**Management of obstructive sleep apnoea in children with modified monobloc appliances**

**P. COZZA**, R. GATTO**, F. BALLANTI*, L. PRETE*

**ABSTRACT.** This was to investigate the effect of the use of an orthodontic appliance in the treatment of obstructive sleep apnoea (OSA) in children using polysomnographic variables. Methods: 10 boys and 10 girls with OSA aged between 4 to 8 years, referred from an otolaryngology clinic because of sleep apnoea, wore modified monobloc devices nightly for 6 months. Polysomnography was used for each patient for baseline diagnosis of OSA and also for post therapy assessment. Results: The median obstructive apnoea-hypopnoea index decreased after 6 months of therapy with oral appliances. The mean (±SD) number of episodes of OSA was 7.88±1.81 before treatment and 3.66±6.80 after 6 months (p<0.001). Conclusions: The modified monobloc appliance is suggested for use in children with OSA and may be an effective therapeutic alternative in children with mild to moderate OSA.

**KEYWORDS:** OSAS, Child, Oral appliance, Polysomnography.

**Introduction**

Obstructive sleep apnoea (OSA) is a common chronic disorder of sleep and breathing. It is characterized by a number of symptoms: intermittent upper airway obstruction during sleep, socially handicapping snoring and excessive daytime sleepiness, with related harmful effects in cognitive functions, and increased risks of car accidents in adults [Lowe et al., 2000; Tangugsorn et al., 2001]. It is caused by partial or complete collapse of the pharyngeal airway during sleep due to a combination of a reduction in muscle tone at sleep onset and abnormal cervical-craniofacial structures such as retrognathia, micrognathia (skeletal relationship tending significantly more toward a Class II), increased lower facial height, reduced anteroposterior size of the bony pharynx, enlarged soft palate and enlarged tongue. A decreased posterior airway space (adenotonsillar hypertrophy) is the most common associated condition in otherwise normal children, together with inferior position of the hyoid bone [Bennet et al., 1998; Bernhold and Bondemark, 1998; Cozza et al., unpublished data].

With the recent interest in sleep apnoea a number of approaches have been tried. A recent review on the subject, with possible methods of therapy, has been published by Lindman and Bondemark [2001]. The methods include oral appliances of various designs [Denbar, 1998; Villa et al., 2002], surgery [Bell and Turvey, 2001] and nasal continuous positive airway pressure [Waters et al., 1995]. However, generally orthodontic type appliances have been proposed and studied and are increasingly used to treat sleep apnoea. Historically used in treating obstructive sleep apnoea appliances are, in terms of their construction design, predominantly derived from functional appliances and can be divided into two categories: mandibular advancing devices (MADs) and tongue retaining devices (TRDs) [Schmidt-Nowara et al., 1995; Hans et al., 1997; Bennet et al., 1998; Lowe et al., 2000; Petijean et al., 2000; Schoem, 2000]. MADs are designed to be attached to one or both dental arches, with the intention of advancing or downwardly rotating the mandible. The rationale for this movement is that the tongue is attached to the genial tubercles of the mandible and positioning the mandible forward moves the tongue forward as well. These mandibular repositioning appliances also change hyoid bone position and modify the lower airway space below the level of the base of the tongue [Hans et al., 1997; Schoem, 2000]. The TRD approach has the intention of securing the tongue by negative pressure into a soft plastic bulb, thereby forcibly holding the tongue anteriorly while sleeping [Schoem, 2000].

While a number of studies in the literature investigated the effects of dental appliances on adult OSA patients, there appear to be no studies on the use of appliances in...
children [Cartwright and Samelson, 1982; Cartwright, 1985; Cartwright et al., 1988; Rider, 1988; Schmidt-Nowara et al., 1991; Knudson et al., 1992; Knudson and Meyer, 1993; Schmidt-Nowara et al., 1995; Ferguson et al., 1996; Hans et al., 1997; Bernhold and Bondemark, 1998; Gale et al., 2000]. Accordingly the purpose of this study was to investigate the effect of using a modified monobloc appliance in the treatment of OSA children measured by polysomnographic variables.

Materials and methods

Subjects. The study group comprised 20 OSA Caucasian subjects (10 boys and 10 girls) with an age ranging from 4 to 8 years (mean age 5.91). The children were referred to the Department of Orthodontics from an otolaryngology clinic because of a history of apnoea. The study population was selected from a larger group of children, who were diagnosed with orthodontic problems, that required a mandibular advancing device, and with complete records. The inclusion criteria were that children should have been diagnosed as suffering from moderate to severe OSA at the otolaryngology clinic, should have no other contraindicating medical condition, nor a history of behavioural problems likely to lead to poor cooperation in the use of an orthodontic appliance and an informed consent from the parents was also required. Exclusion criteria included inadequate medical and dental records concerning the history of OSA, medical histories confounding possible treatment, and evidence of likely poor cooperation.

