomedical Institutional Review Board of the University of North Carolina.

Subjects

Bimaxillary Surgery

Two hundred sixty-two subjects with class III malocclusions who had undergone bimaxillary orthognathic surgery at the University of North Carolina (UNC) Memorial Hospital between 2003 and 2012 were identified from the UNC orthognathic surgery database after accounting for inclusion and exclusion criteria. Subjects were included if they were at least 1 year post-surgery, had current contact information, and were able to understand and read English. The presence of a congenital syndrome led to exclusion from the study.

Orthodontic-Only Control

One hundred seventy-five patients with class III malocclusions who were treated nonsurgically in the UNC graduate orthodontic clinic and who met the same inclusion and exclusion criteria as the surgery group were frequency matched to the surgery group based on gender, age, and time since deband.

Each subject was mailed a packet which included a cover letter for informed consent, a HIPAA authorization, an opt-out form, a set of questionnaires, and a business reply envelope. Demographic data, information on OSA diagnosis or management since their class III treatment, and responses to items on three questionnaires to assess OSA risk and quality of life were requested. The questionnaires were created in Teleform so that returned questionnaires could be easily scanned, verified, and input into a SAS dataset for analysis. Non-responders were mailed a second and, if necessary, a third packet at monthly intervals.

Questionnaires

Subjects were asked to report age in years and months, gender (male/female), height in feet and inches, weight in pounds, race/ethnicity, and information on previous OSA diagnosis or treatment. Three sleep questionnaires (Berlin, Functional Outcomes of Sleep-10, and Epworth Sleepiness Scale) were completed by participants in this study to assess OSA risk and sleep-related quality of life. Although the diagnostic gold standard for assessing OSA is overnight polysomnography (PSG), validated disease-specific questionnaires are frequently used as convenient and cost-effective screening tools for OSA.²³

The Berlin Questionnaire is a validated survey that scores subjects as “high risk” or “low risk” for OSA.²⁴ In a recent systematic review of validated OSA screening questionnaires, the Berlin had a pooled sensitivity and specificity of, 77% and

74%, respectively.²⁵ The Berlin Questionnaire is composed of 10 questions divided among 3 symptom categories: snoring, daytime sleepiness, and obesity/hypertension. Patients with frequent and persistent symptoms in any 2 of the 3 categories are considered at high risk for OSA. At least 2 affirmative answers in either the snoring or daytime sleepiness categories is confirmation of the presence of that symptom. For the obesity/hypertension category, an answer of “yes” to having hypertension or a body mass index (BMI) > 30 kg/m² is considered a positive score. BMI was calculated from the self-reported height and weight.

The Functional Outcomes of Sleep Questionnaire (FOSQ-30) is a valid and reliable 30-item questionnaire that is considered to be the gold standard in assessing the impact of sleepiness on quality of life.²⁶ The FOSQ-10 is a shorter version of the original FOSQ-30 and has been shown to be easier to use and to reach the same statistical conclusions as the longer version regarding comparisons in sleep-related quality of life between normal controls and patients with OSA.²⁷ The FOSQ-10 assesses quality of life via 10 questions measuring 5 subscales: general productivity, activity level, vigilance, social outcome, and intimacy and sexual relationships.²⁶ Total scores range from 5–20 with lower values suggesting poorer sleep-related quality of life.

The Epworth Sleepiness Scale (ESS) assesses daytime sleepiness and is one of the most widely used sleep assessment questionnaires in clinical settings. Although the ESS has been found to have a low predictive value when used as a screening method for OSA,^{28–30} a study using participants from the Sleep Heart Health Study found excessive daytime sleepiness to be strongly associated with reduced quality of life.³¹ The subject rates from 0–3 (0-never, 3-high) his/her chances of dozing off in 8 situations that are often encountered in daily life. ESS scores range from 0–24, and a score > 10 (i.e., 11+) is considered indicative of excessive daytime sleepiness.³²

Statistical Analysis

All statistical analyses were conducted using SAS (SAS Institute Inc. Version 9.3 2011. Cary, NC: SAS Institute Inc.).

The orthognathic surgery and orthodontic-only groups were compared to assess characteristic differences (age, sex, time since surgery/deband, race, BMI, diagnosis of OSA, prescription for OSA treatment) and to assess whether the groups differed with respect to perception of quality of life and risk for OSA. Descriptive and inferential statistics were used to analyze the data. A χ^2 or Fisher exact test was used to compare categorical variables, and a Cochran-Mantel-Haenszel row mean score test was used to compare continuous variables between groups. The level of significance was set at 0.05.

RESULTS

Of the 262 surgery subjects sent questionnaires, 78 patients responded (response rate of 29.8%). Surgery participants included 46 females and 32 males with a median age of 27.6 (19.1, 36.2). They were all at least 2 years post-surgery with a median time since surgery of 5.4 (2.8, 8.1) years. Twenty-four of the 175 subjects in the control group responded (response rate of 13.7%). The control group consisted of 15 females and 9 males with a median age of 22.0 (14.6, 29.4) years. They were

all at least 1 year post deband with a median time since deband of 4.1 (1.8, 6.4) years. The 2 groups were significantly different in median age ($p < 0.01$), time since surgery/deband ($p < 0.05$), and race ($p < 0.01$). Compared to the control group, the surgery group was older, had a longer follow-up time, and was composed of a higher percentage of Caucasians. No statistical difference between gender and BMI was detected (Table 1). One participant in the surgery group acknowledged being treated with an oral appliance, but denied having a previous OSA diagnosis. Either the patient failed to recall a diagnosis or was provided the oral appliance in absence of an official diagnosis. The patient also stated that it had been 2 years since the appliance was used.

The Berlin Questionnaire did not reveal any statistically significant difference in the OSA risk assessment between the surgery and orthodontic-only groups nor were there any statistically significant differences between groups in any of the symptom categories (Table 2). Overall, 9.0% of the surgery group and 16.7% of the orthodontic-only group were found to be at high risk for OSA.

Analysis of the FOSQ-10 indicated no statistically significant difference between the total FOSQ-10 score for the surgery and orthodontic-only groups with median total scores of 18.3 (16.4, 20.1) and 18.1 (15.7, 20.6), respectively. The two groups did not differ significantly in any of the subscales: productivity, activity, vigilance, social outcomes, or intimacy and sexual relations (Table 3).

The difference in the Epworth Sleepiness Scale scores was significantly different between the surgery and orthodontic-only groups ($p < 0.05$). After excluding those with missing data, the median ESS score for the surgery group was 6.3 (3.3, 9.3) compared to 6.9 (2.4, 11.4) for the orthodontic-only group. Both median scores, however, fell within the normal range for daytime sleepiness. When assessed for the proportion of subjects who demonstrated excessive daytime sleepiness, 10.5% of the surgery group and 20.8% of the orthodontic-only group had an ESS total score > 10 ($p = 0.29$; Table 4)

DISCUSSION

Sleep disordered breathing, including OSA, is a serious condition associated with increased morbidity and mortality.^{3,4} Isolated mandibular setbacks are becoming rare in the United States due to both esthetic reasons and concerns over the risk of airway reduction possibly leading to SDB. Studies on the effects of bimaxillary orthognathic surgery with mandibular setback (BOS) on sleep function are limited. Objective measures, primarily from polysomnograms, have traditionally been reported in the literature and have led to varying conclusions. In the PSG study of Foltán et al,²⁰ BOS was found to worsen respiratory parameters with significant decreases in oxygen saturation (SpO_2) and nasal airflow measured before and after (mean 8.5 months)

Table 1—Descriptive statistics for study participants.

| Variable | Surgery (n = 78) Median (P25, P75) | Orthodontic-Only (n = 24) Median (P25, P75) | p value |
|---------------------------|--|---|---------|
| Age | 27.6 (19.1, 36.2) | 22.0 (14.6, 29.4) | 0.006 |
| Time since surgery/deband | 5.4 (2.8, 8.1) | 4.1 (1.8, 6.4) | 0.035 |
| Body mass index | 25.4 (19.8, 27.8) | 23.4 (19.1, 27.8) | 0.110 |
| Gender | n (%) | n (%) | |
| Male | 32 (41.0) | 9 (37.5) | 0.760 |
| Female | 46 (59.0) | 15 (62.5) | |
| Race | | | |
| Caucasian | 64 (83.1) | 12 (52.2) | 0.002 |
| Other | 12 (16.9) | 11 (47.8) | |
| Previous OSA diagnosis | 0 | 0 | — |
| History of OSA treatments | 1 (1.3) | 0 | 1.000 |
| Oral appliance | 1 (1.2) | 0 | — |

P25, 25th percentile; P75, 75th percentile.

Table 2—Berlin Questionnaire results.

| Symptom categories | Surgery n (%) | Orthodontic-Only n (%) | p value |
|--------------------|------------------|---------------------------|---------|
| Snoring | | | |
| Positive | 11 (14.1) | 6 (25.0) | 0.22 |
| Negative | 67 (85.9) | 18 (75.0) | |
| Daytime sleepiness | | | |
| Positive | 17 (21.8) | 6 (25) | 0.74 |
| Negative | 61 (78.2) | 18 (75.0) | |
| Blood pressure/BMI | | | |
| Positive | 15 (19.2) | 2 (8.3) | 0.34 |
| Negative | 63 (80.8) | 22 (91.7) | |
| Risk assessment | | | |
| Low risk | 71 (91.0) | 20 (83.3) | 0.29 |
| High risk | 7 (9.0) | 4 (16.7) | |

Table 3—Functional Outcomes of Sleep Questionnaire-10 results.

| Subscale Scores | Surgery Median (P25, P75) | Orthodontic-Only Median (P25, P75) | p value |
|--|------------------------------|---------------------------------------|---------|
| General productivity subscale | 3.6 (3.1,4.1) | 3.6 (3.0,4.2) | 0.74 |
| Activity level subscale | 3.5 (2.9,4.1) | 3.6 (3.1,4.1) | 0.61 |
| Vigilance subscale | 3.7 (3.2,4.1) | 3.7 (3.1,4.3) | 0.47 |
| Social outcomes subscale | 3.9 (3.4,4.3) | 3.8 (3.3,4.3) | 0.89 |
| Intimacy and sexual relations subscale | 3.7 (3.0,4.3) | 3.5 (2.5,4.4) | 0.06 |
| Total score ^a | 18.3 (16.4,20.1) | 18.1 (15.7,20.6) | 0.90 |

^aTotal score is a mean-weighted item score.

surgery. However, in a different PSG study, Hasebe et al.²¹ was unable to detect significant differences in SDB or changes in

Table 4—Epworth Sleepiness Scale (ESS) results.

| | Surgery Median (P25, P75) | Orthodontic-Only Median (P25, P75) | p values |
|-----------------|--------------------------------------|---|-----------------|
| ESS Score | 6.3 (3.3,9.3) | 6.9 (2.4, 11.4) | 0.049 |
| | n (%) | n (%) | |
| ESS Scores < 10 | 68 (87.2) | 19 (79.2) | 0.29 |
| ESS Scores > 10 | 8 (10.5) | 5 (20.8) | |

SpO₂ or apnea-hypopnea index (AHI) in patients 6 months after BOS. The investigators did note that 2 patients with very large mandibular setbacks were diagnosed with mild OSA after surgery. Turnbull and Battagel³³ compared overnight pulse oximetry and respiratory noises before and after BOS and found no significant changes despite identifying a reduction in the retrolingual airway diameter in all patients. In a recent PSG study by Gokce et al.,¹⁶ sleep quality and efficiency improved significantly after BOS (mean 1.4 years) with significant increases in SpO₂ and decreases in AHI.

While objective measures of SDB have traditionally been reported in the literature, quality of life assessments are increasingly being recognized as an important outcome variable as well.^{31,35} A number of studies have examined quality of life subsequent to jaw surgery for dentofacial deformities,³⁶ however, no studies were identified that explored the impact of Class III jaw surgeries on sleep-related quality of life. Our finding of no significant difference in Berlin Questionnaire scores between the BOS group and the orthodontic-only group is consistent with the objective measure studies that were unable to demonstrate an increased risk of SDB after BOS. The BOS group scores were also found to be similar to a recent population study on OSA risk. For example, the Berlin Questionnaire was used in a national sleep poll of 1,506 people, and 19% of participating adults were found to meet the criteria for high risk of OSA.³⁴ In our study, 9.0% of the surgery group and 16.7% of the orthodontic-only group were found to be at high risk for OSA.

In our study, we used 2 validated sleep questionnaires, the FOSQ-10 and ESS, to focus on how BOS may affect patients' perception of sleep-related quality of life. To our knowledge, this is the first study to assess patients' perception of daytime sleepiness after BOS. With a median post-surgery time of 5.4 (2.8, 8.1) years, our study offered information on subjects with a longer follow-up than any previous study that measured sleep outcomes after BOS.

In a recent study evaluating ESS scores between OSA patients and non-OSA patients, the average values found were 10.9 and 7.7, respectively.³⁰ Although we found a statistically significant difference in ESS scores between our 2 groups ($p < 0.05$), with the surgery group having a lower median daytime sleepiness score, ESS scores in both groups fell within the normal range. Both groups in our study were less than the reported non-OSA score of 7.7, with the surgery group having a median ESS score of 6.3 (3.3, 9.3) and the orthodontic-only group a score of 6.9 (2.4, 11.4). It has been estimated that 10% to 20% of the general population has ESS scores > 10.³⁷ Our results were in that range with 10.5% of the BOS group and 20.8% of the orthodontic-only group having ESS scores > 10. The significantly lower ESS score and lower proportion of scores > 10 in the surgery group

suggest that BOS does not adversely impact daytime sleepiness.

