Craniofacial and upper airway morphology in pediatric sleep-disordered breathing and changes in quality of life with rapid maxillary expansion

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Introduction: The association between pediatric sleep-disordered breathing caused by upper airway obstruction and craniofacial morphology is poorly understood and contradictory. The aims of this study were to evaluate the prevalence of children at risk for sleep-disordered breathing, as identified in an orthodontic setting by validated screening questionnaires, and to examine associations with their craniofacial and upper airway morphologies. A further aim was to assess the change in quality of life related to sleep-disordered breathing for affected children undergoing rapid maxillary expansion to correct a palatal crossbite or widen a narrow maxilla. Methods: A prospective case-control study with children between 8 and 17 years of age (n = 81) at an orthodontic clinic was undertaken. The subjects were grouped as high risk or low risk for sleep-disordered breathing based on the scores from a validated 22-item Pediatric Sleep Questionnaire and the Obstructive Sleep Apnea-18 Quality of Life Questionnaire. Variables pertaining to a screening clinical examination, cephalometric assessment, and dental cast analysis were tested for differences between the 2 groups at baseline. Ten children who underwent rapid maxillary expansion were followed longitudinally until removal of the appliance approximately 9 months later with a repeated Obstructive Sleep Apnea-18 Quality of Life Questionnaire. All data were collected blinded to the questionnaire results. Results: The frequency of palatal crossbite involving at least 3 teeth was significantly higher in the high-risk group at 68.2%, compared with the low-risk group at 23.2% (P <0.0001). Average quality of life scores in the high-risk group indicated reduced quality of life related to sleep-disordered breathing by 16% compared with children in the low-risk group at baseline (P <0.0001). Cephalometrically, mean inferior airway space, posterior nasal spine to adenoidal mass distance, and adenoidal mass to soft palate distance were reduced in the high-risk group compared with the low-risk group by 1.87 mm (P <0.03), 2.82 mm (P <0.04), and 2.13 mm (P <0.03), respectively. The mean maxillary intercanine, maxillary interfirst premolar, maxillary interfirst molar, mandibular intercanine, and mandibular interfirst premolar widths were reduced in the high-risk group compared with the low-risk group by 4.22 mm (P <0.001), 4.24 mm (P <0.001), 1.50 mm (P <0.01), and 1.84 mm (P <0.01), respectively. Children treated with rapid maxillary expansion showed an average improvement of 14% in quality of life scores in the high-risk group compared with the low-risk group, which showed a slight worsening in quality of life related to sleep-disordered breathing by an average of 1% (P <0.04), normalizing the quality of life scores in the high-risk children to the baseline scores compared with the low-risk group. Conclusions: Children at high risk for sleep-disordered breathing are characterized by reduced quality of life, reduced nasopharyngeal and oropharyngeal sagittal dimensions, palatal crossbite, and reduced dentoalveolar transverse widths in the maxillary and mandibular arches. No sagittal or vertical craniofacial skeletal cephalometric predictors were identified for children at high risk for sleep-disordered breathing. In the short term, rapid maxillary expansion
Pediatric sleep-disordered breathing (SDB) caused by upper airway obstruction is associated with the cardinal symptom of snoring. In children, SDB exhibits a spectrum of severity ranging from primary snoring as the mildest form to obstructive sleep apnea (OSA) as the most severe. Primary snoring is not associated with any gas exchange abnormalities or sleep fragmentation, whereas OSA is characterized by repetitive and prolonged partial or complete upper airway obstruction that disrupts normal ventilation during sleep. The spectrum of SDB in children has gained increased attention because of the deleterious health implications if it is left undiagnosed or untreated.

The reported prevalence of primary snoring ranges from 3.2% to 35%, and OSA ranges from 0.7% to 10.3% in children, depending on the diagnostic instrument used to measure SDB, with most authors reporting prevalences of 10% for primary snoring and <3% for OSA. Although snoring was once believed to be benign, it is now recognized that it can be associated with significant sleep disruption and daytime symptoms. Both primary snoring and OSA are known to impact quality of life, behavior, and neurocognition, the cardiovascular system, and lipid regulation in children. Children affected by SDB are more likely to be diagnosed with attention deficit hyperactivity disorder. There is relatively poor recognition of pediatric SDB in clinical practices because approximately 80% of symptomatic habitual snorers are not reported to their general medical practitioners. In addition, there is a 226% (2.3-fold) increase in health care utilization among children with OSA compared with the unaffected population. Hence early diagnosis and intervention should be beneficial and cost-effective.

