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Original Article

Differences in dentofacial morphology in children with sleep disordered breathing are detected with routine orthodontic records

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ABSTRACT

Background: Residual snoring in children with obstructive sleep disordered breathing (SDB) may continue post-adenotonsillectomy. This study aims to identify baseline dentofacial differences in children with SDB using routine orthodontic records that might aid effective early intervention for the upper airway to prevent continued obstruction.
Methods: Children (6–16 years) with clinically diagnosed SDB from a paediatric Otolaryngology Clinic who required adenotonsillectomy were participants ($n = 10$). The control group ($n = 9$) comprised healthy non-snoring children from the community. Baseline overnight polysomnography (PSG), standardised frontal and right profile photographs and alginate impressions were taken of all children. Facial width, length, depth, convexity and mandibular position were measured from the photographs. The occlusion, arch width, arch depth, maxillary arch form, palatal height and volume were recorded from digitised dental models. Inter-group differences were compared.
Results: SDB patients had a significantly increased lower face height, maxillo-mandibular angle (1.73°; 95% CI 0.45–3.0) and a narrower maxillary arch in the upper posterior region. There was a trend towards a decreased palatal volume, increased posterior crossbite and Class II molar relationship.
Conclusion: Dentofacial phenotypic differences between children with SDB and controls can be detected using facial photographs and dental models. Increased awareness of these features may help to identify children who to continue to snore post adenotonsillectomy.

1. Introduction

Habitual snoring and obstructive sleep apnoea (OSA) is reported to affect around 1–15% of children [1]. The effects of SDB on the health of a child are wide ranging and may include poor growth [2], adverse behavioural and learning effects [3], poor academic performance [4], cardiovascular [5] and metabolic [6] complications and impairment in quality of life [7].

Adenotonsillar hypertrophy is one of the most common causes of paediatric obstructive sleep disordered breathing (SDB) in children and adenotonsillectomy (AT) is the primary surgical treatment option in children with moderate to severe obstructive sleep apnoea [8]. Although there are improvements to SDB severity post-operatively, it has been reported to persist between 13 and 29% in low-risk populations and up to 73% in high-risk obese children [9]. While neuromuscular and respiratory control mechanisms contribute towards the maintenance of airway patency, dentofacial abnormalities are a known risk factor for obstructive sleep apnoea (OSA) [10].

Features of a long and narrow face have been associated in children with SDB including an increased facial height, steep mandibular plane angle, transverse maxillary deficiency and habitual mouthbreathing [11]. Two recent systematic reviews that included studies using lateral cephalograms reported that children with SDB had an increased mandibular plane angle (4.2° 95% CI 3.32–5.07), retrusive chin angle (−1.79° 95% CI −2.61 to −0.97), sagittal jaw discrepancy within 2° and a reduced posterior airway space [12,13]. Recently, a reduced mandibular size has been postulated to be a craniofacial contribution to residual SDB [14].

The developing dentition is also known to be associated with breathing obstruction [15] and OSA [16,17]. Class II malocclusion

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and posterior crossbite are among the most commonly reported findings [18]. Preliminary studies have suggested that orthodontic and dentofacial orthopaedic treatments such as maxillary expansion or mandibular advancement with functional appliances might be effective in improving paediatric SDB [19].

As the relationship between dentofacial disharmony and sleep disordered breathing is small but significant, a feasible clinical method that can detect these differences may help to identify appropriate patients for targeted therapy [20]. This study aims to evaluate the baseline differences in dentofacial morphology of children who have SDB validated by polysomnography using routine orthodontic diagnostic records.

2. Materials and methods

2.1. Subjects

Ethics approval was granted by the Adelaide Womens' and Childrens' Hospital Human Research Ethics Committee (HREC) and the University of Adelaide. The parent/s of all subjects gave written consent in accordance with the Declaration of Helsinki. Full inclusion and exclusion criteria are outlined in Table 1.

Children awaiting surgery for the removal of their adenoids and tonsils because of SDB were prospectively recruited as participants from the paediatric otorhinolaryngology (ENT) waiting list at the Women's and Children's Hospital (WCH, Adelaide, Australia). The subjects were referred by their primary physician for the evaluation of SDB because of a history of parental report of snoring more than three nights per week. The subjects were clinically diagnosed with SDB and had hypertrophic adenoids and tonsils $\geq$3 based on a standardised scale of 0–4 [21]. Healthy non-snoring children were prospectively and consecutively asked to participate in the study as a control group from the community via advertisement and through relatives of the children with SDB.