The FOSQ was developed to measure the impact of sleep on quality of life. Higher FOSQ scores reflect better quality of life. In a previous FOSQ-10 study, patients with OSA had an average score of 12.5, while non-OSA participants had an average score of 17.2.²⁷ In our BOS group, the FOSQ-10 score of 18.3 (16.4, 20.1) compared favorably to the reported value in the non-OSA patients. Thus, results from both the ESS and FOSQ-10 in our study suggest that Class III bimaxillary surgery did not significantly affect the patients' sleep-related quality of life post-surgery.

Study Limitations

The median age of both of our study groups was relatively young. Due to the conversion in 2003 from paper charts to the electronic patient record (EPR) at our institution, we were limited in the time frame for which we had current contact information for patients in the UNC surgery database. As such, the median age of both groups was < 28 years and may not reflect OSA outcome differences that may occur with increasing age.³⁸ A well-known risk assessment questionnaire, the STOP-Bang, uses age 50 as a threshold for increased OSA risk.³⁹ If or how our groups differ after age 50 would be valuable information on clarifying whether BOS is associated with an increased risk of OSA over time. Although we attempted to frequency match the age of the orthodontic-only group to the age of the surgery group respondents, the median age of the surgery group was approximately 5 years older, which one might have speculated would have magnified a difference in OSA risk if it existed.

The increased follow-up time of approximately 1 year for the BOS group compared to the orthodontic-only group is understandable because up to a year of orthodontic finishing remains after surgery. We were not able to compare deband dates between groups because we did not have access to the deband dates of the surgery group. The majority of the orthognathic surgery patients seen at UNC have their orthodontic treatment carried out by local orthodontists.

The BMI used in this study was calculated from self-reported height and weight values. Although the BMI was not significantly different between groups, any inaccuracies in BMI could also have altered the scoring of the Berlin Questionnaire which uses BMI as one of its variables. Given that the study design did not evaluate patients clinically, obtaining accurate height and weight data from participants was not possible. The significantly more Caucasians in the surgery group is consistent with the demographics of the surgery patients at UNC.

There was a significant difference in response rate between the BOS group and orthodontic-only group with response rates of 29.8% and 13.7%, respectively. The BOS subjects may have been more likely to participate in our study because many had previously agreed to participate in an ongoing surgery stability study at UNC. In addition, the BOS subjects may have felt more of an obligation to participate because of the intense emotional and psychological impact that comes from the profound positive changes in function and facial esthetics after surgery. The low response rate in both groups is likely related to the transient nature of individuals who have treatment in

their teens and early twenties and then relocate for college and jobs. No institutional effort is made to periodically update the contact information of patients who have completed treatment, which possibly negatively impacts retrospective study response rates, thereby limiting the generalizability of the findings.

To our knowledge, this study provides the longest follow-up information to date on the effects of Class III bimaxillary orthognathic surgery (BOS) on sleep disordered breathing (SDB). Moreover, this is the first study to assess sleep-related quality of life after BOS. The results of this study suggest that young adults receiving this double jaw surgical procedure for the correction of class III malocclusions are at no greater risk for OSA and/or reduction in sleep-related quality of life than patients treated with orthodontics alone. Patients have been shown to be at most risk for SDB if the mandible is set back significantly, preventing adaption to their new respiratory position during sleep.²¹ Bimaxillary orthognathic surgery for Class III malocclusions may be able to limit the risk of SDB by minimizing the amount of mandibular setback required and through compensating increases in the nasopharyngeal and velopharyngeal airways from the maxillary advancement.^{16,40} Prospective research is needed to evaluate sleep-related quality of life before and after BOS and to examine correlations between PSG data, sleep questionnaires, and 3D airway parameters. The ability to more clearly identify an orthognathic surgery patient's presurgical risk of developing SDB is a goal that would guide surgeons and benefit patients in the future.

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Development of a Simplified Pediatric Obstructive Sleep Apnea (OSA) Screening Tool

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STUDY OBJECTIVES: To develop and test a pediatric screening tool to gauge the risk that an individual child would have OSA prior to a dental procedure by a pediatric dentist requiring minimal or moderate oral conscious sedation.

METHODS: 180 pediatric patients completed a polysomnogram at the VCU Center for Sleep Medicine between February 2011 and February 2013. A modified STOP-Bang questionnaire was validated with polysomnography.

RESULTS: A validated adult questionnaire, STOP-Bang, was modified using more typical pediatric risk factors for OSA: presence of snoring (S), tonsillar hypertrophy (T), obstruction (O), daytime tiredness or neuropsychological-behavioral symptoms such as ADHD or daytime irritability (P), BMI percentile for age (B), age at diagnostic screening (A), presence of neuromuscular disorder (N), and presence of genetic/congenital disorder (G). A positive scoring from these variables was measured against the patients acquired in-laboratory polysomnogram using the standard OSA measure, apnea-hypopnea index. A multiple logistic regression analysis found a statistically significant relationship ($p = 0.0007$), with a minimum of 4 variables needed to have a sensitivity of 57% and a specificity of 78%. Only obstruction, BMI, and age showed a strong significant relationship to OSA. The presence of an obstruction was positively related to apnea ($p = 0.0010$). Most of the other components had an odds ratio larger than one (indicating a nominally positive relationship).

CONCLUSIONS: The pediatric modified STOP-Bang screening tools showed a statistically significant relationship. Only obstruction, BMI, and age showed a predictive relationship to OSA. Although the PM-STOP-Bang results do not lend support to including other known risk factors of pediatric OSA, further studies are warranted of a revised screening tool that include recognized risk factors.

KEYWORDS: pediatric obstructive sleep apnea, simplified screening tool

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Sleep disordered breathing encompasses a wide range of upper airway disorders from primary snoring (PS) to obstructive sleep apnea (OSA). OSA results from impedance to airflow in the upper airway during sleep; these periodic obstructions of the upper airway interfere with normal respiratory gas exchange and subsequently interrupt sleep.^{1,2} OSA has become recognized as one of the most common, underdiagnosed chronic diseases.^{3–5} People of all ages are affected with OSA. Recently studies have shown increased numbers among pediatric and adolescent populations.⁶ The prevalence of obstructive sleep apnea (OSA) in children is estimated to be 1% to 3%,⁷ while primary snoring occurs in 3% to 12% of the pediatric population.⁸ Mild cases of pediatric OSA are recognized and at times treated; however, measurable effects on development, cardiopulmonary, or metabolic systems have been difficult to validate. OSA is associated with behavioral problems, poor school achievement, and, in severe cases, pulmonary hypertension.² Many studies have been conducted to identify adverse effects of sleep disorders, yet few studies have examined how health care providers may identify and treat sleep disorders.¹⁰

Dentists see their patients more frequently than their primary care doctors, and so have a greater opportunity to observe signs and symptoms of OSA.⁶ However, many potential sleep disorders in children are unrecognized and underreported, and overall the condition is under-diagnosed.¹¹ Dentists who practice sedation dentistry should exercise extra precautions when treating

patients with risk of sleep apnea. Minimal and moderate oral conscious sedation and general anesthesia are commonly used in pediatric dentistry. During sedation, children with OSA have an increased vulnerability of their airway undergoing pharyngeal collapse and of having upper airway obstruction.⁷ Thus pediatric dentists have an acute responsibility to be able to identify patients who may have OSA.⁵ The risk of postoperative respiratory complications among the pediatric population ranges from 0 to 1.3%; however, for children with OSA, the rates have been reported to be 16% to 27%.^{12,13} The prevalence of OSA in children is most elevated between 2 to 6 years of age. In this age range, pharmacologic measures are most often used to complete diagnostic and therapeutic procedures.⁷

While polysomnography (PSG) remains the gold standard for diagnosing OSA, there are many challenges due to the limited number of sleep laboratories and the high cost of performing a PSG on each child who snores and who may be at risk.⁸ Available non-PSG screening tests have poor sensitivity for milder OSA, and overall poor specificity.⁸ Moreover, there remains a challenge to differentiate PS from OSA in a “cost-effective, reliable, and accurate manner before recommending invasive or intrusive therapies, such as surgery or continuous positive airway pressure.”⁸

Sleep questionnaires that are completed by the parent and child are a crucial component of behavioral and physiological sleep assessment. Pediatric questionnaires are mostly

retrospective in that the parents report on past sleep patterns and behaviors that are typical of their child. In 2008, Chung et al. developed and validated a STOP questionnaire as a screening tool for OSA in patients 18 years and older. This questionnaire asks four yes/no questions: do you snore loudly?, do you feel tired during the daytime?, has anyone observed you stop breathing during your sleep?, and do you have high blood pressure? These questions along with body mass index, age, neck size, and gender (BANG) were found to have a sensitivity of 83.6, 92.9, and 100% (for mild, moderate, and severe OSA, respectively).¹⁵ In 2006, the American Society of Anesthesiologists (ASA) Task Force on Perioperative Management of Patients with Obstructive Sleep Apnea recommended a checklist as a routine screening tool to utilize in surgical patients who may have OSA. The ASA checklist has 12 items for adults and 14 items for children, but was only validated by Chung for its use on adults.¹⁶

In 2011 Spruyt and Gozal published a review on pediatric sleep questionnaires that examined 57 sleep measures that were used to screen children for sleep disorders including OSA.¹⁴ Only 2 questionnaires fulfilled all desirable criteria: The Sleep Disturbance Scale for Children (SDSC) at a cutoff score of 39 provided a sensitivity of 0.89 and a specificity of 0.74; and The Sleep Disorders Inventory for Students-Children (SDIS-C) showed a sensitivity of 0.91 and a specificity of 0.62 for the category of obstructive sleep apnea syndrome.^{17,18} This review documented that few standardized screening tools exist thus far to determine risk for OSA in children.¹⁴ Pediatric dentists and anesthesiologists alike would benefit from a standard screening tool, similar to the STOP-Bang, to determine if OSA may exist in potential sedation and anesthesia pediatric patients.

OSA is often the Achilles heel of pediatric sedation and analgesic programs⁷; thus it is imperative that pediatricians and pediatric dentists be able to identify a child who may be at risk for OSA so that appropriate referrals for a definitive diagnosis can be made. Currently there is no screening tool available to pediatric dentists to aid in recognizing OSA during the preoperative appointment or to help direct specialty consultation for patients undergoing minimal and moderate oral conscious sedation.

The primary aim of the study was to modify the STOP-Bang instrument for use in a pediatric setting and test the scale to screen for sleep apnea in children. The secondary aims were to test each of the components of the instrument.

METHODS

This project was granted an exempt status from the Virginia Commonwealth University Institutional Review Board (# HM15027). This was a retrospective chart review of the routine exam of patients referred for a sleep study. The original data was collected at the Center for Sleep Medicine for pediatric patients (under age 18) from February 1, 2011, to February 1, 2013, with no previous sleep disorder diagnosis. Study data were collected and managed using REDCap.²⁰ To be included, patients had to have a completed polysomnogram and a completed sleep questionnaire (Appendix 2) in the chart record.

Using data from the Medical College of Virginia electronic health record and the Sleep Center database, 180 patients

were eligible. The following variables were collected: age of patient at time of PSG, gender, race, height, weight, body mass index, presence of snoring, presence of tonsillar hypertrophy, obstruction while sleeping, presence of neurobehavioral symptoms, daytime tiredness or irritability, presence of neuromuscular disorders, presence of genetic disorders, and apnea-hypopnea index (AHI). The child's age, gender, height and weight was used to verify the reported BMI and to calculate the BMI-percentile-for-age (using the nccd.cdc.gov/dnpabmi/Calculator.aspx calculator). The presence of snoring, obstruction while sleeping, and daytime tiredness or irritability was determined by the sleep study questionnaire completed by the parent and patient prior to the PSG.

Apnea-Hypopnea Index (AHI)

OSA was diagnosed by a patient's apnea-hypopnea index (AHI). The AHI represents the average number of apneas and hypopneas per hour of sleep.⁸ In pediatric OSA, which has the same prevalence in boys and girls, more than one obstructive apnea event of any length per hour of sleep is considered abnormal.^{21,22,23} Based on these recommendations, apnea was categorized as: none (AHI \leq 1.5), mild (AHI > 1.5), moderate (AHI > 5), or severe (AHI > 15). The primary categorization was a binary outcome: apnea negative (AHI \leq 5) or apnea positive (AHI > 5).

Pediatric Modified (PM) STOP-Bang

PM-STOP-Bang (heretofore simply referred to as STOP-Bang) was the sum of the presence of snoring (S), tonsillar hypertrophy (T), observed obstruction (O), neuropsychological-behavioral symptoms such as ADHD or daytime irritability (P), BMI percentile for age and gender above 95% (B), age at diagnostic screening (A), presence of neuromuscular disorder (N), and presence of genetic/congenital disorder (G). Yes values were scored as 1 and all other values (No and unknown) were scored as zero.

Data Analysis

All analyses were performed using SAS software (JMP pro version 11, SAS version 9.3, SAS Institute Inc., Cary NC). The statistical methods included screening of each diagnostic characteristic (using χ^2 analysis) and a multiple logistic regression analysis of the OSA diagnosis to determine which diagnostics characteristics are associated with the diagnosis. Final reporting included odds ratios and 95% confidence intervals on all estimates.

Using the projected 250 charts that were initially thought to be available, and estimating the prevalence OSA \geq 25% and odds ratios \geq 2, the study had approximately 80% power (at $\alpha = 0.05$).

RESULTS

After excluding 27 because of incomplete data, 153 subjects with usable data were analyzed (see Table 1). Neither gender ($p = 0.4455$) nor race ($p = 0.1368$) appeared related to the AHI scores. Subjects ranged in age from 38 months to 17.5 years. The average BMI percentile for age was 73%. There were 60 subjects (39%) who were described as obese since they were above the 95th percentile for age and gender.