Many studies have reported a positive relationship between craniofacial characteristics such as high palatal vault, narrow maxilla, mandibular retrognathia, increased facial height, and SDB in nonsyndromic children. However, evidence from 2 meta-analyses suggests that the sagittal and vertical craniofacial associations, as measured on a lateral cephalogram, might have low clinical significance in predicting childhood SDB. In contrast, evidence from clinical trials indicates that rapid maxillary expansion (RME) might be an effective treatment for children with a narrow maxilla and OSA. Little attention has been paid to children in the snoring end and the middle of the SDB spectrum. A recent questionnaire-based screening study by Huynh et al assessed patients in an orthodontic setting, with most children being at the milder end of the SDB spectrum. They found that SDB was primarily associated with adenotonsillar hypertrophy and morphologic features such as narrow palate, dolichocephalic pattern, high mandibular plane angle, and severe maxillary and mandibular crowding. Interestingly, Huynh et al did not assess radiographic or dental cast measurements and relied solely on a visual clinical screening examination. Hence, the association between craniofacial and upper airway morphology and pediatric SDB in the orthodontic setting can be regarded as poorly understood and somewhat contradictory.

The primary cause of SDB in children is reported to be adenotonsillar hypertrophy, which results in upper airway obstruction particularly when accompanied by other factors affecting airway patency or muscle tone. Adenotonsillectomy is therefore recommended as the first line of treatment for pediatric SDB and is curative in 25% to 80% of patients. Normalization after adenotonsillectomy surgery is less frequently seen in black children, obese children, and those with severe OSA at baseline. Nasal continuous positive airway pressure is a nonsurgical alternative treatment, but there is evidence of developmental midface hypoplasia and other craniofacial side effects in children with this approach. Dentofacial orthopedics, particularly RME, is an emerging treatment modality in the management of pediatric OSA.

Overnight polysomnography is considered the gold standard for diagnosis of OSA in children; however, polysomnography is expensive, time-consuming, and frequently inaccessible. Various validated screening questionnaires have been developed to aid in the screening of children with SDB by standardizing history taking and evaluating quality of life, behavior, neurocognition, and caregiver concerns. The quality of sleep is related to the quality of life, and the measurement of health-related quality of life provides an assessment of the health status of a clinical sample and the effects of intervention, as perceived by the parent or the patient. To date, there are no data on changes in SDB-related quality of life after RME treatment for children at the snoring end of the SDB spectrum. Therefore, the main aims of this study were to evaluate the prevalence of children at risk for SDB, as identified in an orthodontic setting by validated screening questionnaires, and to
examine associations with craniofacial and upper airway morphology. A further aim was to assess the change in the SDB-related quality of life for affected children having RME to correct a palatal crossbite or widen a narrow maxilla.

**MATERIAL AND METHODS**

Ethical permission was granted by the Royal Adelaide Hospital (Adelaide, Australia) Human Research Ethics Committee. Informed consent was obtained from all parents or guardians, and verbal assent from the children before data collection for the study.

The subjects were children less than 18 years of age who came to the orthodontic clinic for diagnosis and treatment between February 2012 and April 2013. Good general health, normal weight, the availability of study models or lateral cephalometric radiographs within 6 months of the baseline orthodontic examination (T1), and no previous orthodontic treatment were required for inclusion. The initial sample comprised 81 children. Weight and height were measured at the orthodontic examination to calculate the body mass index (weight in kilograms divided by height in meters squared) for each child. Since obesity might be a confounding factor, 3 children with a body mass index above the 95th percentile (>31.9 kg/m²) of the group were removed from the primary statistical analyses, leaving a final study cohort of 78 children (33 boys, 45 girls). Lateral cephalograms for 6 children and dental casts for 11 children were not taken at T1 for patient management reasons.

The parents or guardians at the clinical examination were asked to complete a medical history and 2 questionnaires on behalf of their children to assess sleep, daytime behavior, and sleep duration and quality at T1. The sleep and daytime behavior questionnaire was a modified and validated 22-item Pediatric Sleep Questionnaire (PSQ). All positive responses were grouped under “yes” and all negative responses under “no.” Each “yes” response was given a score of 1. The second questionnaire was the validated and modified version of the OSA-18 to assess the children’s quality of life in 4 domains that included sleep disturbance, physical discomfort, emotional distress, and caregiver concerns.