2.2. History and Anthropometrics

On the night of testing, height, weight and neck circumference at the level of the cricothyroid membrane were measured. Body mass index (BMI) and BMI z-score were calculated [22].

2.3. Overnight polysomnography

Baseline overnight polysomnography (PSG) was conducted for all children and scored as previously described in Kontos et al., [23] including the obstructive apnoea/hypopnea index (OAHI).

2.4. Orthodontic records

Each child had a set of standardised facial photographs and an upper and lower alginate impression taken by a single blinded clinician.

Extra-oral digital photographs (frontal and right profile) of the head and neck were obtained with a standardised setup similar to Lee et al., [24]. Subjects were photographed standing upright while assuming natural head position. Gonion and infraorbital rim were pre-identified and marked on the subjects. A single lens reflex digital camera with a single external flash unit (D7000 with AF-S 105 mm f/2.8 VR Micro-NIKKOR and SB-700; Nikon Corp., Japan) was mounted on a tripod at a distance of 270 cm from the subject alignment plane. Standardised camera settings (focal length 300 cm, aperture f/5.6, shutter speed 1/125th, ISO 400) were used to ensure consistency of the JPEG images (4928 $\times$ 3264 pixels). A grid to be used as a reference scale was included adjacent to the patient in their alignment plane.

A set of upper and lower alginate impressions (Kromopan; Lascod, Italy) were taken in disposable plastic trays (Algi-Grip; Select Dental, USA). The impressions were poured in a 50:50 combination of white plaster and die-stone (Investo; Ainsworth Dental, Australia) and trimmed using a wax bite registration taken in centric occlusion. The plaster study casts were scanned with the OrthoInsight 3D scanner (Motionview; Hixson, TN) to produce digital dental models.

2.5. Facial image analysis

Using image analysis software (Image J v1.50; NIH, Bethesda, MD), the extra-oral photographs were uploaded and landmarks were identified and digitised. A total of 16 facial measurements (10 linear, 6 angular) were recorded. The measurements represented the width, depth and height of the face as well as relative maxillo-mandibular positions within the face replicated from Farkas [25] (Fig. 1).

2.6. Dental model analysis

Inter-canine width, inter-second premolar width, inter-first molar width and arch depth were measured (Fig. 2). The molar classification, stage of dentition, presence or absence of posterior crossbite, anterior overbite and overjet were also recorded. All registrations were carried out with the proprietary software included with the model scanner.

2.7. Palatal vault analysis

Digitised maxillary models were exported to CAD-CAM software (Spaceclaim; ANSYS, Massachusetts, USA) so that the maxillary

Table 1
Inclusion and Exclusion criteria.

Inclusion	Exclusion
Age 6–16 years (child). Clinical diagnosis of sleep disordered breathing (SDB) defined as caregiver report of habitual snoring that occurs most of the night on at least three nights per week and has been present for at least three months.	Previous tonsillectomy, including partial tonsillectomy. Severe obesity (body mass index z-score $\geq$ 3).
Tonsillar hypertrophy $\geq$3 based on a standardised scale of 0–4.	Failure to thrive, defined as either height or weight being below the 5th percentile for age or gender.
Deemed to be a candidate for AT by otolaryngologist (ENT) evaluation (ie no technical issues that would be a contraindication for surgery).	History of orthodontic treatment
Primary indication for AT is nocturnal obstructive symptoms (ie, not recurrent infections or other indications).	Severe chronic health conditions that might hamper participation or confound the study variables including: - significant asthma - significant craniofacial abnormalities - medications that affect sleep or respiration - developmental and psychiatric disorders English as a second language

Fig. 1. Photographic Landmarks: *sup* – infraorbital rim; *me'* – ST mentum; *gol'* – ST gonion (L); *gor'* – ST gonion (R); *g'* – ST glabella; *n'* – ST nasion; *sn* – subnasion; *A'* – ST A' point; *B'* – ST B' point; *pog'* – ST pogonion; *tl* – tragion (L); *tr* – tragion (R); *all* – alare (L); *alr* – alare (R); ST = soft tissue; (L) = left side of the patient; (R) = right side of the patient.