AHI

The primary outcome variable was observed apneas and hypopneas, as indicated by AHI. The raw AHI values ranged from 0 to 85.7 apnea-hypopnea events per hour, with a median value of 0.8. The strongly skewed values yielded a mean of 4.08 (SD 9.53). There were 82% considered negative for apnea (96 = none, and 29 = mild), and therefore 18% (16 moderate and 12 severe) were considered positive.

Scale Values

The components of the STOP-Bang scale are summarized in the prevalence column of Table 2. For instance, over 59% of all subjects had a positive indication in the medical record for snoring (n = 91). Thus the prevalence of each of these components ranged from a high of 60% (psychological symptoms) to a low of 14% for both neuromuscular disorders and for genetic/congenital disorders.

For the STOP-Bang scale the scores ranged from 0 to 6 (Mean = 2.76, SD = 1.34). Logistic regression was used to test for a relationship between the STOP-Bang scale

and apnea. There was a statistically significant relationship (likelihood ratio $\chi^2 = 11.5$, $p = 0.0007$). The stacked bar chart in Figure 1 shows that for STOP-Bang scores ≤ 3 , at least 72% were categorized as OSA = none (the white area of the graph), and that this white proportion decreases with increasing

Table 1—Demographic characteristics of study subjects (n = 153).

| Characteristic | n (%) | | | |
|------------------------------------|-------------|-----------|---------------|--------------|
| Gender | | | | |
| Female | 70 (45.8) | | | |
| Male | 83 (54.2) | | | |
| Race/ethnicity | | | | |
| Asian | 4 (2.6) | | | |
| Black or African American | 66 (43.1) | | | |
| Hispanic | 5 (3.3) | | | |
| White | 69 (45.1) | | | |
| Unknown/not reported | 9 (5.9) | | | |
| | Mean | SD | Median | Range |
| Age (years) | 10.59 | 4.10 | 10.50 | 3.17–17.50 |
| Age (months) | 127.06 | 49.21 | 126.00 | 38.00–210.00 |
| BMI (kg/m ²) (n = 152) | 23.32 | 8.85 | 21.17 | 11.26–59.69 |
| BMI percentile | 72.73 | 33.52 | 91.42 | 0.00–100.00 |

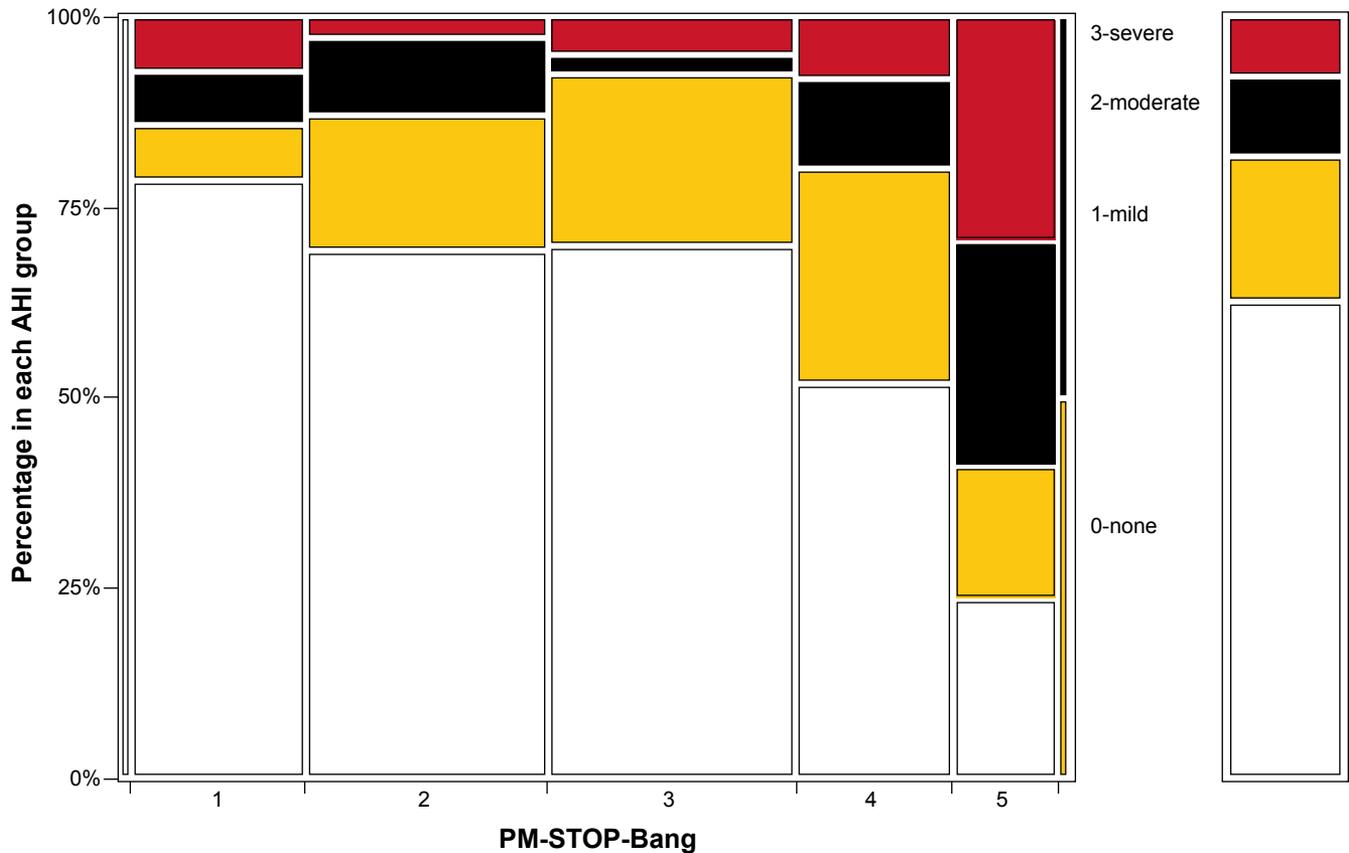
SD, standard deviation.

Table 2—Components of STOP-Bang and the relationship with obstructive sleep apnea.

| Risk Indicator | Prevalence | OSA | | Unadjusted | | Adjusted | |
|--|------------|----------|----------|------------|---------|----------|---------|
| | | Negative | Positive | OR | p value | OR | p value |
| Snore | | | | | | | |
| Yes | 59% | 70 | 21 23% | 2.36 | 0.064 | 0.74 | 0.677 |
| No | 33% | 45 | 6 11% | | | | |
| Unknown | 7% | 10 | 1 | | | | |
| Tonsillar hypertrophy | | | | | | | |
| Yes | 20% | 23 | 8 26% | 1.77 | 0.226 | 1.96 | 0.245 |
| No | 33% | 40 | 10 16% | | | | |
| Unknown | 47% | 62 | 10 | | | | |
| Obstruction | | | | | | | |
| Yes | 39% | 39 | 20 34% | 5.51 | < 0.001 | 7.56 | 0.001 |
| No | 52% | 72 | 8 9% | | | | |
| Unknown | 9% | 14 | 0 | | | | |
| Neuropsychological symptoms or tiredness | | | | | | | |
| Yes | 60% | 77 | 15 16% | 0.72 | 0.433 | 0.61 | 0.319 |
| No | 31% | 37 | 10 21% | | | | |
| Unknown | 9% | 11 | 3 | | | | |
| BMI-per-age percentile > 95 | | | | | | | |
| Yes | 39% | 45 | 15 25% | 2.05 | 0.085 | 1.90 | 0.255 |
| No | 61% | 80 | 13 14% | | | | |
| Age < 3 or age > 13 | | | | | | | |
| Yes | 31% | 37 | 11 23% | 1.54 | 0.318 | 2.42 | 0.100 |
| No | 69% | 88 | 17 16% | | | | |
| Neuromuscular disorder | | | | | | | |
| Yes | 14% | 17 | 4 19% | 1.06 | 0.924 | 3.06 | 0.148 |
| Not indicated | 86% | 108 | 24 18% | | | | |
| Genetic or congenital disorder | | | | | | | |
| Yes | 14% | 16 | 5 24% | 1.48 | 0.482 | 3.71 | 0.065 |
| Not indicated | 86% | 109 | 23 17% | | | | |

All 8 components were jointly significant by logistic regression ($p = 0.0024$). OSA positive, obstructive sleep apnea (AHI > 5); OR, odds ratio.

Figure 1—Results for PM-STOP-Bang.



Mosaic plot showing the percentage in each AHI group by Pediatric-Modified STOP-Bang score. The legend panel on the right shows the colors used for each AHI group and the percentage overall. The width of panels on the horizontal axis is proportional to the sample size of each PM-STOP-Bang score. The colored panels show the percentage of each AHI group separately for each PM-STOP-Bang score.

STOP-Bang. And as STOP-Bang increases the proportion of patients with OSA = severe (red) and the proportion with OSA = moderate (black) is < 11% for those with STOP-Bang scores ≤ 3, and it increases to 20% OSA positive for STOP-Bang = 4 and 58% positive for STOP-Bang = 5 or 6.

Table 3 shows the relationship between each scale value and the sensitivity and specificity. For instance, if STOP-Bang ≥ 6 is used as a cutoff, then 2 subjects were predicted to be positive. Of the 28 actual positives, one had a cutoff ≥ 6 and so the sensitivity was 4% (1/28). Of the 125 actual negatives, all but one had a cutoff < 6, so the specificity was 99% (124/125). If the risk of false positives and false negatives were equal, then the cutoff yielding the largest sensitivity + specificity would be the optimal cutoff. For a cutoff of STOP-Bang ≥ 4, sensitivity was 57% and specificity was 78%.

Analysis of the Components

Each of the individual components was first screened using an unadjusted χ^2 test (Table 2). There were 91 patients with a snoring risk indicator, and 21 of them (23%) were positive for OSA. This is compared to 11% positive for OSA in the group of 62 patients without a snoring risk indicator. Although the odds ratio was large (2.36), it was not statistically significant ($p = 0.064$). The results indicate that the only statistically significant risk factor was sleep obstruction ($p = 0.001$). However

all components had a relative risk value > 1 except for neuropsychological-behavioral symptoms/tiredness (OR = 0.72). A multiple logistic regression analysis was used to test the significance of each of the components of the scales and shows the results for the components of STOP-Bang (adjusted columns in Table 2). Although the test that all 8 components provided predictive value was significant ($p = 0.0024$), only one component was individually significant. The presence of an obstruction was positively related to apnea ($p = 0.001$). Most of the other components had an odds ratio larger > 1 (indicating a nominally positive relationship). However, two components—snoring and neurobehavioral symptoms/daytime tiredness had odds ratios < 1, which indicates that the presence of the component is negatively related to apnea.

DISCUSSION

In this retrospective chart review, specific variables were compared with AHI scores in order to develop a screening tool with a high sensitivity and specificity for pediatric obstructive sleep apnea. The literature indicates that less than half of children with OSA symptoms actually have the syndrome.²⁴ As a result, screening for OSA is challenging and causes many children to go undiagnosed. Presently, pediatric OSA is under-diagnosed and thus undertreated because of the high

Table 3—STOP-Bang scale results.

| STOP-Bang | Percent | Specificity | Sensitivity | True Pos | True Neg | False Pos | False Neg |
|-----------|---------|-------------|-------------|----------|----------|-----------|-----------|
| | | 100% | 0% | 0 | 125 | 0 | 28 |
| 6 | 1% | 99% | 4% | 1 | 124 | 1 | 27 |
| 5 | 11% | 94% | 39% | 11 | 117 | 8 | 17 |
| 4* | 16% | 78% | 57% | 16 | 97 | 28 | 12 |
| 3 | 26% | 48% | 68% | 19 | 60 | 65 | 9 |
| 2 | 25% | 21% | 86% | 24 | 26 | 99 | 4 |
| 1 | 18% | 2% | 100% | 28 | 2 | 123 | 0 |
| 0 | 1% | 0% | 100% | 28 | 0 | 125 | 0 |

*Cutoff yielding the largest sensitivity + specificity. Logistic regression $p = 0.0007$.

Table 4—Revised instrument.

| Risk Indicator | | OSA | | RR | Adjusted | | |
|--|-----|----------|----------|------|----------|---------------|---------|
| | | Negative | Positive | | OR | (95% CI) | p value |
| S Snore | No | 55 | 7 11% | 2.04 | 0.93 | (0.21, 3.82) | 0.9249 |
| | Yes | 70 | 21 23% | | | | |
| T Tonsillar hypertrophy | No | 102 | 20 16% | 1.57 | 2.11 | (0.66, 6.72) | 0.2057 |
| | Yes | 23 | 8 26% | | | | |
| O Sleep obstruction | No | 86 | 8 9% | 3.98 | 7.84 | (2.41, 31.41) | 0.0004* |
| | Yes | 39 | 20 34% | | | | |
| B BMI percent > 85 or < 10 | No | 45 | 4 8% | 2.85 | 4.02 | (1.28, 15.93) | 0.0155* |
| | Yes | 79 | 24 23% | | | | |
| A Age younger than 4, older than 16 | No | 111 | 22 17% | 1.81 | 4.33 | (1.06, 18.34) | 0.0410* |
| | Yes | 14 | 6 30% | | | | |
| N Neuro/Muscular disorder | No | 108 | 24 18% | 1.05 | 3.79 | (0.76, 18.29) | 0.1007 |
| | Yes | 17 | 4 19% | | | | |
| G Genetic/Congenital disorder | No | 109 | 23 17% | 1.37 | 5.03 | (1.19, 21.22) | 0.0289* |
| | Yes | 16 | 5 24% | | | | |

* $p < 0.05$.

cost to test for OSA and the limited number of pediatric sleep laboratories.