At the end of the OSA-18 Quality of Life Questionnaire, the parents were asked to mark the perceived quality of life of their child caused by issues related to sleep and breathing on a 0 to 10 visual analog scale, with higher scores indicating a better SDB-related quality of life. The scoring for the PSQ varies from 0 to 22 points, with higher scores indicating greater severity; the scoring for OSA-18 varies from 15 to 126 points, with higher scores indicating a worse quality of life. Studies have validated that if a child’s PSQ score is greater than 7 “yes” responses or the OSA-18 score is greater than 60, a high probability of SDB can be expected. Children were grouped as “high risk” or “low risk” according to the results of the 2 questionnaires, with the high-risk group having more than 7 “yes” responses to the PSQ or a score of 60 or more on the OSA-18 questionnaire.

Fifteen participants (68.2%) in the high-risk group and 13 (23.2%) in the low-risk group were diagnosed with a palatal crossbite or a narrow maxilla and were recommended for RME treatment. Five children in the high-risk group and 5 in the low-risk group (n = 10) who underwent RME (mean age, 10.3 ± 1.3 years) were followed longitudinally until removal of the appliance (T2) approximately 7 to 9 months later (mean age, 10.9 ± 1.3 years). The RME appliance was a 4-banded hyrax-type with a rate of activation of 0.5 mm daily.
The expansion was stopped between 14 and 21 days, or once the palatal cusps of the maxillary molars were in line with the buccal cusps of the mandibular molars, to allow for some relapse. After expansion, the RME device served as a passive retainer to allow sutural and bony adaptation. Since the OSA-18 Quality of Life Questionnaire has been validated to measure changes in SDB-related quality of life in a pediatric sample, it was repeated at T2 to assess the quality of life changes in the children treated with RME.

All subjects were clinically evaluated at T1 under the supervision of orthodontists, blinded to the questionnaire results, using a standardized orthodontic evaluation form covering dental, skeletal, functional, and esthetic factors. Sagittal craniofacial form was recorded as skeletal Class I, Class II, or Class III. Vertical evaluation included the visual categorization of face height as mesofacial, brachyfacial, and dolichofacial. The presence or absence of a palatal crossbite involving at least 3 teeth was recorded.

Fifty-five children in the low-risk group and 17 in the high-risk group (n = 72) had lateral cephalograms taken at T1, all taken on the same machine (Kodak Carestream CS 9000; Eastman Kodak Company, Rochester, NY), with the patient erect in natural head position, the teeth in maximum intercuspation, and the lips relaxed. The enlargement factor (8%) was adjusted to provide true size measurements. The cephalograms were digitized and analyzed using Dolphin Imaging software (version 11.5; Dolphin Imaging and Management Solutions, Chatsworth, Calif). The cephalometric landmarks and analyzed planes are shown in Figure 2. Cephalometric measurements included 17 morphologic, 3 airway, and 1 hyoid position variables. The cephalometric variables tested were as follows.

1. Cranial base assessment: cranial base length (SN length) and cranial base flexion (NSBa angle).
2. Maxillary and mandibular skeletal assessment: maxillary position in relation to cranial base (SNA angle), mandibular position in relation to cranial base (SNB angle), maxillomandibular sagittal differential (ANB angle and Wits appraisal), maxillary length (Co–A-point), mandibular length (Co–Gn), and maxillomandibular length differential (Mx–Md, difference between maxillary and mandibular lengths).
4. Dental measurements: angulation of maxillary incisor to cranial base (U1–SN angle), angulation of mandibular incisor to mandibular plane (L1–MP angle), and occlusal plane in relation to cranial base (OP–FH angle).
5. Airway dimensions: oropharyngeal airway dimension (IAS), nasopharyngeal airway dimension (PNS–AD1), and patency of nasopharyngeal airway (AD1–SP).
6. Hyoid position: perpendicular distance from anterio-
superior point on hyoid body to mandibular plane
(Hy–MP).

Forty-six children in the low-risk group and 21 chil-
dren in the high-risk group had dental casts available
(n = 67) at T1. Photocopies of dental models were taken
for measurements and analyzed to true size (photocopi-
er Afioc MP C5502a; Ricoh, Tokyo, Japan). The dental
casts were not used for direct data collection to prevent
damage during repeated measurements. Centroids
of the crowns of the canines, first premolars, and first
molars were located in the occlusal plane according to
the method of Moyers et al.39 The centroid of a dental
crown in the occlusal plane is defined as the point
halfway between 2 points calculated by joining the 2
approximal midpoints and the buccal and lingual mid-
points. The corresponding centroids were used for
intra-arch linear measurements. Calibrated digital
sliding calipers were used for all measurements includ-
ing maxillary intercanine width (MxIC), maxillary inter-
first premolar width (MxIPM), maxillary interfirst molar
width (MxIM), mandibular intercanine width (MdIC), man-
dibular interfirst premolar width (MdIPM), and mandib-
ular interfirst molar width (MdIM). When permanent
teeth were absent or unerupted, their deciduous counterparts
were used as substitutes.