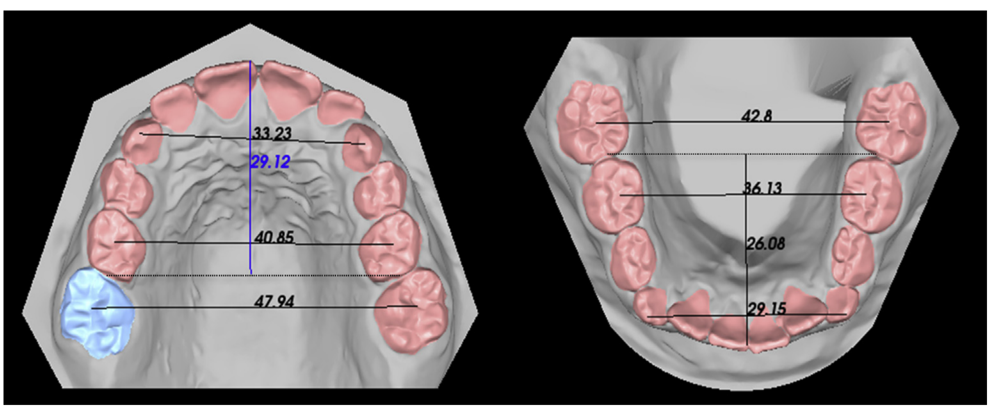

Fig. 2. Dental Landmarks: Inter-canine width – distance between the crown tips of the canines; Inter-second premolar width – distance between the central fossae of the second primary molars; Inter-first molar width – distance between the central fossae of the first permanent molars; Arch depth – perpendicular distance from the labial surface of the central incisors to the distal surfaces of the second primary molars at the midline.

palatal height and width at three different cross-sections could be measured. The palatal index was calculated as the palatal height divided by the palatal width. A solid volume defining the boundaries of the maxillary palate was separately analysed to determine the palatal volume [26] (Fig. 3).

2.8. Measurement error

Technique validation (landmark digitisation accuracy and test-retest reliability) was performed in a subgroup of subjects for all methods. Based on 2 standard deviations of error, measurement error was within 1.8 mm, 1.1 mm and 1.0° for linear facial, dental and angular measurements respectively.

2.9. Data and statistical analysis

Data were stored and analysed on a software package used for statistical analysis (SPSS v.23; Chicago, Illinois, USA). To exclude age-related growth differences in facial and dental arch sizes and to compare the morphology between the two groups, all linear measurements were normalised by age and gender using standards of facial [25] and dental arch size [27] measured using the same methodology. The normalised values (z-scores) were calculated as follows: Normalised value = [value of study sample−mean value of the standard value]/standard deviation of the standard value.

Each variable was examined for its normality and distribution and analyses were interpreted with and without outliers. Intergroup differences were compared using independent sample t-tests for continuous variables and Fisher's Exact test (non-parametric) for categorical variables. Linear modelling of significantly different dentofacial variables was performed to assess if the differences between the groups were due to an interaction with obesity. All values are presented as mean ± standard deviation with 95% confidence intervals, unless otherwise stated. A p value of less than 0.05 was considered statistically significant.

3. Results

3.1. Subject characteristics

The sample consisted of nine controls and 10 SDB children. There was no difference between the groups for gender, age, ethnicity, height, weight and neck circumference. However, the SDB group had greater BMI ($p < 0.046$) and BMI z-scores