Consequently screening for OSA has become essential.²⁵ Canto's recent systematic review and meta-analysis explored the diagnostic value of alternative methods such as clinical history and physical examination to identify pediatric OSA, and also validated the role dentists play in screening patients.²⁶ In the following discussion, the findings of the current study will be compared to the results of Canto's systematic review where applicable. The results of the current study found a clinically significant correlation between the proposed STOP-Bang scale and AHI. However, only one individual component was strongly related to AHI. This suggests that certain variables that present together in a single individual may predispose that person to OSA more than individual parameters.

Below each variable evaluated in this study is dissected along with present findings and suggestions for a revised screening tool based on these results.

STOP-Bang: Snoring is the first component of both the adult STOP-Bang and the PM-STOP-Bang. In a review on sleep disordered breathing in children, Padmanabhan et al. ascertained that snoring, apnea, and difficulty in breathing were the three main symptoms of OSA in children and infants.

Snoring occurs in almost all children with a sleep disorder; often it is the catalyst for parents to believe there is a problem and to pursue a medical evaluation.²⁷ Furthermore, snoring remains the most common complaint in sleep disordered breathing for children under five years old.²¹ However, only a fraction of children who snore have OSA,^{27,28} and the presence of snoring alone cannot accurately predict OSA.⁸ The correlation between snoring and AHI in our study overall had a weak relationship both individually ($p > 0.06$), and once all values were adjusted for, it became nonsignificant ($p > 0.6$). Thus, the presence of snoring does not automatically indicate that the child has OSA.⁸ Young et al. determined that 10% to 14% of children snore at least every other night, and found a prevalence of OSA in 10% to 20% of habitual snorers.²⁹ Our results are similar. One limitation of our study was that snoring was not uniformly described in the medical charts, and so parents may have reported their child snored even when it was infrequent. Also problematic is that parents likely have varying subjective standards for what they consider "snoring" and also vary in their opportunity to observe the behavior. Snoring alone is not a sensitive indicator of OSA, but because it is a prevalent symptom of OSA it remains a useful variable in our screening tool.

STOP-Bang: We used the presence of tonsillar hypertrophy for the T component of our modified STOP-Bang but it did not show a significant correlation with AHI ($p > 0.2$). However, the most common identified risk factor in childhood OSA is adenotonsillar hypertrophy.^{2,31,32} The primary treatment for OSA in children is adenotonsillectomy.^{2,31,32} In Marcus' Childhood Adenotonsillectomy Trial (CHAT), a watchful waiting group was compared to early removal of the tonsils in school-age children. This study found that patient's symptoms overall improved as well as quality of life and polysomnography findings. However surgical treatment did not improve attention or function evaluated through neuropsychological testing.³¹ Like snoring, the presence of large tonsils does not necessarily result in OSA. Several studies have reported that no relationship exists between the size of the tonsil and adenoids and the presence of OSA.^{8,33,34} Canto's systematic review found overall weak results concerning tonsils: with sensitivity = 69% and specificity = 53% for tonsillar hypertrophy and sensitivity = 81% and specificity = 58% for Grade 3 tonsil size.²⁶ Although our study found no relationship with the size of tonsils, it was not always reliably recorded. In this study a little less than half (72 of 153) of the subjects' tonsil size was unable to be determined. These unknowns were recorded as "no" in the data analysis. These limitations likely cause the data in this study to underreport tonsillar hypertrophy. Despite these results and the lack of literature ascertaining tonsillar hypertrophy to predict OSA, it continues to a major cause of OSA² and consequently will remain in the revised screening tool.

STOP-Bang: Sleep obstruction is another common symptom of children with OSA and represented the O in our study. Obstructive apnea occurs when there is respiratory effort and lack of airflow.³⁰ Our results exemplified a strong ordinal relationship with AHI and obstruction ($p = 0.0010$), with a sensitivity of 71% and a specificity of 69%. There is a strong correlation for two reasons; first the obstruction that parents report most likely represent the apnea and hypopnea events significant for OSA, and second, choking and gasping during sleep is a distinct sound that may be definitively distinguished from that of snoring. In this study, obstruction represents the variable with the strongest correlation to AHI and thus remains in our revised screening tool.

STOP-Bang: The P in our screening tool represents neuropsychological-behavioral symptoms in which excessive tiredness and irritability during the daytime was combined with daytime neurobehavioral symptoms. Positive scores of neurobehavioral symptoms required a diagnosis from a medical professional of either ADHD, ADD, or ODD. Daytime hyperactivity and inattention have been shown to be associated with restless sleep and improved sleep patterns have led to positive changes in behavior.^{19,36,37} Relationships between OSA, hyperactivity, and inattentive behavior have been documented.^{19,38–43} Yet excessive tiredness, irritability, and hyperactivity are widely prevalent in children without OSA.^{19,35,44–48} In this retrospective chart review, parents completed the sleep questionnaire for the majority of subjects under the age of 12, their subjective answers were naturally influenced by their own thoughts, feelings, and attitudes on tiredness and their child's irritability. The results of this study indicated no relationship to AHI score and the reporting of excessive tiredness/

irritability ($p > 0.3$). Literature on neurobehavioral symptoms exemplifies a wide range of results. In a study by O'Brien et al., 26% of children with mild symptoms of ADHD were shown to have OSA via a polysomnograph.^{11,49} A more recent study found that in children 6 to 14 years old with ADHD, OSA was not a common underlying disorder or etiologic factor.^{11,50} Yet there is evidence to show persistent sleep disturbance can affect cognition, mood, behavior, and family function.^{11,51} As mentioned previously, the CHAT study ascertained that surgical treatment for OSA in school-age children did not improve attention or executive function.³¹ Canto's review showed attention deficit hyperactivity disorder to have a low sensitivity = 52% and a specificity = 67%.²⁶ Based on the lack of evidence that psychological-behavioral symptoms and reports of tiredness/irritability have significant predictive value, this variable will be omitted in the revised scale.

STOP-Bang: The B in this retrospective chart review denotes body mass index (BMI) percentile for a given age. BMI $\geq 95\%$ indicates an obese child. BMI $\geq 85\%$ to 94% represents children who are overweight. Underweight children are in the BMI $\leq 5\%$ category.⁵² It was proposed that BMI percentiles $> 95\%$ would place a patient at risk for OSA. Obesity has been found to predispose patients to OSA due to the mass loading of upper airway and respiratory muscles, in addition to impairment of ventilation. OSA in obese children ranges from 13% to 36%, based on the severity of obesity.^{53,54} In our study not only are patients above the 85th percentile at risk for OSA, but also patients below the 10th percentile for BMI. So, using these revised cutoffs, BMI-by-age percentile will remain in the revised screening tool.

STOP-Bang: In our study, age presents the A in STOP-Bang and was defined as a risk factor for patients younger than 3 or older than 13. Evidence of systematic variability with age in pediatric OSA is lacking.⁵⁵ Our original age parameters are based on the theory that children younger than 3 may have underdeveloped airways, and patients older than 13 are nearing their full growth potential and may start to develop adult risk factors for OSA such as obesity and high blood pressure. After analysis of the data, a reconsideration of age cutoffs is proposed, as it appears in this study that children younger than 4 and older than 16 are at most risk for OSA, yielding specificity as high as 88% and sensitivity as high as 61%. Thus these changes are taken into account in our revised scale.

STOP-Bang: The N in the modified STOP-Bang screening tool represents neuromuscular disorders related to abnormalities of muscle tone, hypotonia, and spasticity influence a child to have OSA.⁷ The results of this study show a weakly positive relationship to AHI ($p = 0.15$). This study was limited in that there was a very low sample size of patients who had a neuromuscular disorder (only 21 of 153). This low sample size may have prevented a predictive value with AHI. Neuromuscular deficits, along with craniofacial abnormalities and soft tissue hypertrophy, are frequently the origin of airway narrowing.⁵⁵ Although neuromuscular disorders did not show a strong correlation to AHI in this study, it remains in the revised scale as it is cited as one of the main causes of OSA.^{27,29}

STOP-Bang: The G in the modified STOP-Bang represents genetic disorders and congenital disorders and like neuromuscular disorders, did not show a strong correlation to AHI in

the current study ($p = 0.06$). Many of these disorders are the underlying etiology of upper airway obstruction as a result of craniofacial malformation.^{57,58} In the systematic review and meta-analysis by Canto, micrognathia/retrognathia had a sensitivity = 0% with a specificity = 95%. Furthermore, midface hypoplasia overall had a sensitivity = 16% and a specificity = 100%.²⁶ These results ascertain that craniofacial anomalies are not highly predictive of pediatric OSA. Like neuromuscular disorder patients, in our study there was a very small sample size from the data collected—only 21 of 153 subjects had a genetic/congenital disorder. We propose to keep genetic/congenital disorders in the screening tool, as craniofacial anomalies and syndromes were ascertained to be a cause of OSA.²⁷

If the seven revised components mentioned above were used to score the likelihood of OSA, an exploratory multiple logistic regression indicates that more components would have been statistically significant (see Table 4). Even though the p values in the table are not entirely fair, as they are the result of post hoc data mining, it does suggest that the additional factors of BMI risk, age risk, and instances of neuromuscular disorders or genetic/congenital disorders may be important indicators of higher OSA risk. This is consistent with the known subset of children who have the highest risk for OSA including those with underlying abnormalities, such as craniofacial disorders; Down syndrome; cerebral palsy; neuromuscular disorders; chronic lung disease; sickle cell disease; genetic, metabolic, and storage diseases; and laryngomalacia.²³

There were several limitations in this retrospective chart review. The collection of data from the sleep questionnaire proved challenging because many of the sleep questionnaires had inconsistent answers recorded. There remained a lack of verification from the parents reporting and it was not clear whether the patient or parent had filled out the questionnaire. Expectation bias most certainly may have existed in this study, as the examiner was recording the PSG results and also the presence of specific variables that suggest OSA. Prior to 2011, all sleep questionnaires and patient information was recorded in paper charts. These charts were unable to be accessed at the time of data collection; as a result, the sample size was less than originally projected. The sample size does not represent an average population of children as all patients were believed to have a sleep disorder problem and as a result were seeking a diagnosis; thus this study may contain selection bias. Lastly, because this study was retrospective, researchers were limited in what variables could be used in the screening tool as to what information had been previously collected.

There were several variables that this study did not focus on but may be relevant to pediatric OSA. It has been frequently mentioned that there is a genetic component to children with OSA. Future studies may want to include evaluation of whether the parents or siblings currently have a sleep disorder. The siblings of children who have been treated for sleep disorders are more likely have sleep disordered breathing.^{9,59} In addition, children with a family history of OSA are four times more likely to have OSA than children from families with no OSA diagnosis.^{60,61} It is also recommended to define snoring both quantitatively and qualitatively to not score children whose snoring is infrequent and not really suggestive of OSA. Mouth breathing during the daytime (sensitivity = 26%, specificity = 79%) and

during sleep (sensitivity = 68%, specificity = 42%) was evaluated for diagnostic quality in a previous study.²⁶ Despite the mediocre results, mouth breathing is easily diagnosed by dentists and may be a variable useful for screening patients. Ethnicity may also play a role in screening at risk pediatric patients for OSA. Literature cites that being African American is a risk factor^{11,62–64}; however, this was not found in our study. Kheirandish-Gozal et al. found that the prevalence of OSA was increased in poorly controlled asthmatic children⁵⁶; perhaps this variable should be included in future studies. Worthy of attention would be a prospective study in which variables typical of pediatric OSA and commonly diagnosed clinically by dentists could be evaluated to determine a predictive value. Further evaluation is recommended to continue to strive and find a highly predictive screening tool for pediatric OSA.

Polysomnography studies have proven labor intensive and have low availability for children. Moreover, Gozal ascertains that “development of simple, cheap, and reliable diagnostic tools that permit more expanded screening of at-risk populations, and enable accurate identification of the children with definitive disease or with definitive absence of disease would revolutionize the field and provide timely access to clinical care to a large sector of the pediatric population, thereby reducing the health burden of OSA.”²⁵ This study attempted to further clarify which variables were strongly associated with childhood OSA, and thus could be used to develop a screening tool that would accurately predict the disorder in at risk children.

CONCLUSIONS

The purpose of the study was to develop a concise and easy-to-use questionnaire as a screening tool to aid in the recognition of OSA in pediatric patients. The screening scale proposed (PM-STOP-Bang) proved to be predictive of pediatric OSA. Based on the results of this study and the review of the literature the following components are recommended to remain in a revised screening tool: presence of snoring, sleep obstruction, tonsillar hypertrophy; BMI, age, neuromuscular disorders and genetic/congenital disorders. Worthy of attention would be to explore ethnicity factors, presence of asthma, and family history of OSA in future studies.

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DISCLOSURE STATEMENT

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Appendix 1—Data sheet.**Obstructive Sleep Apnea in Children**

Study # _____

Demographics

1. Date of PSG: _____
2. Age at time of PSG (__y__m): _____
(< 4 or > 12 years = 1, otherwise 0)
3. Race (Choose one): Caucasian, African American, Asian, Hispanic
other: _____
4. Gender (M or F): _____

Sleep Center Information

5. Snore (No, Yes, unknown): _____
6. Tonsillar hypertrophy (No, Yes, unknown): _____
7. Obstruction (No, Yes, unknown): _____
8. Daytime neurobehavioral symptoms (No, Yes, unknown): _____
 - a. ICD 314.01 Attention deficit disorder with Hyperactivity
 - b. ICD 314.00 Attention deficit disorder without mention of Hyperactivity
 - c. ODD oppositional defiant disorder
 - d. _____
9. Excessive tiredness/irritability during daytime (No, Yes, unknown): _____
10. Weight: ____lb/kg and Height: ____inches/cm
BMI if provided in chart: _____
BMI calculated by recorders: _____
11. Neuro/Muscular disorder (fill in): _____
12. Genetic/Congenital disorder (fill in): _____
13. Epworth scale: _____
14. AHI score: _____

Appendix 2—VCU Center for Sleep Medicine Questionnaire.