All cephalometric and dental cast measurements
were made by the same investigator (V.K.), who was
blinded to the results of the 2 questionnaires. Ten radi-
ographs and study models were chosen at random and
analyzed at least 2 weeks apart to calculate the error
of the method. Intraclass correlation coefficients (ICC)
were calculated using a 2-way mixed model and abso-
lute agreement type for all angular and linear cephalo-
metric variables. ICC values varied from 0.973 to 0.997
for the angular cephalometric measurements, from
0.912 to 0.981 for the linear cephalometric measure-
ments, and from 0.992 to 0.998 for the dental cast mea-
surements. This indicates a satisfactory level of
intraobserver reliability.

Statistical analysis

Sample-size calculations were done a priori using
cephalometric variables (ANB angle, FH–MP angle, and
PNS-AD1) from previous meta-analyses.21,22 The
calculated power of the study exceeded 0.90 at an
alpha of 0.05, with sample sizes of the examined
groups from 55 to 70 subjects.

All data were analyzed using SPSS Statistics for
Windows software (version 21; IBM, Armonk, NY). The
assumptions behind each statistical test were assessed
and validated. Data are presented as means and the
standard deviations for continuous variables, and fre-
quencies or percentages for categorical variables. Pear-
son correlations were performed to check the
associations between the questionnaire scores and
patient data. The differences between the 2 groups for
continuous variables at T1 were tested for statistical sig-
nificance with a t test for independent samples and for
matched pairs with a paired-sample t test. The high-
risk and low-risk participants were further subdivided
by age at T1 into young children (ages, 8–12.9 years)
and teenagers (ages, 13–18 years) for subgroup analyses.
When an independent t test indicated a significant
difference, subgroup analysis was performed to enable
greater resolution of the results using univariate analysis
of variance test by pairwise comparison with the Bonfer-
roni correction applied. Odds ratios were calculated for
exposure and outcome categorical variables, and signif-
ance was tested with a 2 × 2 chi-square test. Statistical
significance was assessed at P < 0.05 (2 tailed). The data
at T1 from the 3 excluded children were analyzed sepa-
рately in a sensitivity analysis.

RESULTS

The mean age of the final cohort of 78 children at T1
was 12.3 ± 2.5 years (range, 8.3–17.6 years). Sixty-eight
children (87.1%) were white. Sixty (76.9%) question-
naires were completed by mothers, 10 (12.8%) by fa-
thers, and 8 (10.3%) by guardians or grandparents of
the children. The low-risk group (control) comprised
23 boys and 33 girls, and the high-risk group comprised
10 boys and 12 girls. No child in the low-risk group was
reported as a habitual snorer based on the 2 question-
naires.

Demographic data at T1 for both groups are presented in
Table I. Quality of life scores in the high-risk group
were 39.9 ± 15.6 compared with the low-risk group at
22.6 ± 6.9, indicating worsening in SDB-related quality
of life by 16% in the high-risk group (P < 0.0001). The
prevalence of a palatal crossbite involving at least 3 teeth
was significantly higher in the high-risk group at 68.2%
than in the low-risk group at 23.2% (P < 0.0001). At
T1, the OSA–18 Quality of Life scores and the PSQ scores
were correlated highly with each other (r = 0.81; P < 0.0001),
but the perceived quality of life correlated moderately
with OSA–18 Quality of Life scores (r = −0.61; 
P < 0.0001) and PSQ scores (r = −0.53; P < 0.0001). The
frequencies of positive responses to the PSQ survey are shown in
Table II. Odds ratios and their 95% confi-
dence intervals (CI) with statistical significance are also
presented in Table II for a palatal crossbite involving at
least 3 teeth and its association with each question of
the PSQ.
Table III summarizes the statistically significant differences in cephalometric and dental cast variables found between the 2 groups. The mean IAS, PNS-AD1, and AD1-SP values were lower in the high-risk group compared with the low-risk group by 1.87 mm (P < 0.03), 2.82 mm (P < 0.04), and 2.13 mm (P < 0.03), respectively. There were highly statistically significant differences between the groups in all width measurements except for MdIM (P = 0.20). The mean MxIC, MxIPM, MxIM, and MdIPM widths were reduced in the high-risk group compared with the low-risk group by averages of 4.22 mm (P < 0.0001), 3.92 mm (P < 0.0001), 4.24 mm (P < 0.0001), 1.50 mm (P < 0.01), and 1.84 mm (P < 0.01), respectively.
Table IV provides a summary of quality of life changes after maxillary expansion at T2. There was a statistically significant difference before and after RME in the OSA-18 Quality of Life scores between the groups, indicating an average improvement of 14% (mean score, 15.2 – 13.8) for children in the high-risk group compared with the low-risk group, which had a slight worsening in quality of life by an average of 1% (mean score, 1.2 – 3.9) (P > 0.04). At T2, the perceived quality of life changes did not correlate with the calculated OSA-18 score changes (r = −0.62; P = 0.06).

