Randomized Controlled Study of an Oral Jaw-Positioning Appliance for the Treatment of Obstructive Sleep Apnea in Children with Malocclusion

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To evaluate the clinical usefulness and tolerability of an oral jaw-positioning appliance in the treatment of obstructive sleep apnea syndrome in children, we studied 32 patients (mean age, 7.1 ± 2.6 yr; 20 males) with symptoms of obstructive sleep apnea, malocclusion, and a baseline apnea index > 1 event/h. A group of 19 subjects was randomly assigned to a 6-mo trial of an oral appliance; the remainder acted as control subjects. At baseline and after the trial all patients underwent physical examination, a standard polysomnography, and orthodontic assessment. A modified version of the Brouillette questionnaire related to obstructive sleep apnea symptoms was administered to parents before and after the trial and a clinical score was calculated. Of the 32 subjects enrolled, 4 treated subjects and 5 control subjects were lost to follow-up. Polysomnography after the trial showed that treated subjects all had significantly lower apnea index (p < 0.001) and hypopnea index values (p < 0.001) than before the trial, whereas in untreated control subjects these values remained almost unchanged. Clinical assessment before and after treatment showed that in 7 of the 14 subjects (50%) the oral appliance had reduced (a fall of at least 2 points in the respiratory score) and in 7 had resolved the main respiratory symptoms, whereas untreated patients continued to have symptoms. In conclusion, treatment of obstructive sleep apnea syndrome with an oral appliance in children with malocclusion is effective and well tolerated.

Keywords: children; malocclusion; obstructive sleep apnea; oral appliance

Although the most common treatment for obstructive sleep apnea syndrome (OSAS) in childhood is adenotonsillectomy, this approach is limited by its surgical risks (1) and, in some patients, by recurrence (2). Children who do not improve after adenotonsillectomy tend to have a narrower epipharyngeal air space, a more poorly developed maxilla, and mandibular retrusion (3). Nasal continuous positive airway pressure (nCPAP) is an effective and safe treatment for OSAS, but is generally used in the more severe cases (4) and is limited by parents' and patients' acceptance (5). In a study conducted in a small number of children with OSAS and adenotonsillar hypertrophy, Brouillette and coworkers (6) reported that a 6-wk course of nasal corticosteroids decreased the frequency of apnea, hypopnea, and desaturation events. The long-term efficacy of and tolerance to nasal corticosteroid therapy unfortunately remains unclear.

A useful alternative therapy for patients with sleep-disordered breathing, currently studied only in adults, is the use of an oral appliance (7–10). Despite their varying designs and mechanisms of action, advancing the mandible or the tongue or both (11–13), all these appliances induce their therapeutic effect by enlarging the upper airway. In 73–100% of adult patients oral appliances reportedly reduce snoring and in 50–100% improve polysomnographic indexes, decreasing the apneahypopnea index (AHI) by at least 50% from baseline (14). Whereas several studies found an oral device less effective in adult patients with severe disease (15–17), others reported that the device normalized the apnea index also in patients with severe OSAS (18). Although oral appliances are generally less effective than nCPAP in eliminating OSAS, adult patients tolerate them better (19, 20).

No data exist on the use of oral jaw-positioning appliances in the management of sleep-related respiratory disturbances in children. Nor is it known whether children will tolerate these devices. Guilleminault and coworkers suggest that children with retroposition of the mandible, steep mandibular plane, high hard palate, long oval-shaped face, or long soft palate were highly likely to have sleep-disordered breathing (21). Children with habitual snoring and OSAS have a special morphological craniofacial type, showing a high angle and an accompanying increase in both upper and lower gonion angles (22). This 6-mo randomized controlled trial was aimed at assessing the clinical usefulness of and tolerance to a personalized oral jaw-positioning appliance for the treatment of childhood OSAS accompanied by malocclusion.

METHODS

The 32 children (age range, 4 to 10 yr; mean age, 7.1 ± 2.6 yr; 20 males) who participated in the study were selected from among patients referred to our Pediatric Sleep Disorders Center (Rome, Italy) for symptoms of OSAS, who had an apnea index (AI) of > 1 event/hr of sleep during diagnostic polysomnography and who had evident clinical signs of dysgnathia.