Sleep Questions: Please respond to what extent a statement (item) has been applicable to you during the past 4 weeks. Score each item on a 4-point-scale: 1 (not at all) 2 (somewhat) 3 (rather much) 4 (very much)

Section 1: ____

- | | | | | |
|--|---|---|---|---|
| 1. I am told that I snore. | 1 | 2 | 3 | 4 |
| 2. I sweat during the night. | 1 | 2 | 3 | 4 |
| 3. I am told that I hold my breath when sleeping. | 1 | 2 | 3 | 4 |
| 4. I am told that I wake up gasping for air. | 1 | 2 | 3 | 4 |
| 5. I wake up with a dry mouth. | 1 | 2 | 3 | 4 |
| 6. I wake up during the night while coughing or being short of breath. | 1 | 2 | 3 | 4 |
| 7. I wake up with a sour taste in my mouth. | 1 | 2 | 3 | 4 |
| 8. I wake up with a headache. | 1 | 2 | 3 | 4 |

Section 2: ____

- | | | | | |
|---|---|---|---|---|
| 9. I have difficulty in falling asleep. | 1 | 2 | 3 | 4 |
| 10. Thoughts go through my head and keep me awake. | 1 | 2 | 3 | 4 |
| 11. I worry and find it hard to relax. | 1 | 2 | 3 | 4 |
| 12. I wake up during the night. | 1 | 2 | 3 | 4 |
| 13. After waking up during the night, I fall asleep slowly. | 1 | 2 | 3 | 4 |
| 14. I wake up early and cannot get back to sleep. | 1 | 2 | 3 | 4 |
| 15. I sleep lightly. | 1 | 2 | 3 | 4 |
| 16. I sleep too little. | 1 | 2 | 3 | 4 |

Section 3: ____

- | | | | | |
|--|---|---|---|---|
| 17. I see dreamlike images when falling asleep or waking up. | 1 | 2 | 3 | 4 |
| 18. I sometimes fall asleep on a social occasion. | 1 | 2 | 3 | 4 |
| 19. I have sleep attacks during the day. | 1 | 2 | 3 | 4 |
| 20. With intense emotions, my muscles sometimes collapse during the day. | 1 | 2 | 3 | 4 |
| 21. I sometimes cannot move when falling asleep or waking up. | 1 | 2 | 3 | 4 |

Section 4: ____

- | | | | | |
|--|---|---|---|---|
| 22. I am told that I kick my legs when I sleep. | 1 | 2 | 3 | 4 |
| 23. I have cramps or pain in my legs during the night. | 1 | 2 | 3 | 4 |
| 24. I feel little shocks in my legs during the night. | 1 | 2 | 3 | 4 |
| 25. I cannot keep my legs at rest when falling asleep. | 1 | 2 | 3 | 4 |

Section 5: ____

- | | | | | |
|--|---|---|---|---|
| 26. I would rather go to bed at a different time. | 1 | 2 | 3 | 4 |
| 27. I go to bed at very different times (more than 2 hr difference). | 1 | 2 | 3 | 4 |
| 28. I do shift work. | 1 | 2 | 3 | 4 |

Section 6: ____

- | | | | | |
|---|---|---|---|---|
| 29. I sometimes walk when I am sleeping. | 1 | 2 | 3 | 4 |
| 30. I sometimes wake up in a different place than where I fell asleep. | 1 | 2 | 3 | 4 |
| 31. I sometimes find evidence of having performed an action during the night I do not remember. | 1 | 2 | 3 | 4 |

Section 7: ____

- | | | | | |
|---|---|---|---|---|
| 32. I have frightening dreams (if not, go to Item 37). | 1 | 2 | 3 | 4 |
| 33. I wake up from these dreams. | 1 | 2 | 3 | 4 |
| 34. I remember the content of these dreams. | 1 | 2 | 3 | 4 |
| 35. I can orientate quickly after these dreams. | 1 | 2 | 3 | 4 |
| 36. I have physical symptoms during or after these dreams (e.g., movements, sweating, heart palpitations, shortness of breath). | 1 | 2 | 3 | 4 |

Section 8: ____

- | | | | | |
|--|---|---|---|---|
| 37. It is too light in my bedroom during the night. | 1 | 2 | 3 | 4 |
| 38. It is too noisy in my bedroom during the night. | 1 | 2 | 3 | 4 |
| 39. I drink alcoholic beverages during the evening. | 1 | 2 | 3 | 4 |
| 40. I smoke during the evening. | 1 | 2 | 3 | 4 |
| 41. I use other substances during the evening (e.g., sleep or other medication). | 1 | 2 | 3 | 4 |
| 42. I feel sad. | 1 | 2 | 3 | 4 |
| 43. I have no pleasure or interest in daily occupations. | 1 | 2 | 3 | 4 |

Section 9: ____

- | | | | | |
|--|---|---|---|---|
| 44. I feel tired at getting up. | 1 | 2 | 3 | 4 |
| 45. I feel sleepy during the day and struggle to remain alert. | 1 | 2 | 3 | 4 |
| 46. I would like to have more energy during the day. | 1 | 2 | 3 | 4 |
| 47. I am told that I am easily irritated. | 1 | 2 | 3 | 4 |
| 48. I have difficulty in concentrating at work or school. | 1 | 2 | 3 | 4 |
| 49. I worry whether I sleep enough. | 1 | 2 | 3 | 4 |
| 50. Generally, I sleep badly. | 1 | 2 | 3 | 4 |

Efficacy versus Effectiveness in the Treatment of Obstructive Sleep Apnea: CPAP and Oral Appliances

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Obstructive sleep apnea (OSA) is a chronic disorder and effective long-term treatment is necessary to prevent associated health risks. Standard treatment remains continuous positive airway pressure which is highly efficacious but has well-recognized limitations, with suboptimal patient acceptance and adherence rates, which in turn obviates the desired health benefits. The leading alternative device treatment is oral appliances. Patients often report preferring oral appliances to CPAP treatment, with better usage rates. However, unlike CPAP, inter-individual variability in the efficacy of oral appliance therapy means that patients are often left with some residual OSA. Despite discrepancies in efficacy (apnea-hypopnea index [AHI] reduction) between CPAP and oral appliances, randomized trials show similar improvements in health outcomes between treatments, including sleepiness, quality of life, driving performance, and blood pressure. Similar results in terms of health outcomes suggests that although the two treatments have different efficacy and treatment usage profiles, these result in similar overall effectiveness. In this narrative review, we discuss efficacy versus effectiveness in relation to CPAP and oral appliance treatment of OSA.

KEYWORDS: obstructive sleep apnea, treatment effectiveness, efficacy, CPAP, oral appliances

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Efficacy and effectiveness are important concepts to distinguish when evaluating treatment performance. Treatment efficacy refers to how well an intervention works under ideal circumstances whereas, effectiveness is how well an intervention performs in the real world where conditions are not controlled. Therefore treatment effectiveness is particularly important in management of chronic disease. Obstructive sleep apnea (OSA) is a common sleep disorder characterized by repetitive upper airway obstruction leading to intermittent hypoxia and sleep fragmentation. There has been a dramatic increase in OSA prevalence over the last two decades, attributable to the obesity epidemic, with at least moderate OSA now evident in 17% of middle-aged men and 9% of middle-aged women.¹ OSA is associated with excessive daytime sleepiness and lower quality of life as well as increased risk of workplace and motor vehicle accidents, hypertension and cardiovascular disease, type 2 diabetes, and all-cause mortality.^{2–9} Therefore effective management of this chronic disorder is imperative to not only improve symptoms but to prevent long-term health risks. Standard care is the highly efficacious treatment, continuous positive airway pressure (CPAP). This therapy involves delivery of pressurized air to the upper airway during sleep via a nasal mask interface and tube connected to a pump. The pressurized air acts to splint open the upper airway preventing it from collapsing during sleep. The effectiveness of this therapy is therefore dependent upon its ability to overcome airway collapse (efficacy) as well as the time course over which a patient applies it during sleep (compliance). While the efficacy of CPAP is generally high, in the real world long-term health effects of CPAP are likely to be compromised by low compliance and suboptimal hours of treatment use. Treatment usage as a proportion of the total sleep period when a patient is vulnerable to OSA is often overlooked as a confounder of efficacy.

However, treatment usage compared to sleep time is an important aspect of real-world effectiveness. Importantly, treatment effectiveness warrants consideration when comparing effects of other OSA treatment options which may not have the same level of efficacy as CPAP but may have a better usage profile. In this review we discuss efficacy and effectiveness between first line OSA treatment CPAP and the leading alternative device treatment, oral appliances.

EFFICACY VERSUS EFFECTIVENESS IN OSA

Efficacy, in the context of OSA, reflects the ability of treatment to prevent the occurrence of obstructive breathing events during periods when the treatment is being physically applied. This is assessed by the number of obstructive breathing events *per hour of sleep* or apnea-hypopnea index (AHI). An AHI < 5 events/h indicates absence of disease or a completely efficacious treatment. In a fully compliant patient (using treatment for 100% of sleep time) efficacy measured as AHI on treatment (AHI_{Treatment}) will give an accurate reflection of OSA treatment effectiveness. However sleep time off treatment becomes an important consideration when compliance is suboptimal. The potential impact of suboptimal CPAP compliance on AHI has been considered using formulas that adjust AHI_{Treatment} for sleep time off treatment when AHI can presumably revert to untreated levels (AHI_{Untreated}).^{10,11} When the untreated portion of the night with OSA reoccurrence is taken into consideration, CPAP effectiveness can dramatically decrease depending on OSA severity and total sleep time. Good CPAP adherence is generally set at a benchmark of 4 h/night; however, the rationale for this benchmark is not overly evidence based. Moreover when taking into consideration sleep time off treatment, 4 h of CPAP use during an 8-h sleep period may only reduce

the AHI by 50% due to reoccurrence of moderate OSA during the remaining 4 h without CPAP.¹⁰ In this case, the true AHI is poorly represented by $AHI_{\text{Treatment}}$. It has therefore been proposed that treatment comparisons should be made on overall effectiveness after adjustment of efficacy for hours of usage over total sleep time.¹² In this context, although other OSA treatments such as surgery and oral appliances may be less efficacious, they offer more favorable compliance profiles (100% in the case of surgery), which may be an important determinant of the overall effectiveness, and may correlate more strongly with downstream health outcomes.

CPAP COMPLIANCE AND EFFECTIVENESS

Adequate CPAP compliance, based on reported average usage rates, is generally accepted as > 4 h on $\geq 70\%$ of nights.¹³ However, even with strategies to enhance patient acceptance and usage, only ~50% of patients use CPAP ≥ 4 h per night after 6 months.¹⁴ The proportion of patients maintaining this minimally acceptable level of CPAP usage further drops to 17% after 5 years.¹⁵ Furthermore this 4-h threshold is arbitrary and not necessarily adequate to convey benefits for *all* important health outcomes. In reality, a dose response relationship has been observed between hours of CPAP use and a range of subjective and objective health benefits with differing benefit thresholds for different outcomes.^{16–18} For example, normalization of subjective sleepiness (ESS), objective sleepiness (multiple sleep latency test), and disease specific functional status (functional outcomes of sleep questionnaire [FOSQ]) requires 4, 6, and 7.5 h, respectively, of nightly CPAP usage.¹⁸ In hypertensive OSA patients, ≥ 5.6 h of CPAP usage is required to sustain a long-term reduction in blood pressure.¹⁹ CPAP usage > 6 h per night shows greatest reduction in mortality risk.²⁰ Therefore to maximize treatment benefits for all important health outcomes, CPAP needs to be consistently used for the majority, if not all, of the sleep period. Given that this is generally not a reality for most CPAP users, there is a clear rationale for conducting comparative effectiveness trials against alternative less efficacious treatments which may still be equally effective at improving health outcomes due to higher compliance rates.