Subgroup analyses by age (young children and teens) and risk category (high and low risks) enabled greater resolution of the associations between risk for SDB and dental cast variables (Fig 3). Narrower MxIC, MxIPM, and MxIM widths indicated an increased risk of SDB across all age groups, whereas the associations of MdIC, MdIPM, and MdIM widths were not significant in the groups. Mean MdC width less than 27 mm was highly associated with an increased risk for SDB in young children (P <0.01) and teens (P <0.0001), whereas MdIC width less than 24 mm, on average, was associated with an increased risk for SDB only in young children (P <0.05). Mean MxIPM and MxIM widths had a high predictive value in the young (P <0.01) and teen (P <0.0001) subgroups. Mean MdIPM width was associated with an increased risk for SDB only in the teen subgroup (P <0.02). The results of the sensitivity analyses showed no statistically significant differences in any analyzed variable at T1 by the inclusion of the 3

**Table III. Summary of differences between high-risk (HR) and low-risk (LR) groups for cephalometric and dental cast variables at T1**

<table>
<thead>
<tr>
<th>Variable</th>
<th>Mean difference between groups (LR – HR)</th>
<th>95% CI</th>
<th>Significance P value</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Cephalometric analysis (n = 72)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cranial base analysis</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>S-N (mm)</td>
<td>1.22</td>
<td>−0.68-3.13</td>
<td>0.20 (NS)</td>
</tr>
<tr>
<td>N-S-Ba (°)</td>
<td>−0.10</td>
<td>−3.06-2.86</td>
<td>0.95 (NS)</td>
</tr>
<tr>
<td>Maxillary and mandibular skeletal analysis</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SNA (°)</td>
<td>−0.19</td>
<td>−2.07-1.68</td>
<td>0.84 (NS)</td>
</tr>
<tr>
<td>SNB (°)</td>
<td>0.21</td>
<td>−1.40-2.81</td>
<td>0.51 (NS)</td>
</tr>
<tr>
<td>ANB (°)</td>
<td>−0.66</td>
<td>−2.44-1.11</td>
<td>0.46 (NS)</td>
</tr>
<tr>
<td>Wits (mm)</td>
<td>−0.87</td>
<td>−3.26-1.53</td>
<td>0.47 (NS)</td>
</tr>
<tr>
<td>Co-A (mm)</td>
<td>1.18</td>
<td>−1.73-4.10</td>
<td>0.42 (NS)</td>
</tr>
<tr>
<td>Co-Gn (mm)</td>
<td>−0.12</td>
<td>−4.29-4.04</td>
<td>0.95 (NS)</td>
</tr>
<tr>
<td>Mx-Md (mm)</td>
<td>−1.30</td>
<td>−4.50-1.90</td>
<td>0.42 (NS)</td>
</tr>
<tr>
<td>Vertical skeletal analysis</td>
<td></td>
<td></td>
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<tr>
<td>SN-PP (°)</td>
<td>0.35</td>
<td>−1.31-1.99</td>
<td>0.68 (NS)</td>
</tr>
<tr>
<td>FH-MP (°)</td>
<td>−0.51</td>
<td>−3.45-2.43</td>
<td>0.73 (NS)</td>
</tr>
<tr>
<td>PP-MP (°)</td>
<td>−1.34</td>
<td>−4.57-1.90</td>
<td>0.41 (NS)</td>
</tr>
<tr>
<td>Co-Go (mm)</td>
<td>0.28</td>
<td>−2.42-2.97</td>
<td>0.84 (NS)</td>
</tr>
<tr>
<td>ANS-Me (mm)</td>
<td>−0.77</td>
<td>−3.67-2.12</td>
<td>0.59 (NS)</td>
</tr>
<tr>
<td>Dental analysis</td>
<td></td>
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</tr>
<tr>
<td>U1-SN (°)</td>
<td>−0.68</td>
<td>−5.18-3.81</td>
<td>0.76 (NS)</td>
</tr>
<tr>
<td>L1-MP (°)</td>
<td>2.26</td>
<td>−2.33-6.85</td>
<td>0.33 (NS)</td>
</tr>
<tr>
<td>OP-FH (°)</td>
<td>0.75</td>
<td>−1.67-3.16</td>
<td>0.54 (NS)</td>
</tr>
<tr>
<td>Airway analysis</td>
<td></td>
<td></td>
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<tr>
<td>IAS (mm)</td>
<td>1.87</td>
<td>0.24-3.49</td>
<td>&lt;0.03*</td>
</tr>
<tr>
<td>PNS-AD1 (mm)</td>
<td>2.82</td>
<td>0.26-5.39</td>
<td>&lt;0.04*</td>
</tr>
<tr>
<td>AD1-SP (mm)</td>
<td>2.13</td>
<td>0.34-3.92</td>
<td>&lt;0.02*</td>
</tr>
<tr>
<td>Hy-MP (mm)</td>
<td>−0.61</td>
<td>−3.68-2.47</td>
<td>0.69 (NS)</td>
</tr>
<tr>
<td>Dental cast analysis (n = 67)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maxillary arch widths</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>MxIC (mm)</td>
<td>4.22</td>
<td>2.73-5.70</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>MxIPM (mm)</td>
<td>3.92</td>
<td>2.31-5.53</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>MxIM (mm)</td>
<td>4.24</td>
<td>2.57-5.91</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>Mandibular arch widths</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>MdIC (mm)</td>
<td>1.50</td>
<td>0.46-2.53</td>
<td>&lt;0.01*</td>
</tr>
<tr>
<td>MdIPM (mm)</td>
<td>1.84</td>
<td>0.55-3.13</td>
<td>&lt;0.01*</td>
</tr>
<tr>
<td>MdIM (mm)</td>
<td>0.98</td>
<td>−0.54-2.50</td>
<td>0.20 (NS)</td>
</tr>
</tbody>
</table>