The parents of all participants completed a modified version of the Brouillette questionnaire on the symptoms of OSAS (23). The questions sought information about the child’s daytime symptoms (including sleepiness, irritability, tiredness, school problems, morning headache, morning thirstiness, oral breathing, and nasal stuffiness) and nighttime symptoms (including habitual snoring, apneas, restless sleep, and nightmares). Each respiratory symptom (oral breathing, nasal stuffiness, snoring, and apnea) was assigned a score of one point (maximum score, four points). All participants also underwent a complete physical examination to exclude the presence of an acute upper airway infection and to assess the presence of tonsillar hypertrophy according to clinical criteria (tonsillar size, scored on a scale ranging from 0 to 4: 0 = tonsils not visible, 4 = tonsils touching).

From among the 32 children recruited, 19 subjects (mean age, 6.86 ± 2.34 yr; 10 males) were randomly assigned alphabetically, by surname, to undergo a 6-mo trial of a personalized oral appliance. The remaining 13 subjects (mean age, 7.34 ± 3.10 yr; 10 males) acted as a control group and did not undergo therapy.

After the 6-mo trial, the children’s parents again completed the same questionnaire and children underwent a new physical examination and standard overnight polysomnography.

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To meet the parents’ wishes and in accordance with the American Sleep Disorders Association (13), none of the subjects underwent cephalometry.

On recruitment, all children underwent an orthodontic assessment and clinical inspection of mandibular posture, on the three spatial planes (sagittal, horizontal, and frontal) to detect possible jaw deviation from normal occlusion: deep bite, retrusive bite, and cross-bite. To evaluate the contact relationship of the occlusal surfaces of the upper and lower teeth and to obtain the information needed to prepare a personalized oral appliance, an alginate impression was taken of the two dental arches. A wax check bite was obtained to identify the degree of correction needed for the maximum alignment of the maxillary and mandibular basal bone. During the wax bite procedure, the operator held the two dental arches in the position considered ideal for correcting the patient’s mandibular anomaly. The wax bite therefore served as a guide for constructing the oral appliance that treated patients had to wear.

**Oral Appliance**

All children in the treatment group were fitted with an acrylic resin oral bite plate for mandibular positioning. The orthodontist designed an oral appliance for use in children (Figure 1A–C). All appliances were designed to correct each patient’s mandibular malpositioning compared with the specific reference occlusal planes: a receding bite was therefore advanced, a deep bite raised, and cross-bite recentered (Figure 2). Each appliance also had a lingual “target” (Figure 1B, b1), an acrylic ring that stimulated the child’s tongue toward its proper position on the palatine folds immediately behind the upper incisors.

Each child chose the color of their appliance: most of our patients chose from among the colors of the local football team. Because the treatment served not only to solve the sleep-disordered breathing but also to correct the orthodontic defect, children were required to wear the appliance continuously except at mealtimes. Treated children who had side effects or failed to comply with instructions for using the oral device, and control subjects who refused or whose parents refused on the basis of unforeseen circumstances to allow them to continue the study, dropped out.

Patients treated with an oral appliance were assessed monthly by the orthodontist to monitor functioning of the oral appliance and to resolve any problems caused by eventual changes in dental structure and occlusal position.

Tolerance was assessed by the orthodontic specialist who questioned patients, during the monthly assessment, about difficulties in using the oral appliance as prescribed.

**Polysomnography**

All subjects underwent a standard overnight polysomnography performed with a Grass multichannel instrument (model Heritage; Grass Instruments, Quincy, MA). Polysomnographic data included a standard montage consisting of two-channel electroencephalograms (C4/ A1, C3/A2), electro-oculograms, a submental electromyogram, bilateral leg, and an electrocardiogram. Abdominal and chest movements were measured by inductive plethysmography. Airflow was monitored with an oronasal thermocouple transducer (model F-ONTT; Grass Instruments). Arterial blood oxyhemoglobin saturation (SaO2) was recorded with a pulse oximeter. Polysomnography variables were scored according to the criteria of Rechtschaffen and Kales (24) and to the international criteria of the American Thoracic Society for sleep studies in children (25).