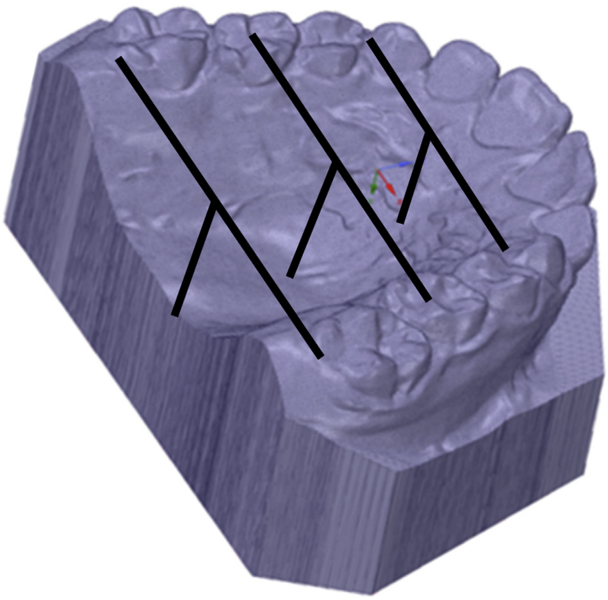

Fig. 3. Palatal width – distance between the disto-gingival points of the upper first molars; Palatal height – perpendicular distance from the palatal width line to the palate at the midline; Palatal volume – The volume bounded by the palate, an axial plane (disto-gingival points of the upper first molars and the tip of the incisive papilla) and a coronal plane (perpendicular to the axial plane through the disto-gingival points of the upper first molars).

($p < 0.022$). Based on the polysomnography, the mean obstructive apnoea-hypopnea index (OAHI) was significantly greater in the SDB children (1.26 ± 1.3) compared to controls (0.16 ± 0.3) ($p < 0.026$) (Table 2).

3.2. Facial morphology

There were three variables that were significantly different between the groups. The SDB group had an increased z-score for lower anterior face height ($p < 0.003$) and lower jaw length ($p < 0.039$). There was also an increased maxillo-mandibular angle ($p < 0.011$) of 1.73° (95% CI 0.45–3.0). There was a trend for a shorter and narrower nose, increased facial convexity and steeper mandibular plane angle, however, these were not significantly different (Table 3).

Linear modelling was used to assess the effect of BMI and BMI z-scores on the effect of lower jaw length and lower anterior face height, since soft tissue coverage can affect these measurements. After controlling for BMI and BMI-z scores, there was no difference in lower jaw length; however, there was still a difference in lower anterior face height.

3.3. Dental morphology

There were two variables that were significantly different between the groups. The SDB group had decreased z-scores for maxillary inter-second premolar ($p < 0.009$) and maxillary inter-first molar width ($p < 0.033$). No significant differences were found in any other measure of maxillary or mandibular dental arch width, depth and occlusion. Notably, there was a trend for the SDB group to have an increased frequency of posterior crossbite (50% vs 12.5%), Class II molar relationship (60% vs 25%) and a wider and longer mandibular arch although this was not significant (Table 4). No subjects in this study had a Class III molar relationship.

3.4. Palatal vault morphology

The palatal index distal to the upper first molars was significantly greater in the SDB group indicating an impression of a higher and narrower palate posteriorly. However, no difference in the palatal height was found when the values were normalised.

There were no known reference values for palatal volume and the non-normalised data were presented. Although no significant difference was found between the groups, one would expect that growth could have an effect on this value as the SDB group had a trend for a greater proportion of older males. Yet, it is noteworthy that the SDB group had a mean decrease in their palatal volume by 621.7 mm^3 (95% CI −465.93–1709.35).

4. Discussion

This study has demonstrated that routine orthodontic records are able to identify quantitative differences in dentofacial morphology in children with SDB confirmed by PSG. Significant differences included an increase in lower anterior face height, a

Table 2
Subject characteristics.

	Controls (n = 9)	SDB (n = 10)	*p*-value
Males (%)	7 (78%)	8 (80%)	<1.0[a]
Age (yrs)	9.56 ± 2.14 (8–17)	11.85 ± 3.29 (7–14)	<0.09
Ethnicity (% Caucasians)	100%	100%	<1.0
Anthropometry			
Height	132.78 ± 14.04	147.76 ± 18.17	<0.06
Weight	33.6 ± 19.67	55.49 ± 25.84	<0.052
BMI	**18.33 ± 5.9 (14.2–31.9)**	**24.15 ± 4.93 (14.8–35.0)**	**<0.046**
BMI z-score	**0.16 ± 1.26**	**1.55 ± 0.85**	**<0.022**
Neck circumference (cm)	29.78 ± 4.14	33.6 ± 4.95	<0.085
Polysomnography			
Total sleep time (hrs)	7.01 ± 0.94	6.64 ± 0.93	<0.401
S1 (%TST)	7.8 ± 4.91	11.24 ± 6.38	<0.203
S2 (%TST)	36.51 ± 5.38	35.51 ± 7.7	<0.745
Slow wave sleep (%TST)	39.89 ± 7.04	35.94 ± 9.69	<0.321
REM (%TST)	15.78 ± 3.44	17.32 ± 3.45	<0.344
Nadir SpO_2	92.22 ± 3.27	88.7 ± 3.83	**<0.045**
Respiratory arousals/hr	1.42 ± 1.65	7.27 ± 6.4	**<0.019**
OAHI	0.16 ± 0.3 (0–0.93)	1.26 ± 1.3 (0.02–4.02)	**<0.026**
Tech reported snoring	n = 1 (11%)	n = 5 (50%)	<0.14[a]