NS, Not significant.
*P <0.05; †P <0.001; ‡P <0.01.
previously excluded children, except for an increase in prevalence of children at high risk for SDB; this changed from 22 (28.2%) to 24 (29.6%) children.

**DISCUSSION**

The prevalence of children at high risk for SDB in the orthodontic sample was 28.2%, higher than the estimated 10% from a recent questionnaire-based study by Huynh et al 25 with a large sample of 604 children. This might have been due to different sample sizes, study methodologies, questionnaires, and evaluation and scoring methods used in the 2 studies. At T1, children in the high-risk group showed a worse SDB-related Quality of Life score by approximately 16% compared with children in the low-risk group. The frequency of palatal crossbite involving at least 3 teeth was significantly higher in the high-risk group at 68.2% compared with the low-risk group at 23.2%. A palatal crossbite involving at least 3 teeth most likely suggests a transversely narrow maxilla.40 Children with this were 4 times more likely to be frequent and loud snorers, and 6 to 12 times more likely to have heavy breathing or troubled breathing at night. They were also more likely to wake up with a dry mouth, have morning headaches, and have parent-reported behavioral concerns such as “not listening when spoken to directly” and being “constantly on the go.”

Our inclusion criteria had a wide age range to reflect routine orthodontic practice, making this study clinically applicable. Although overall a priori sample-size calculations for the cephalometric variables were met, the 2 study groups were unequal in numbers of subjects.

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**Table IV. Statistical analyses after maxillary expansion (T2) for children at high risk (HR) and low risk (LR or control group) for pediatric SDB**

<table>
<thead>
<tr>
<th>At RME removal</th>
<th>HR group n = 5 Mean ± SD</th>
<th>LR group n = 5 Mean ± SD</th>
<th>Significance P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (y)</td>
<td>11.37 ± 1.64</td>
<td>10.45 ± 0.83</td>
<td>0.30 (NS)</td>
</tr>
<tr>
<td>OSA-18 QoL score</td>
<td>28.40 ± 13.35</td>
<td>22.00 ± 2.45</td>
<td>0.32 (NS)</td>
</tr>
<tr>
<td>OSA-18 QoL parental score</td>
<td>9.00 ± 1.00</td>
<td>8.80 ± 1.10</td>
<td>0.77 (NS)</td>
</tr>
<tr>
<td>After RME QoL score change (T2–T1)</td>
<td>−15.20 ± 13.83</td>
<td>1.20 ± 3.96</td>
<td>&lt;0.04</td>
</tr>
<tr>
<td>After RME QoL parental score change (T2–T1)</td>
<td>1.80 ± 1.64</td>
<td>−1.10 ± 1.75</td>
<td>&lt;0.03</td>
</tr>
</tbody>
</table>

QoL, Quality of life; NS, not significant.