ORAL APPLIANCES IN TREATMENT OF OSA

Oral appliances are the leading device alternative to CPAP. Oral appliances cover the upper and lower dental arches and are configured so that the lower jaw is held forward in a more protruded position. The action of mandibular advancement results in an increase in pharyngeal airway space and reduces airway collapsibility.^{21,22} Oral appliances have a demonstrated role in improving snoring, obstructive apneas and hypopneas, and oxygen desaturation measures.²³ Oral appliances also have demonstrated benefit on health outcome measures such as daytime sleepiness and blood pressure.^{23,24} However unlike CPAP which will prevent airway collapse in most people as long as sufficient pressure is applied, therapeutic response to oral appliance treatment shows intra-individual variability. In general terms, over a third of patients will show a complete response to oral appliance therapy with a reduction in AHI to < 5/h (or no OSA). Another third will have

a clinically important response showing > 50% reduction in AHI,²⁵ although AHI remains > 5/h and a third will not achieve > 50% reduction in AHI. There are many factors which may contribute to differences in therapeutic response to oral appliance therapy including differences in devices and treatment protocols but also craniofacial, upper airway, and obesity characteristics of the patient.²⁵ Currently there is no validated clinical method to reliably pre-select patients who will receive sufficient benefit from oral appliance therapy from those who show minimal therapeutic response. Uncertainty around efficacy has essentially restricted oral appliance implementation to milder cases of OSA with consideration only in more severe OSA if CPAP fails.²⁶

COMPARISON OF HEALTH EFFECTS OF CPAP AND ORAL APPLIANCE THERAPY

Although CPAP is clearly superior to oral appliances in terms of eliminating obstructive breathing events and improving nocturnal oxygen saturation,²⁷ this is not the case for health outcomes. In randomized controlled trials comparing CPAP to oral appliance treatment, CPAP consistently demonstrates normalization of AHI, whereas average AHI remains in the range of mild OSA on oral appliance treatment.^{28–35} However the superiority of CPAP in terms of efficacy is generally not carried through to the actual health outcomes of treatment. A summary of randomized controlled trials comparing CPAP and oral appliances with commonly reported health outcomes is summarized in Table 1. Subjective daytime sleepiness, assessed by the Epworth Sleepiness Scale, does not differ following CPAP and oral appliance treatment.³⁶ This has also been shown in objective tests of sleepiness^{32,37} and simulated driving performance.^{35,38} Furthermore, in terms of cardiovascular outcomes there is no demonstrated difference between treatments in short-term effects on blood pressure.^{29,34,35} In a small study both CPAP and oral appliances were found to improve endothelial function to the same degree.³⁹ To date short-term treatment studies comparing CPAP and oral appliance overall consistently show minimal to no difference in health outcome measures despite demonstrating a higher $AHI_{\text{Treatment}}$ with oral appliances. Longer term studies are lacking, although a recent 6-year observational study of untreated and treated (either CPAP or oral appliance) OSA patients found OSA treatment reduced the cardiovascular mortality rates regardless of whether CPAP or oral appliance treatment was used.⁴⁰

A likely explanation for similarity in key health outcomes is that oral appliances are more consistently used for a greater proportion of the total sleep period, compared to CPAP. Greater usage may counterbalance the lower treatment efficacy and result in overall equivalent treatment effectiveness. Oral appliances were preferred to CPAP in four of six crossover trials asking for treatment preference at the end of the trial.^{30–32,35} This preference for oral appliance treatment may translate to significantly more hours of usage. A review of reported treatment times in oral appliance studies suggests usage remains at a median of 77% of nights after one year of treatment.⁴¹ However, it has been possible to objectively verify CPAP usage by data download for many years, while comparison to oral appliance

Table 1—Efficacy and effectiveness of oral appliances versus CPAP: AHI and health outcome results from randomized trials.

| Study | Study Design | N | Baseline AHI | Treatment AHI | | Health Outcomes | | | |
|-----------------|--------------|-----|-------------------------|----------------------|-----------------------|--------------------|------------|--------------------------------|----------------|
| | | | | CPAP | OA | Daytime Sleepiness | | Health-Related Quality of Life | Blood Pressure |
| | | | | | | Subjective (ESS) | Objective | | |
| Aarab, 2010 | parallel | 57 | 20.9 ± 9.8 | 1.4 ± 13.1 | 5.8 ± 14.9 | ↔ | N/A | ↔ | N/A |
| Barnes, 2004 | crossover | 80 | 21.5 ± 1.6 | 4.8 ± 0.5 | 14.0 ± 1.1 | ↔ | ↔ (MWT) | N/A | ↔ |
| Engleman, 2002 | crossover | 48 | 31 ± 26 | 8 ± 6 | 15 ± 16 | CPAP | CPAP (MWT) | CPAP | N/A |
| Ferguson, 1997 | crossover | 20 | 26.8 ± 11.9 | 4.0 ± 2.2 | 14.2 ± 14.7 | ↔ | N/A | N/A | N/A |
| Gagnadoux, 2009 | crossover | 59 | 34 ± 13 | 2 (1–8) [#] | 6 (3–14) [#] | ↔ | ↔ (OSLER) | OA | N/A |
| Hoekema, 2008 | parallel | 103 | 40.3 ± 27.6 | 2.4 ± 4.2 | 7.8 ± 14.4 | ↔ | N/A | ↔ | N/A |
| Lam, 2007 | parallel | 101 | 23.8 ± 1.9 [^] | 2.8 ± 1.1 | 10.6 ± 1.7 | CPAP | N/A | CPAP | ↔ |
| Phillips, 2013 | crossover | 108 | 25.6 ± 12.3 | 4.5 ± 6.6 | 11.1 ± 12.1 | ↔ | N/A | ↔ | ↔ |
| Tan, 2002 | crossover | 21 | 22.2 ± 9.6 | 3.1 ± 2.8 | 8.0 ± 10.9 | ↔ | N/A | ↔ | N/A |

[#]Median (interquartile range). [^]Mean ± standard error. Summary of AHI data with CPAP and oral appliances (OA) in randomized trials comparing treatments. Summary of commonly reported health assessments are presented. “CPAP” or “OA” indicates superior results were found with that treatment, ↔ indicates equivalent findings observed with both treatments. AHI data is mean ± standard deviation, unless otherwise indicated. ESS, Epworth Sleepiness Score; MWT, maintenance of wakefulness test; OSLER, oxford sleep resistance test.

usage has been limited to self-report until recently. Therefore, even though self-reported oral appliance usage appears to exceed that of objectively downloaded CPAP usage, it has been difficult to compare usage profiles between therapies. The recent advent of objective compliance monitors for oral appliances in the form of small embedded temperature-sensing chips⁴² now makes verification of usage patterns possible. Initial studies of objective oral appliance usage confirm good usage of > 7 hours a night in the initial 3 months of oral appliance treatment⁴² which is maintained at > 6 hours per night after one year.⁴³ Furthermore the discrepancy of over an hour between subjective and objective CPAP usage¹³ does not seem to be apparent with oral appliance treatment, with initial studies reporting < 30 minutes difference between subjective estimates and objective data.⁴³ Regardless, initial evidence from oral appliance compliance monitors lends support to greater usage of oral appliance therapy than CPAP.

SLEEP ADJUSTED RESIDUAL AHI (SARAH INDEX) FOR ASSESSMENT OF TREATMENT EFFECTIVENESS

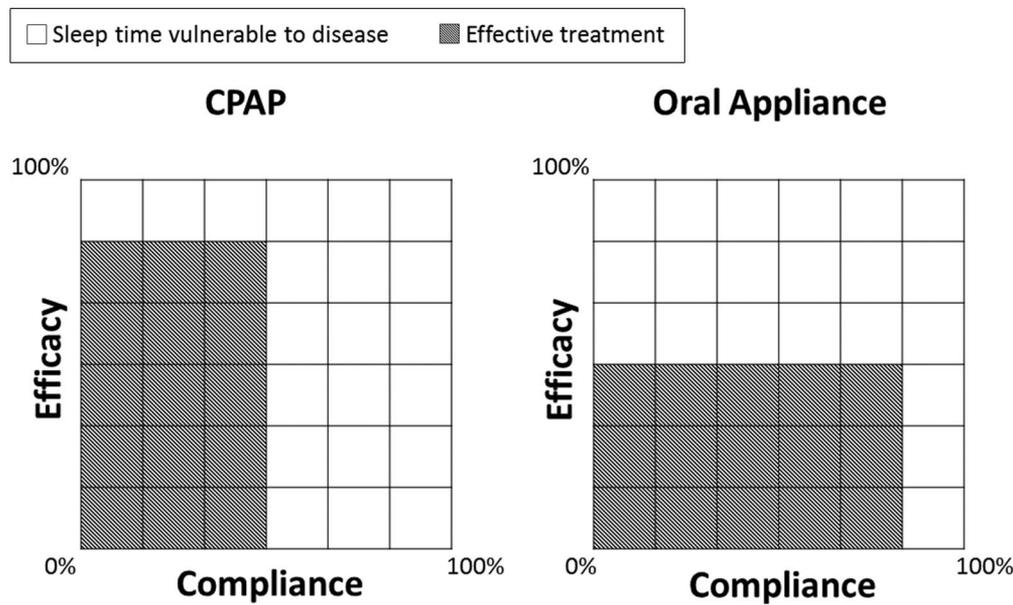
Evidence of equivalent health outcomes between oral appliances and CPAP suggest that real-world treatment effectiveness is not captured by the efficacy measure $AHI_{Treatment}$. However this is the metric on which clinical decisions are primarily made, although it is well recognized that CPAP is not used for all hours of sleep. The different treatment profiles of CPAP (high efficacy/low adherence) and oral appliances (moderate efficacy/high adherence) may conceptually result in similar profiles of treatment effectiveness. In the schematic in Figure 1, two identical sleep periods in which OSA can occur is represented by a grid (white boxes) for which CPAP and oral appliance are applied. Treatment effectiveness is a composite of efficacy (represented on the y axis of the grid) and hours of

treatment usage (represented on the x axis). In this example MAS is only half as efficacious as CPAP, but compliance is two-fold greater. Despite these different treatment profiles, both treatments have similar overall effectiveness in relieving OSA (shaded area). This conceptual example likely reflects many patients in the real world, for whom CPAP is highly efficacious but treatment usage is modest, while oral appliances may have more modest efficacy but are used for relatively more of the sleep period. Potentially a more representative measure of treatment effectiveness than $AHI_{Treatment}$ should also take into account hours ON treatment ($AHI_{Treatment}$) and hours OFF treatment ($AHI_{Untreated}$) for the TOTAL sleep period. We adopt the formula of Ravesloot and colleagues,¹² which accounts for these additional factors in order to assess a more accurate measure of treatment effectiveness, which we have called the Sleep Adjusted Residual AHI or SARAH Index. Potentially such an index which incorporates these currently overlooked factors could be a more accurate measure of treatment effectiveness and will better align with downstream health benefits. The formula is expressed below:

$$\text{Sleep Adjusted Residual AHI (SARAH Index)} = \frac{[AHI_{Treatment} \times \text{Hours}_{Treatment}] + [AHI_{Untreated} \times \text{Hours}_{Untreated}]}{\text{Hours}_{\text{Total Sleep Time}}}$$

COMPARISON OF AHI AND SLEEP ADJUSTED RESIDUAL AHI (SARAH INDEX) IN CPAP AND ORAL APPLIANCE TREATMENT

We have previously published a large cross-over study (108 completers) of one month each of optimized CPAP and oral appliance treatments.³⁵ This study found that oral appliances were non-inferior to CPAP across a range of health outcomes in predominantly moderate-severe patients. There were no between-treatment difference in cardiovascular (24-h

Figure 1—Comparison of treatment effectiveness profile of CPAP and oral appliances.

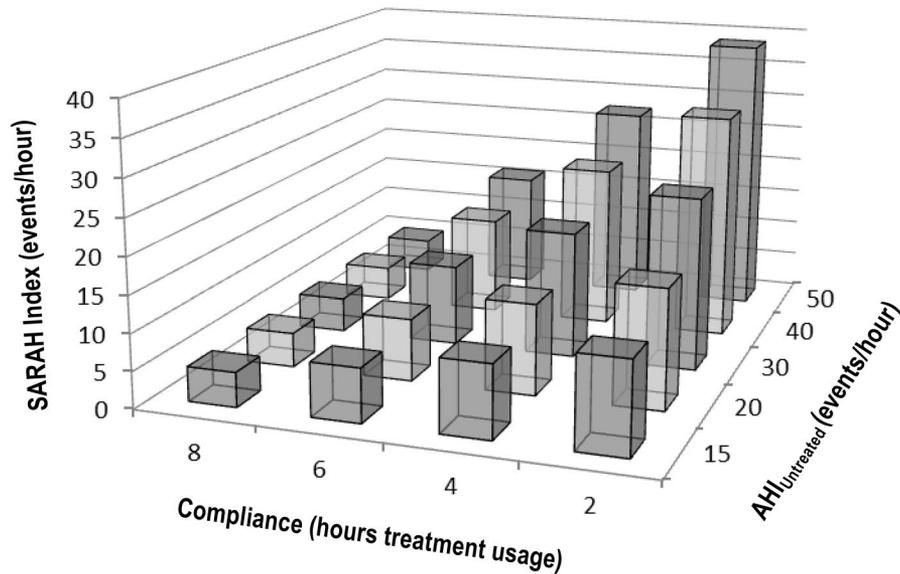
Efficacy (y axis) reflects the ability of treatment to prevent obstructive breathing events when it is physically applied. Compliance (x axis) reflects the hours the treatment is applied for over the total sleep time when obstructive events can occur. “Effectiveness” requires both efficacy and compliance and the balance of these likely reflects over health outcomes. This schematic illustrates the scenario of an oral appliance which is only half as efficacious as CPAP but has two-fold greater compliance which results in equivalent effectiveness (shaded area).

blood pressure, arterial stiffness), neurobehavioral (subjective sleepiness, driving simulator performance), or quality of life outcomes. In a subgroup of hypertensive patients, blood pressure during sleep reduced from baseline with both treatments, but more importantly, with no difference between them. In comparing the efficacy profiles of the two treatments, as expected, polysomnography confirmed OSA resolution on CPAP, whereas residual mild OSA was evident with oral appliance treatment ($AHI\ 4.5 \pm 6.6$ vs. $11.1 \pm 12.1/h$). However, self-reported compliance favored oral appliances at an average 1.3 h more usage per night than CPAP. These efficacy and compliance profiles of CPAP and oral appliance treatment suggest that superior CPAP efficacy may be offset by greater oral appliance usage. We now use real data from this trial to compare AHI and SARAH Index between CPAP and oral appliance treatments across the spectrum of OSA severity.