Mean ± SD; BMI = body mass index; OAHI = obstructive apnoea hypopnea index.
Bold represents significant difference (*p-value <0.05*).
[a] Fisher's Exact test.

Table 3
Facial analysis.

Facial variables	Landmarks	Control (n = 9)	SDB (n = 10)	*p*-value
Upper face depth[a]	t-n'	1.3 ± 0.99	2.91 ± 2.48	<0.084
Mid face depth[a]	t-sn	1.84 ± 1.07	3.43 ± 2.61	<0.102
Lower face depth[a]	t-me'	1.42 ± 0.94	2.44 ± 1.55	<0.101
Lower jaw length[a]	**go'-me'**	**0.94 ± 1.45**	**2.44 ± 1.48**	**<0.039**
Upper face (nose) height[a]	n'-sn	0.84 ± 1.75	0.79 ± 1.36	<0.949
Total anterior face height[a]	n'-me'	0.37 ± 0.94	1.14 ± 0.95	<0.094
Lower anterior face height[a]	**sn-me'**	**0.31 ± 0.72**	**1.77 ± 1.04**	**<0.003**
Face width[a]	tl-tr	1.79 ± 1.20	2.35 ± 1.52	<0.381
Mandibular width[a]	gol'-gor'	4.28 ± 1.8	5.19 ± 1.34	<0.238
Nose width[a]	all-alr	2.05 ± 1.11	1.97 ± 1.20	<0.886
Maxillary depth angle	t-n'-A'	80.87 ± 4.85	81.85 ± 3.54	<0.626
Mandibular depth angle	t-n'-B′	72.87 ± 4.28	72.11 ± 3.31	<0.675
Maxillo-mandibular angle (°)	**A′-n'-B′**	**8.01 ± 1.4**	**9.74 ± 1.21**	**<0.011**
Facial convexity	g'-sn-pog'	168.51 ± 4.07	166.16 ± 4.16	<0.232
Natural head position:FH′	t-sup:TH	1.21 ± 3.29	3.83 ± 3.13	<0.094
Mandibular plane angle:FH′	t-sup:go'-me'	30.93 ± 4.79	31.60 ± 7.31	<0.815

Mean ± SD.
Bold represents significant difference (*p-value <0.05*).
[a] z-scores.

maxillo-mandibular jaw discrepancy indicative of a retrognathic mandible [28] and narrower maxillary arch. The data also revealed a trend towards an increased frequency of posterior crossbite, Class II malocclusion, wider and longer mandibular arch and an overall reduction in maxillary palatal vault volume.

A polysomnogram was performed on all children which confirmed the diagnosis of SDB based on the OAHI. SDB patients clearly demonstrated the OAHI was significantly greater than one, which represents the diagnostic threshold for OSA [29]. The OAHI is used as an objective parameter for OSA with an OAHI≤1/hr considered to be normal, 1 < OAHI ≤ 5 to be mild OSA, 5 < OAHI ≤ 10 to be moderate OSA and OAHI > 10 as severe OSA [30]. Further differences in dentofacial morphology might be expected in a group of subjects with increased severity of OSA.

Standardised clinical photography was used as it provided a detailed quantification of facial features. It is a simple, safe and quick image acquisition technique that has minimal measurement error. While other available imaging technologies are available (lateral cephalometry and cone beam volumetric tomography) and allow a detailed examination of skeletal, soft tissue and upper airway structures, they involve a degree of radiation exposure. This was not ethically possible in this study where the diagnostic information gained would not alter clinical management. Sophisticated three-dimensional facial scanning is also increasingly accessible [31]; however, it has limited clinical application due to its large data sets and time-consuming analyses.