*Negative value implies improvement; †P <0.05; ‡negative value implies worsening.

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**Fig 3.** Analyses of transverse dental cast measurements (millimeters) by pairwise comparisons of high-risk and low-risk patients in the 8 to 12.9 and 13 to 18 years subgroups. A Bonferroni correction was applied to control type I errors. Error bars indicate 1 SD. ns, Not significant.
This could be why the study was underpowered in the high-risk group, and hence the finding of no significant differences in cephalometric measurements between the high-risk and low-risk groups. However, the sample determination was based on previous studies of children with OSA because of a lack of available standardized cephalometric data for children at the snoring end of the SDB spectrum. The absence of statistically significant vertical and sagittal skeletal differences as measured on cephalometric films in our sample is in contrast to 2 meta-analyses that reported that children with SDB showed an increased ANB angle caused by mandibular retrognathia by a marginally clinically significant value of 1.6° and an increased mandibular plane angle of approximately 4° when compared with the controls.  

The primary studies chosen in those meta-analyses included polysomnography-proven OSA, whereas the children in our sample were more likely to be at the snoring end of the SDB spectrum. In addition, most primary studies in those meta-analyses were considered to have low to moderate quality primarily because of lack of blinding. Our results support those of Schiffman et al., who found no differences in mandibular length, width, area, or volume as measured by magnetic resonance imaging between children with OSA and the controls. At T1, the 2 sample groups differed in airway dimensions (PNS-AD1, AD1-SP, and IAS), which were reduced by approximately 2 to 3 mm in the high-risk group compared with the low-risk group. The reduction of the nasopharyngeal dimension (PNS-AD1) in the high-risk group was on average 2.8 mm; this agrees with the meta-analysis by Katyal et al.  

Without any clinically significant sagittal and vertical craniofacial skeletal disharmony, this finding might be due to either adenotonsillar hypertrophy or a thicker than usual soft palate.

Dental cast analyses confirmed that the high-risk group had a reduced transverse maxillary dimension by an average of 4 mm in all measured widths and approximately 1.5 mm in the MdIC and MdIPM areas. Löfström-Tidestrom et al. and Pirilä-Parkkinen et al. reported reduced maxillary widths by 2 mm between OSA-affected children and controls; this was slightly lower than in our study. We found no differences in intermandibular transverse widths. However, the highly statistically significant reductions of mandibular intercanine and interfist premolar widths by approximately 1.5 mm in this study can be regarded as having low clinical significance. We found no significant reduction in MdIM width, whereas Cozza et al. reported reduced MdIM width by an average of 2 mm in OSA-affected children. This might have been due to different study populations and facial skeletal patterns. Subgroup analysis showed a highly significant predictive value of MxIC, MxIPM, and MxIM in assessing SDB risk in all age categories, whereas MdIC was significantly predictive only in the young subgroup and MdIPM in the teen subgroup. MdIM measurements were not associated with increased risk for SDB in any age category. Since intercanine widths stabilize much earlier than other dentoalveolar transverse dimensions, it might be a useful clinical predictor across different age groups.

Our data suggest that children between 8 and 17 years of age with MxIC width less than 27 mm, on average, are at high risk for SDB, and this could be used clinically as an efficient screening tool.

To our knowledge, a change in SDB-related Quality of Life scores for children treated with RME at the milder end of the SDB spectrum has not been reported previously. Although the sample size at T2 was small and caution is recommended in interpretation of such data, this study shows an improvement in SDB-related Quality of Life scores by an average of 14% with an RME device in the high-risk group compared with the low-risk group, which showed a slight worsening by 1%. The worsening in the low-risk group might have been due to the appliance, which reduces inraoral volume and affects oral hygiene maintenance and speech. The improvement in the OSA-18 Quality of Life scores after RME in the high-risk group can be regarded as having some clinical significance as these children were “normalized” in the SDB-related Quality of Life scores, which were comparable with those of the low-risk group at T1. However, long-term follow-up and a larger sample size are required to assess the stability of such changes in SDB-related Quality of Life after RME treatment. The prevalence of pediatric SDB can change with time. Marcus et al. reported normalization of polysomnography scores in nearly 47% of the OSA-affected children randomized to watchful waiting for 7 months compared with the adenotonsillectomy surgery group. This might have been due to growth of the airway, regression of the lymphoid tissue, routine medical care, or regression to the mean. Interestingly, at T2, a change in the perceived quality of life did not correlate well with the calculated OSA-18 Quality of Life score change. This could have been caused by parental attitudes toward the overall health of their children rather than the SDB-related effects on quality of life.