The local ethics committee approved the study protocol and all children’s parents gave their informed consent to the procedures.

**Statistical Analysis**

Data were analyzed with the SPSS (Chicago, IL) software program. Data are expressed as means ± the standard deviation (SD). Pretreatment versus posttreatment values were analyzed with two-tailed paired and unpaired Student t tests and Pearson χ2 analysis. The significance level was set at p = 0.05.

**RESULTS**

All subjects had normal height and weight for age and none of them were obese. Most patients (87%) had deep or retrusive bite or both dysfunctions (Table 1). No significant differences were found between polysomnographic values for subjects who completed the trial and those who dropped out. No patients had acute medical illnesses at the time of recruitment, nor were significant differences found in the frequency of symptoms reported in answer to the questionnaire or the clinical score for daytime and nighttime symptoms at baseline (before the trial) between patients who underwent the trial and control subjects who did not.

Of the 19 subjects who had appliances fitted, 5 (26%) subsequently abandoned therapy; and of the 13 control subjects, 4 (31%) preferred to discontinue the study and were unavailable for the 6-mo assessment. All the 23 subjects who completed the study (14 treated and 9 untreated control subjects) had a similar frequency of tonsillar hypertrophy (clinical scale ≥ 2) at baseline (12 of 14, 85.7% versus 7 of 9, 77.8%; p = NS). After the 6-mo trial, 8 of the 12 treated subjects with adenotonsillar hypertrophy (66.7%) but only 1 control (14.3%) had a reduction in tonsillar hypertrophy (66.7 versus 14.3%, χ2 = 54.8, p < 0.001).

Answers to the questionnaires administered after the 6-mo treatment trial showed that daytime and nighttime symptoms in treated subjects diminished but in control subjects remained unchanged (Table 2).

After the 6-mo trial, the respiratory symptoms in all patients improved (a fall of at least two points in the score) and in 50% of patients completely regressed (score = 0), whereas in control subjects they remained unchanged.

Polysomnograms obtained after the 6-mo trial showed that in treated children the AI and AHI were significantly lower after the trial (p < 0.001), whereas in control children both respiratory variables remained unchanged. In 9 of the 14 treated subjects (64.2%) the AHI fell by at least 50% (Figure 3). The desaturation index (DI) decreased in treated patients, but the difference failed to reach significance.
Of the 19 children fitted with the oral appliance, 14 (73.7%) tolerated the treatment well and only 5 discontinued therapy. A few patients reported excessive salivation that lasted only briefly. Of the five children (26%) who discontinued therapy, one child found the oral appliance intolerable because putting it into the mouth triggered violent, uncontrollable coughing that stopped only when the appliance was removed. Because this reaction raised a suspicion of an allergy to the metal present in the device, the metal hooks were replaced with acrylic fibers. Although the new device stopped the coughing for a few days, the symptom reappeared and the child refused definitively to continue treatment. Two children claimed that they lost their appliances three times and then refused to wear them again. Two children found wearing the oral appliance at school embarrassing and discontinued therapy.

During monthly orthodontic assessment, none of the patients reported temporomandibular joint discomfort, nor did the orthodontist find pathological changes in occlusal alignment.

**DISCUSSION**

The distinguishing feature of our report is that it provides hitherto unavailable information about the use of a personalized oral jaw-positioning appliance in the treatment of OSAS in children. In this study, children with OSAS and dysgnathia

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<th>TABLE 1. ANTHROPOMETRIC VALUES AND ORTHODONTIC CHARACTERISTICS OF THE STUDIED SUBJECTS*</th>
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* Definition of abbreviation: BMI = Body mass index.
* Data for age, weight, height, and BMI are expressed as means ± SD; orthodontic characteristics are expressed as a percentage of the total category subjects.
who completed the 6-mo trial of an oral appliance for mandibular positioning and wore the device continuously, except at mealtimes, had significantly lower AI and AHI after treatment.