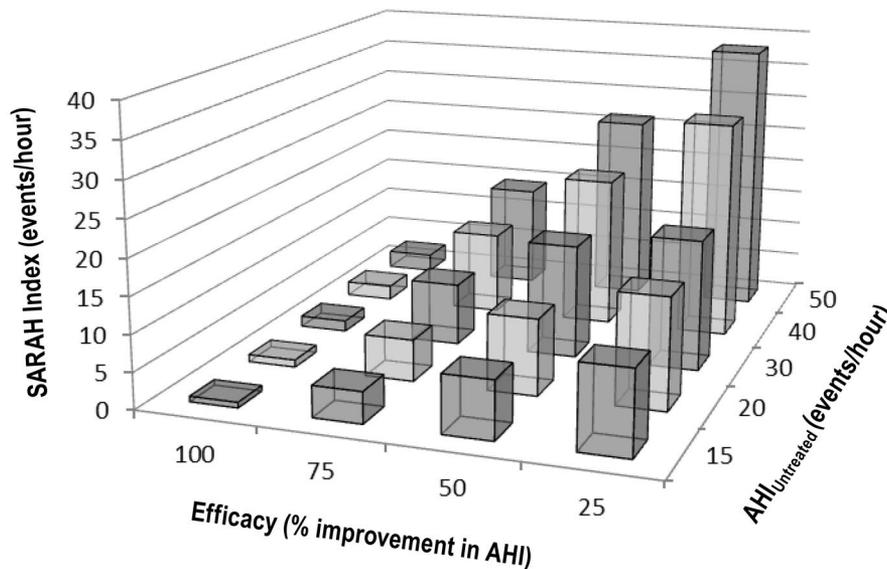
Median treatment AHI on CPAP from this trial was 4.7/h (i.e., elimination of OSA). We have used $AHI_{Treatment}$ of 4.7/h to calculate the SARAH Index at different levels of treatment usage hours for an 8-h sleep period (healthy sleep time range⁴⁴). Figure 2 shows the results from calculation of SARAH Index across a range of OSA severity ($AHI_{Untreated}$). If CPAP is used for the total 8-h sleep, OSA is indeed resolved ($AHI = 4.7$) for all levels of OSA severity. However, it is recognized that as many as 50% of CPAP treated patients are using their treatment < 4 h of total sleep time.¹⁵ Using this example of an 8-h sleep period, the graph demonstrates that patients using their device for 4 and 2 h per night have at least mild OSA assessed by the SARAH Index, with much higher levels in those with more severe OSA. As total sleep time decreases, the SARAH Index reduces; however, for an average 8-h sleep period, the majority of CPAP users would be effectively under-treated based on

known compliance rates. As CPAP usage further declines long term, CPAP treatment effectiveness may additionally become worse over time. This graph illustrates that when taking into consideration CPAP hours used over sleep time, OSA may not be well controlled, and even moderate-severe OSA may still be present in more severe and less compliant patients who sleep for longer periods. The SARAH Index calculation raises the possibility that despite high efficacy, CPAP users may not be effectively treated in practice.

Oral appliance usage data from this same trial³⁵ found median reported usage time to be 95% of total sleep time. We have used this 95% compliance rate to assess oral appliance treatment effectiveness by the SARAH Index. With good self-reported usage of nearly 100% of sleep time the influencing factor on treatment effectiveness for oral appliances is their efficacy, expressed as a percentage improvement in OSA from baseline levels. We show SARAH Index for different OSA severities across different levels of oral appliance efficacy of 25%, 50%, 75%, and 100% improvement in Figure 3. Oral appliance treatment effectiveness expressed by SARAH Index varies with efficacy and OSA severity. We have shown in a large audit of oral appliance treated patients that the majority (70%) will have $\geq 50\%$ improvement in OSA using an oral appliance.⁴⁵ If we compare Figures 2 and 3, CPAP and oral appliance treatment effectiveness measured by the SARAH Index, conceptually we can see that many patients may be effectively undertreated with either treatment. However, with half of all CPAP treated patients using it < 4 h per night and two-thirds of oral appliance treated patients reducing OSA by at least half, theoretically many patients with incomplete efficacy on oral appliance may be no worse off than when on fully efficacious CPAP in terms of treatment effectiveness.

Figure 2—CPAP effectiveness assessed by the Sleep Adjusted Residual AHI (SARAH Index).

This figure illustrates SARAH Index for different levels of OSA severity ($AHI_{Untreated}$) for varying hours of treatment usage for an average 8-h sleep time. An $AHI_{Treatment}$ of 4.7 events/h is used (elimination of OSA). When taking into consideration CPAP hours used over sleep time, OSA may not be well controlled in moderate-severe patients using CPAP 4 hours or less for 8 h of sleep.

Figure 3—Oral appliance effectiveness assessed by the Sleep Adjusted Residual AHI (SARAH Index).

In contrast to CPAP, oral appliance hours of usage are reported to be high (95% of sleep time). However efficacy levels are variable with oral appliances. This figure illustrates SARAH Index for different levels of efficacy (% improvement in AHI). The majority of patients have 50% or greater improvement in $AHI_{Treatment}$ using an oral appliance. Therefore by SARAH Index calculation, many patients may not be worse off on oral appliance treatment despite $AHI_{Treatment} > 5/\text{hour}$ compared to CPAP used for minimal hours compared to total sleep time.

POTENTIAL CONFOUNDERS OF EFFECTIVENESS CALCULATION

Although treatment efficacy is not an adequate indicator of health benefit, effectiveness measures, such as the calculation presented as the SARAH Index, also have potential limitations. The formula assumes that OSA will return to baseline levels once treatment is removed before the end of the sleep period.

Withdrawal of CPAP results in return of OSA.^{46–48} However, short-term carryover effects after CPAP removal may occur resulting in reduced OSA despite being without treatment. Sustained effects of CPAP may be due to an ongoing increase in pharyngeal volume and airflow due to reduced soft tissue edema as a consequence of CPAP use.^{49,50} The evidence for existence and duration of CPAP washout effects has been recently reviewed.⁵¹ Studies re-assessing OSA after CPAP

withdrawal for several nights to weeks find lower AHI levels than recorded at baseline, potentially more evident in severe OSA patients,⁴⁸ although this is not always observed.^{47,52,53} Regardless of baseline severity, AHI does appear to deteriorate between the first and seventh night of CPAP withdrawal.⁵⁴ Furthermore, although some CPAP washout effect is observed in studies, the extent and duration is highly variable and potentially confounded by issues of night to night variability in measurement of sleep-disordered breathing.^{55,56} In particular, it is unknown whether such a phenomenon occurs within a single night. In terms of oral appliances, OSA levels return to baseline after a week of a placebo oral appliance (no active advancement).⁵⁷ However residual effects of mandibular advancement once the lower jaw returns to normal position, or a washout effect, may be less plausible with oral appliances than CPAP.

This effectiveness assessment also does not take into account differences in OSA severity due to body position and sleep stage. OSA may become more severe in the supine position and REM sleep and treatment effectiveness, particularly of oral appliances, may vary under these conditions.⁴⁵ CPAP removal after several hours may leave the patient exposed to the portion of the night with more concentrated REM sleep, and hence more severe OSA. Treatment carryover effects and OSA variability due to body position and sleep stage are not captured in the simple assessment of time on versus off treatment at $AHI_{\text{Treatment}}$ and AHI_{Baseline} , and would be difficult to do so routinely. However, whether this approximation of effectiveness will be more clinically useful than relying only on a potentially false reassurance of $AHI_{\text{Treatment}}$ needs further assessment. If proven to give a more reliable measure of effectiveness, another obstacle to adopting an index such as SARA Index would be related to technological limitations with estimating sleep time in the home setting. Although the growing adoption of lifestyle wearable devices that monitor aspects of sleep may prove useful in this regard.

CONCLUSIONS AND FUTURE DIRECTIONS

Although effectiveness, as a combined measure of real world usage and efficacy, is difficult to accurately assess, proposed formulas which account for sleep time on and off treatment potentially may be a more accurate marker of health outcome responses. However this remains to be assessed in prospective trials. There is limited evidence of comparative effectiveness of CPAP and oral appliance treatments longer-term. If equivalent short-term health outcomes are found to be sustained in the long term, this opens up treatment options for patients with this chronic disease. Comparative-effectiveness and Patient-Centered Outcomes Research aims to help patients (and their healthcare providers) to make informed decisions about health and healthcare options base on outcomes that are important to them.⁵⁸ We propose a greater emphasis on treatment effectiveness rather than efficacy as part of a chronic disease management approach. Future comparative effectiveness research of CPAP and Oral appliance treatment could allow patients more freedom to choose their preferred treatment over all aspects of treatment effectiveness and health outcomes.

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The American Board of Dental Sleep Medicine Announces Significant Changes for the 2016 Exam and Case Presentation

Nancy Addy, DDS, Diplomate, President ABDSM

Leawood, KS

The directors of the American Board of Dental Sleep Medicine (ABDSM) have announced substantial changes to the Diplomate exam and case presentation that will make becoming a Diplomate more accessible and convenient. The ABDSM has transitioned to a computer-based exam, extended the timelines in the application process, broadened requirements for case studies and shortened the suggested reading list.

As dental sleep medicine has gained increasing recognition from the medical community, I have seen ABDSM certification grow in importance and demand, with 60 candidates sitting for and passing the 2015 board exam in Seattle. The ABDSM exam and case presentation certifies a thorough knowledge in dental sleep medicine, and I believe the significant changes for the 2016 exam and case presentation process create a path to certification that is more accessible and more convenient for those interested in growing their dental sleep medicine practice.

A Worthy Investment

Established in 2004, the ABDSM is an independent, nonprofit, self-designated board that certifies licensed dentists who treat sleep-related breathing disorders. Earning certification, or “Diplomate status,” by the ABDSM allows dentists to demonstrate their proficiency in dental sleep medicine to patients, physicians and insurers. Diplomate status is more than just an opportunity to distinguish yourself and prove your expertise in the field of dental sleep medicine. A Diplomate of the ABDSM meets a quality benchmark of care that is nationally recognized not only by the American Academy of Dental Sleep Medicine (AADSM) but also recognized and supported by the physicians of the American Academy of Sleep Medicine (AASM).

Significant Changes in 2016

The changes to the Certification Guidelines were implemented to improve the quality of the test itself, while also creating a better experience for applicants. The changes below are grouped into three main categories: convenient locations, expanded timelines and adjusted requirements.

Convenient Locations

Starting in 2016, the ABDSM exam will no longer be given in conjunction with the AADSM annual meeting. Instead, the exam will be computer based and available at hundreds of testing centers across the U.S. and Canada. This change offers convenience and cost-savings to dentists interested in sitting for the exam. Applicants will now have the flexibility of using Kryterion Global Testing Solutions to choose a testing center near their home and selecting a convenient exam date and time during the two-week testing window of April 10-23, 2016. The

proctored, 200-question exam will still be a mix of multiple choice and true or false questions with an allocated time of four hours.

Extended Timelines

New extended dates provide candidates more time to complete case studies, which can be submitted after sitting for the exam. The application process will start in the fall each year and is open this year from Oct. 1 to Nov. 16, 2015. Once the application is submitted, applicants will have an extended period of 18 months to submit their 15 case studies. In addition, to help applicants achieve the required 50 continuing education (CE) hours within three years prior to applying, candidates are allowed to submit up to 15 approved CE credits after the application deadline. Applicants for the 2016 exam have until March 1, 2016 to submit these 15 CE credits.

Adjusted Requirements

Revisions also have been made to improve the quality of the ABDSM Board Exam and provide flexibility for dentists when collecting required data from physician partners.

- **Acceptance of RDI Measurements:** Case studies can now use either the apnea/hypopnea index (AHI) or respiratory disturbance index (RDI) to quantify sleep apnea severity, as long as the same measure is used for both pre- and post-treatment sleep studies.
- **Unlimited Sleep Center Options:** The new Certification Guidelines now allow dentists to work with any sleep center to fulfill Diplomate requirements. This offers dentists more opportunities to collaborate with any sleep center.
- **Condensed Recommended Reading List:** The recommended reading list has been shortened considerably to approximately 50 of the most recent and relevant articles, the vast majority of which will be made available to those who take the AADSM’s Board Review Course.

Starting the Journey to Diplomate Status

All journeys start with a single step. Being a Diplomate of the ABDSM distinguishes you as a professional in the rapidly growing and rewarding field of dental sleep medicine. I encourage you to take your first step by learning about the certification process. Being aware of the nuances required can help you reach this prestigious designation smoothly. You can begin by treating each patient as a potential board case, and each CE hour as an investment in your knowledge of dental sleep medicine (see tips in Figure 1). Once you have started down the path, you will quickly find that you have accumulated

Figure 1**Board Case Requirement Tips**

- The five detailed case studies will require multiple *pretreatment* intraoral photographs showing the patient's dentition using lip and cheek retractors. Taking these photographs for each patient who has a pre-treatment AHI or RDI of 10 or greater will increase your options when selecting cases for submission.
- The detailed case studies require documentation of at least three follow-up appointments, the third appointment being at least three months post oral appliance insertion. Continue to document follow-up visits by taking thorough SOAP notes in the patient record so you only have to make copies of these notes for cases that qualify as a detailed case.
- Templates and other resources developed to assist you in putting together your application can be found on abdsm.org.
- Diagnostic sleep studies for all 15 board cases must have been conducted no more than five years prior to when the oral appliance was delivered.

Tips for Earning CE Hours

- 50 ADA CERP recognized or AGD PACE approved continuing education hours are required to sit for the exam, and 35 need to be earned before applying.
- Of the 50 required CE credits, up to 10 hours may be AMA PRA Category 1 Credits in sleep medicine.
- The AADSM has many ongoing education offerings to help fulfill this requirement. There are two opportunities in particular that I recommend to potential applicants: the AADSM Annual Meeting and the Board Review Course. Combined, these two events enable you to earn more than half the needed hours in just a few days.
- All 50 CE hours must further your clinical knowledge in dental sleep medicine. CE credits from practice management or billing courses, for example, don't help improve your understanding of dental sleep medicine and will not be accepted.

both the knowledge and the experience you need to sit for the ABDSM Board Exam and present your cases.