Table 4
Dental Analysis.

	Control (n = 9)	SDB (n = 10)	*p*-value
Maxilla			
Mx intercanine[a]	0.37 ± 1.19	−0.68 ± 0.59	<0.053
Mx inter-second premolar[a]	**0.35 ± 0.69**	**−1.15 ± 1.35**	**<0.009**
Mx inter-first molar[a]	**0.27 ± 1.22**	**−1.03 ± 1.1**	**<0.033**
Mx arch depth[a]	1.4 ± 1.13	1.18 ± 1.47	<0.740
Mandible			
Mn intercanine[a]	−0.05 ± 1.43	−0.58 ± 0.99	<0.388
Mn inter-second premolar[a]	−0.01 ± 0.7	0.08 ± 1.22	<0.827
Mn inter-first molar[a]	0.17 ± 1.04	0.43 ± 1.19	<0.636
Mn arch depth[a]	1.54 ± 1.67	1.75 ± 4.5	<0.895
Occlusion			
Overbite (mm)	2.8 ± 0.84	2.66 ± 1.93	<0.840
Overjet (mm)	4.02 ± 0.66	4.26 ± 1.37	<0.638
Posterior crossbite	n = 1 (12.5%)	n = 5 (50%)	<0.37[b]
Class II molar	n = 2 (25%)	n = 6 (60%)	<0.07[b]
Palate			
Palatal height[a]	−0.89 ± 1.67	−0.61 ± 1.25	<0.69
Palatal index (anterior)	0.21 ± 0.07	0.19 ± 0.08	<0.591
Palatal index (mid arch)	0.39 ± 0.06	0.45 ± 0.1	<0.124
Palatal index (posterior)	**0.26 ± 0.3**	**0.34 ± 0.07**	**<0.004**
Palatal volume[c]	6584.23 ± 968.3	5962.53 ± 1208.38	<0.243

Mean ± SD.
Bold represents significant difference (*p-value <0.05*).
[a] z-scores.
[b] Fisher's Exact test.
[c] Not adjusted.

Dental impressions were taken to form digital dental models as these are another routine orthodontic record. Objective measurements of the dental arches could be analysed in three-dimensions and this allowed a detailed investigation of the palatal vault which could not otherwise be done with clinical photography or examination alone. Although alginate impressions can be unpleasant for some children [32], the use of intra-oral scanning might replace the conventional method and should be considered in future investigations.

One of the limitations of this study was patient recruitment. A relatively small number of patients were examined but despite this, significant differences were found in children who snored, but these finding will need to be corroborated in a larger study. The data is largely in agreement with the craniofacial phenotype associated with airway obstruction which has been known as the 'long face syndrome' [11–13,31]. The skeletal maxillary constriction and retrognathic mandibular position may benefit from maxillary expansion and orthopaedic mandibular advancement. Recently, there has been data to suggest rapid maxillary expansion is beneficial in children with SDB [33,34] and use of a mandibular advancement splint can produce clinically relevant reductions in supine AHI [35].

Dentofacial analysis using clinical photographs is readily applicable to clinical paediatric sleep practise but it is not commonly utilised to examine children who are referred with a history of habitual snoring and suspected SDB. It may even be possible to use intraoral clinical photographs instead of dental models as a number of features such as posterior crossbite and Class II malocclusion can be detected. In further studies, this technique may provide a discriminating method to evaluate whether dental and facial structure analysis is a significant factor in children with residual SDB who continue to snore post adenotonsillectomy.

5. Conclusion

There were significant dentofacial differences between children with and without SDB detected using routine orthodontic records including facial photography and dental models. After controlling for differences in age, gender and obesity, SDB children had a longer lower anterior face height, larger maxillo-mandibular angle of 1.73° (95% CI 0.45–3.0), narrower maxillary arch and a trend towards an increased frequency of posterior crossbite, Class II molar relationship and decreased palatal volume. Increased awareness of these features may indicate early referral for orthodontic and dentofacial orthopaedic management. Further investigation is required to determine if these features are associated with residual SDB post-adenotonsillectomy.

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Conflict of interest

None.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2018.12.019.

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