Among the various indexes and cutoff points used to define successful treatment of OSAS with an oral appliance in adults, for this study of children we considered appropriate a decrease of at least 50% in the AHI. According to this definition, treatment with the personalized oral appliance used in this study achieved a success rate of 64.2%, even though it did not return the respiratory variables of our patients to normal pediatric reference values.

Many of the oral appliances used in adults induce their therapeutic effect by increasing the anteroposterior diameter of the retroglossal space through anterior displacement of the jaw and tongue, thereby reducing the degree of pharyngeal collapse (14). Schwab and coworkers provided evidence of jaw and tongue, thereby reducing the degree of pharyngeal changes in a study using magnetic resonance imaging, showing collapse (14). Schwab and coworkers provided evidence of jaw and tongue, thereby reducing the degree of pharyngeal changes in a study using magnetic resonance imaging, showing collapse (14).

The therapeutic rationale is that all orthodontic anomalies (except Class III) benefit from mandibular advancement capable of enlarging the retrolingual space and at the same time improving tongue posture could well have enlarged the retropharyngeal space, thus making the tonsils look smaller. By encouraging children to breath through the nose, treatment may also have reduced respiratory infections. Among the various indexes and cutoff points used to define successful treatment of OSAS with an oral appliance in adults, for this study of children we considered appropriate a decrease of at least 50% in the AHI. According to this definition, treatment with the personalized oral appliance used in this study achieved a success rate of 64.2%, even though it did not return the respiratory variables of our patients to normal pediatric reference values. Many of the oral appliances used in adults induce their therapeutic effect by increasing the anteroposterior diameter of the retroglossal space through anterior displacement of the jaw and tongue, thereby reducing the degree of pharyngeal collapse (14). Schwab and coworkers provided evidence of changes in a study using magnetic resonance imaging, showing that advancing the jaw increases the lateral and anteroposterior dimensions of the airways (26). The therapeutic rationale is that all orthodontic anomalies (except Class III) benefit from mandibular advancement capable of enlarging the retrolingual space and at the same time promoting lingual advancement. Conversely, the young patients with OSAS whom we studied all had defective dental occlusion. Rather than striving for mandibular advancement at all costs, we therefore designed a device that would reposition the dental arches, thus correcting occlusal anomalies in the three spatial planes. This therapeutic approach seemed to us indispensable because jaw advancement in a growing child could run the risk of misdirecting the plane of development. Evidence supporting the therapeutic approach we successfully used in this study comes from numerous reports. For example, children with certain orocraniofacial features and those with enlarged tonsils are at known risk of having some type of sleep-disordered breathing (21, 27, 28). Rees and coworkers have identified a phenotype characterized by micrognathia and retrognathia (28) and Guilleminault and coworkers (21) described a facial phenotype present in 34% of children with sleep-disordered breathing. In many of the children we studied (66.7%), therapy with the oral appliance reduced tonsillar hypertrophy. It presumably did so by enlarging the pharyngeal space so that the tonsils had more space and thus appeared smaller. By encouraging children to breathe through the nose, treatment may also have reduced respiratory infections.

Our oral appliance was designed to correct the individual patient’s occlusal or mandibular dys harmony or both faults and also to keep the tongue in its proper position. Hence, we fitted the device with a tongue target.
malocclusion is a predictive factor for the result of therapeutic intervention. Another unanswered issue is whether the initial orthodontic anomaly is a factor that influences the result. Because we preferred not to use cephalometry, neither can we say whether our therapeutic intervention brought about changes in facial structure. In contrast to the usual practice in clinical orthodontics, none of the children underwent cephalometry. Instead, they all had a detailed clinical orthodontic assessment.

In conclusion, treatment of OSAS with a personalized oral jaw-positioning appliance in children with malocclusion is effective and well tolerated. Essential matters to address are how long these benefits last and how long children can and must continue to wear the appliance.

References


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