Diplomate certification has the power to help build a firm foundation for the ongoing success of your dental sleep medicine practice, and it also plays an important role in building the reputation of dental sleep medicine nationwide. I encourage anyone who is considering applying this fall for the 2016 exam, or for future examinations, to review the detailed Certification Guidelines posted on the ABDSM website. I hope that the changes made this year will help ease the path of your dental sleep medicine journey and provide you with the extra encouragement needed to submit an application and embark on the road toward Diplomate status.

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Severe Obstructive Sleep Apnea Treated with Combination Hypoglossal Nerve Stimulation and Oral Appliance Therapy

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Hypoglossal cranial nerve stimulation therapy via an implantable neurostimulation system is a promising new therapy for moderate-to-severe obstructive sleep apnea patients who are unable to adhere to positive pressure therapy and who meet certain anatomical and clinical characteristics. Treatment for non-responders or incomplete responders to this novel therapy has not been studied. Particularly for patients who fail positive pressure therapy, a management plan combining multiple treatment modalities is increasingly common. This case report highlights a participant from the multicenter Inspire STAR trial who previously failed to achieve benefit with oral appliance monotherapy but then subsequently reintroduced oral appliance therapy successfully in an adjunctive role to augment the effectiveness of hypoglossal nerve stimulation therapy. Discussion will focus on the factors associated with design and selection of the oral appliance as well as the electrical parameters and titration of the neurostimulation in this unique clinical situation.

KEYWORDS: sleep apnea, oral appliance, hypoglossal nerve stimulation, combination therapy, inspire therapy

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Hypoglossal nerve stimulation (HNS) using the Inspire neurostimulation system (Inspire Medical Systems, Minneapolis, MN) is an emerging treatment modality for obstructive sleep apnea (OSA), involving synchronization of tongue protrusion stimulation and ventilatory effort during sleep. In a prospective multicenter trial, participants who met specific clinical and anatomical inclusion criteria had significant improvements in subjective and objective OSA outcome measures, and the treatment effect was maintained at long-term follow-up.^{1,2} There is no literature, however, on management of partial responders. This case report details the combination of HNS and oral appliance therapy (OAT) to successfully treat severe OSA after incomplete response with either treatment in isolation.

REPORT OF CASE

A 75-year-old male presented with a 12-year history of severe OSA. Sleep-related symptoms included loud disruptive snoring, witnessed apnea, nocturnal awakenings, and unrefreshing sleep. Although CPAP therapy provided years of subjective and objective improvement, he sought alternative treatment options due to multiple mask- and pressure-related side effects, persistent equipment-related nocturnal awakenings, and cumbersomeness with travel and camping. Oral appliance monotherapy with a custom mandibular repositioning device (TAP, Airway Labs, Carrollton, TX) was initiated two years prior to presentation, but inadequate symptom improvement and discomfort resulted in discontinuation (prior to repeat sleep testing). He later enrolled in the Inspire STAR Trial and underwent implantation of the HNS system after meeting the study inclusion criteria.¹

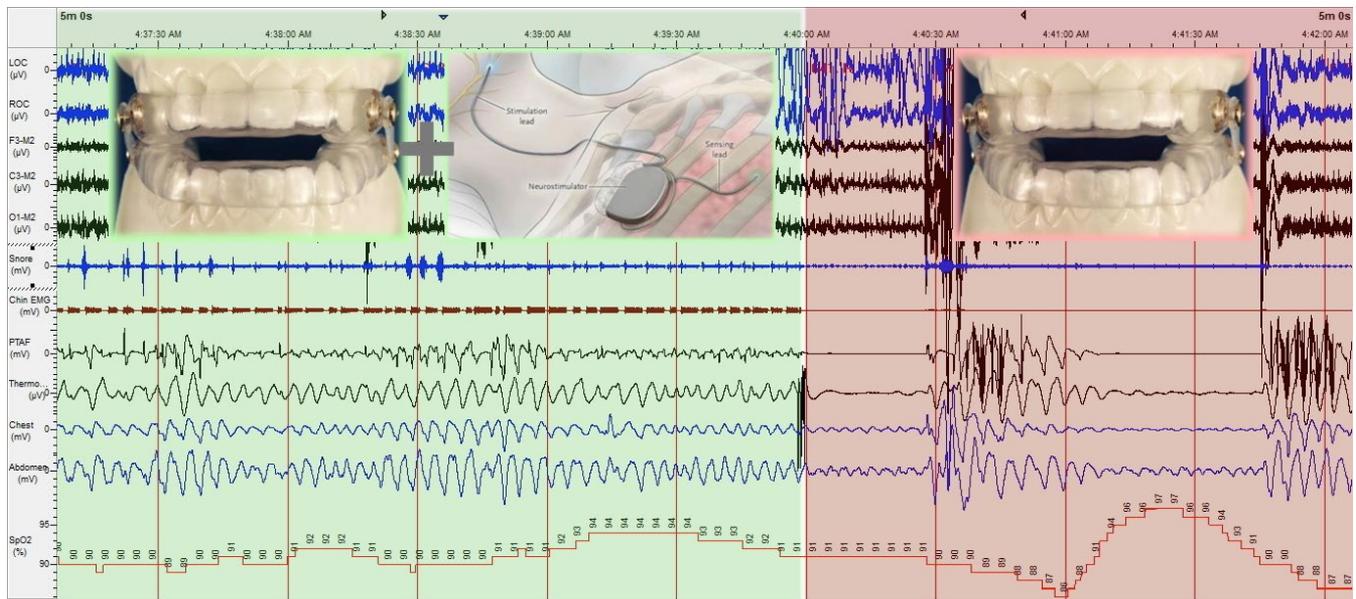
Baseline STAR Trial diagnostic polysomnography (PSG) showed an AHI of 43.7, lowest arterial oxygen saturation

(LSAT) of 75%, and arterial oxygen saturations below 90% (T90) for 33.8% of total sleep time. Six months after implantation, PSG with HNS therapy showed significant reduction in sleep apnea severity with an AHI of 11.6, LSAT 82%, and T90 of 10.2%. Patient-reported measures of daytime sleepiness and sleep-related quality of life were in the normal range at baseline and remained unchanged with HNS therapy. Per self-report and bedpartner report, snoring was significantly reduced with treatment from “loud/bedpartner leaves room” to “soft” on a visual analog scale but was not completely eliminated.

Over the subsequent year, attempts to further augment the effectiveness of HNS were made by the sleep physician/surgeon with positional therapy, topical nasal therapy, and adjustment of the electrical stimulation parameters. Although clinical, PSG, and satisfaction outcome measures remained significantly improved with nightly HNS use, mild OSA and bothersome snoring persisted.

The patient was then refitted with a new mandibular repositioning device to augment the HNS therapy. The Medley Gold (see Figure 1; TMD Technologies, Lilburn, GA) appliance was selected to allow increased anterior space for tongue protrusion that occurs with HNS therapy. Accommodation to the new appliance was successful with good subjective adherence; however, he developed intraoral discomfort, reporting that the stimulation now was too strong with combination therapy. OAT alone (HNS turned off) was tried for several weeks but again sleep-related symptoms, including loud snoring, recurred. The patient then underwent HNS reprogramming with the oral appliance in place to reduce stimulation amplitude (1.3V to 0.8V) and improve comfort.

Combination therapy with the new Medley Gold appliance and reduced HNS settings resulted in excellent subjective adherence and clinical improvement including resolution of snoring. A new PSG was completed with combination therapy.

Figure 1—Polysomnography.

Five-minute polysomnography window showing improved control of breathing in the 3 minutes on the left with combination oral appliance and hypoglossal nerve stimulation (HNS) compared to the recurrence of obstructive sleep apnea in the 2 minutes on the right immediately after the HNS is turned off.

Early in the study, HNS was wirelessly turned off to assess the effect of OAT alone (18.6 min) with clear return of sleep-disordered breathing and an AHI of 29.2 during this time. HNS therapy was then re-activated for the remainder of the night (286.9 min) during which combination therapy resulted in an AHI of 2.1 (Figure 1). The final PSG report for the entire night (305.5 min) confirmed complete objective control, with an overall AHI of 3.5, LSAT 86%, and T90 of 0.8%.

DISCUSSION

Interdisciplinary collaboration with multimodality treatment is an increasingly common approach particularly for severe OSA, although guidelines on patient phenotyping, sequencing of treatment, and specific algorithms are still lacking in the literature.³⁻⁵ In this first reported case of combination OAT and HNS therapy, symptoms and objective control of breathing were normalized as compared to partial OSA improvement with each in isolation. Furthermore, both the HNS stimulation amplitude and the degree of mandibular advancement could be reduced compared to when each was used as monotherapy.

Two key learning points may be noted in this case: (1) Stimulation parameters may be reduced on the HNS system with introduction of OAT, perhaps analogous to prior reports of reduced CPAP requirements with combination CPAP and OAT.^{4,5} (2) An oral appliance design with sufficient anterior room to accommodate tongue protrusion during active stimulation should be considered in HNS patients.

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DISCLOSURE STATEMENT

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Comparing Apples to Apples: Did This Patient Get Worse with a Mandibular Advancement Appliance?

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A 64 year-old male was assessed by a sleep medicine specialist for snoring and unrefreshing sleep. He had a past medical history of paroxysmal atrial fibrillation, for which he was taking bisoprolol and rivaroxaban. His diagnostic polysomnogram (PSG 1) demonstrated moderate obstructive sleep apnea, with an overall apnea-hypopnea index of 26.2 events per hour, and nadir oxygen saturation of 95.8%.

The patient was not keen on CPAP therapy and elected instead to consult with a dental sleep medicine practitioner regarding a mandibular advancement appliance. At this assessment, his BMI was 20.0 kg/m² and neck circumference was 15.5 inches. He had normal range of motion of his temporomandibular joints with no clicks. There was no tenderness to palpation of the temporomandibular joints or muscles of mastication. He was fully dentate with a stable occlusion and class I molar and canine relationship bilaterally.

A mandibular advancement appliance (SomnoDent Classic) was prepared for the patient, set at 70% maximum mandibular protrusion. He returned to the clinic for several minor adjustments. He subsequently reported wearing the appliance comfortably, with improvements in snoring and sleep quality. The first of two therapeutic polysomnograms was performed with the appliance in place (PSG 2). Surprisingly, this demonstrated severe obstructive sleep apnea, with an apnea-hypopnea index of 61.6 events per hour.

Upon further consideration, it became apparent that the patient spent a much greater proportion of the test sleeping supine during the first therapeutic study (PSG 2). It was suspected that differences in sleep position may have driven the apparent worsening in sleep apnea. Consequently, a second therapeutic polysomnogram was conducted (PSG 3) using both the mandibular advancement appliance and positional therapy (Rematee Bumper Belt) to avoid supine sleep. The salient findings from the three polysomnograms are summarized in Table 1.

It is a common practice in sleep laboratories to instruct patients to sleep supine while titrating CPAP therapy, as

obstructive sleep apnea is often worst in the this position.¹ In our case, the patient was instructed by the sleep technologist to sleep supine with the mandibular advancement appliance during the first therapeutic study (PSG 2). While this practice aims to ensure adequate treatment in all positions, it can be discordant with the “real world” application of the therapy and may make comparisons between studies difficult.

Further history from the patient revealed that his preference, when at home, was to sleep in the lateral position; in fact, he found it quite difficult to sleep supine during the first therapeutic study (PSG 2). Comparison of the diagnostic study (PSG 1) to the second therapeutic study (PSG 3) reveals a 32% reduction in lateral AHI and a 39% reduction in overall AHI compared to the diagnostic study. This result is more in keeping with general response rates to mandibular advancement appliances, and may account for the patient's improvement in symptoms. The appliance did not improve the patient's sleep apnea when supine, but this may be irrelevant if supine sleep can be avoided completely in the home setting. Practitioners are encouraged to take into account between-night discrepancies in sleep position when comparing the results of diagnostic and therapeutic studies, as examination of only the overall apnea-hypopnea index can sometimes be misleading.

CITATION

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Table 1—Summary of apnea-hypopnea index and position data from polysomnography.

| | %TST Supine | Supine AHI (events/h) | %TST Lateral | Lateral AHI (events/h) | Overall AHI (events/h) |
|--------------------|-------------|-----------------------|--------------|------------------------|------------------------|
| PSG 1: Diagnostic | 13% | 48.0 | 86% | 22.8 | 26.3 |
| PSG 2: Therapeutic | 94% | 60.6 | 6% | 78.0* | 61.6 |
| PSG 3: Therapeutic | 2% | 37.1* | 98% | 15.6 | 16.0 |

*The reliability of these indices is questionable given the very short sleep time. PSG, polysomnogram; %TST, percentage of total sleep time; AHI, apnea-hypopnea index.

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This was not an industry supported study. Dr. Li has served as medical director and has been a shareholder of Somnaform Corporation, which has developed a positional device for management of snoring. This was not the positional device used during the therapeutic polysomnogram in the case reported. The other authors have indicated no financial conflicts of interest. The sleep studies for this case were carried out at the sleep laboratory, St. Michael's Hospital, Toronto, ON, Canada. There was no off-label or investigational use.

AADSM 2015–2016 Educational Calendar of Events

AADSM Staff

AADSM National Office, Darien, IL

Upcoming 2015 Education

November 7–8

Advanced Dental Sleep Medicine Course
Orlando, FL

November 7–8

Essentials of Dental Sleep Medicine Course
Orlando, FL

December 5

Practical Demonstration Course
Darien, IL – AADSM National Office

Upcoming 2016 Education

February 20–21

Essentials of Dental Sleep Medicine Course
Charleston, SC

February 20–21

Board Review Course
Charleston, SC

March 18

Practical Demonstration Course
Darien, IL – AADSM National Office

June 9–11

25th Anniversary Meeting
Denver, CO