The floor of the nose and the maxillary vault are anatomically related. When the midpalatal suture is opened by RME, the nasal cavity’s lateral walls are also displaced laterally; this increases nasal volume and decreases upper airway resistance. This increase in nasal cavity width after RME might be a reason for the increase in total pharyngeal and retropalatal airway volumes in the children treated with RME. The
changes after RME, as measured by objective tests of nasal airway patency such as rhinomanometry and acoustic rhinometry, show improved pressures for nasal breathing up to 11 months after RME. Maxillary constriction can also lead to decreased oral volume because of a lower tongue position that might decrease further in a supine sleeping position. This lowered tongue posture has been shown to improve after maxillary expansion. Since maxillary width changes little after adenotonsillectomy, orthodontic widening of a narrow maxilla in patients whose snoring persists or relapses after adenotonsillectomy is gaining support.

Many therapies might act synergistically in treating pediatric SDB, which is a complex multifactorial problem. Management of the child with a suspected diagnosis of primary snoring or OSA should consider the severity of upper airway obstruction during sleep, and the presence of morbidity or other coexisting conditions. There is no consensus presently; however, Kaditis et al proposed an integrated and hierarchical stepwise evidence-based algorithm for the diagnosis and multitherapeutic management of childhood SDB. This approach starts with weight control followed hierarchically with nasal corticosteroids, adenotonsillectomy, dentofacial orthopedics such as mandibular advancement or maxillary expansion, continuous positive airway pressure, and maxillofacial surgery.

One drawback of our study is the reliance on the 2-dimensional lateral cephalograms to assess 3-dimensional structures. The intraobserver reliability values for landmark identification and measurements were high, but errors in projection and anatomic interpretations might have been overlooked because these are inherent problems of the technique. Although Major et al found that there was at best a moderate correlation (r = 0.68) between linear measurements of the upper airway in a cephalometric film and the diagnosis of upper airway blockage, Pirila-Parkkinen et al showed that the cephalometric film is a reliable tool to measure nasopharyngeal and retropalatal dimensions but not oropharyngeal width in children with adenotonsillar hypertrophy. Nevertheless, it is a valid screening tool with greater accessibility, lower costs, and lower radiation doses than 3-dimensional volumetric cone-beam computed tomography in an orthodontic setting. It also remains controversial whether a lateral cephalogram should be taken in an upright or a supine position to screen for pediatric SDB; however, it has been shown that the state of consciousness might be a more important factor affecting upper airway muscle tone rather than head position. An issue in this study stems from parental reporting of their children’s SDB-related symptoms and quality of life. Although no identified study has compared parental reporting of SDB symptoms with children’s self-reported perceptions, relatively high agreement between parental and self-reported childhood sleep symptoms related to attention deficit hyperactivity disorder have been reported. Since most of our subjects were white, the results might not apply to other ethnicities. Other limitations of this study include failure to record adenotonsillar size clinically, inaccessibility to polysomnography for diagnosis of pediatric SDB, the small sample size at T2 having RME treatment, and a short follow-up of the children treated with RME.

Future research should include 3-dimensional assessment of craniofacial morphology and the nasopharyngeal and oropharyngeal airways to help in further understanding such anatomic factors related to pediatric SDB, particularly in refractory patients. In addition, future studies should compare changes in SDB-related quality of life changes for various treatment modalities in the management of pediatric SDB. There is an urgent need for research to establish protocols for multitherapies to account for the relative contributions of each therapy in the management of pediatric SDB. More collaboration between sleep medicine physicians, ear, nose and throat surgeons, and orthodontists is required to establish individualized approaches for successful treatments or cures.

CONCLUSIONS

Children at high risk for SDB are characterized by reduced SDB-related quality of life, reduced nasopharyngeal and oropharyngeal sagittal dimensions, palatal crossbite, and reduced dentoalveolar transverse widths in the maxillary and mandibular arches. No sagittal or vertical craniofacial skeletal cephalometric predictors were identified for children at high risk for SDB. In the short term, RME might aid in the improvement of SDB-related quality of life for children with a narrow maxilla at the milder end of the SDB spectrum. Long-term follow-ups and larger sample sizes of children treated with RME at risk for SDB are required to confirm and assess the stability of the changes seen.

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REFERENCES