The mean body mass index (BMI) was 16.02. The demographic data are summarized in Table 1. All patients had a history of disturbed sleep characterized by recurrent apnoeic periods, as well as excessive daytime sleepiness and heavy snoring. All subjects had their diagnosis of OSA confirmed by overnight polysomnography. In addition, all OSA patients were screened with a validated questionnaire to assess the degree of symptoms of excessive daytime sleepiness.

The Italian version of the Epworth sleepiness scale (ESS) was used to assess excessive daytime sleepiness. The ESS, which asks patients to estimate the likelihood that they would doze off or fall asleep in a sedentary situation, is a simple self-administered questionnaire which has been shown to provide a measurement of a subject’s general level of daytime sleepiness [Johns, 1991; Johns, 1993; Vignatelli et al., 2003].

Diagnosis. The diagnosis of OSA can be confirmed only by overnight polysomnography. Nocturnal polysomnography, as opposed to afternoon nap assessment [Marcus et al., 1992], was recorded during sleep by a trained sleep laboratory technician as a screening procedure to verify that OSA was occurring. In this study polysomnography was, therefore, used for the initial screening and diagnosis of OSA and post therapy assessment. Calculated respiratory variables were AHI (the number of apnoeas and hypopnoeas for hours of sleep) and minimum arterial oxygen saturation (min SaO₂) during apnoeas. Apnoea was defined as cessation of airflow for at least 10 seconds. Hypopnoea was defined as a reduction in amplitude of airflow or thoraco-abdominal wall movement of greater than 50% of the baseline measurement for more than 10 seconds (O₂ desaturation need not occur), or the same reduction with an accompanying O₂ desaturation of at least 3% (no time limit), and associated with arousal.

Oral appliances. For each patient the intraoral appliance used was a modified monobloc (MM), fabricated with full tooth coverage in both arches and with a central screw. The screw was activated only to follow the maxillary transversal growth. The MM was designed like an activator to avoid undesirable anterior dental movements. The incisal edge and superior labial surfaces of the mandibular incisors were capped to prevent tipping.

The MM was produced from a construction bite that positioned the mandible anteriorly into an edge-to-edge incisal relationship. The lower jaw was postured forward to increase intermaxillary space. As a general rule, the bite registration was taken 3 mm short of maximum

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Mean</th>
<th>SD</th>
<th>Minimum</th>
<th>Maximum</th>
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</thead>
<tbody>
<tr>
<td>Age in years</td>
<td>5.91</td>
<td>1.14</td>
<td>4.00</td>
<td>8.00</td>
</tr>
<tr>
<td>Height in m</td>
<td>1.26</td>
<td>0.14</td>
<td>1.00</td>
<td>1.45</td>
</tr>
<tr>
<td>Weight in Kg</td>
<td>27.7</td>
<td>8.04</td>
<td>15.00</td>
<td>38.00</td>
</tr>
<tr>
<td>BMI* as kg/m</td>
<td>16.02</td>
<td>3.40</td>
<td>8.23</td>
<td>20.00</td>
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</tbody>
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*BMI = body mass index

| Table 1 - Baseline anthropometric data for a study population of children with OSA. |
protrusion, with care being taken to ensure that lateral displacement did not occur. The height of bite exceeded the freeway space by 2 to 3 mm. An example of the device is illustrated in Figure 1.

This device was fabricated from transparent acrylic resin, which is physiologically harmless, insoluble in water, odour free, and inactive (Fig. 1a). If necessary a lingual arch for anchorage for the Class II intermaxillary elastics was inserted to prevent any jaw opening. The splint was to be used with Tucat’s pearl with sliding wire for references for the tip of the tongue. Tucat’s pearl allows the placement of the tongue tip against the palatal aspect of the alveolar process, behind the maxillary incisors, to improve muscle function and the habitual position of the tongue [Cozza et al., 2002].

Following a one week habituation period (Fig. 1b), the subjects wore the device nightly for 6 months.

During treatment, contact was maintained between the appliance and the maxillary posterior teeth; the mandibular posterior teeth were encouraged to erupt by trimming acrylic occlusally and lingually to them. Appliances were checked at regular recall. A polysomnographic registration was carried out 6 months after appliance insertion.

Statistics. Descriptive statistics include mean and standard deviation. The mean differences in polysomnographic data between baseline (pre) and post therapy were examined using Wilcoxon’s matched paired test. The level of significance was set at p<0.05.

Results

Initial problems with use of MM include excessive salivation and discomfort at awakening after having a repositioning device in the mouth all night. These adverse effects were found to gradually diminish within a few days after beginning of treatment and following this period all children and their parents reported a good compliance with the MM. The appliances were, therefore, tolerated well. No dysfunctions of the dentition were noted.

The baseline data concerning the subjects in this study are shown in Table 1. The mean±SD of Epworth sleepiness scale (ESS) score in OSA patients before treatment was 15.2±4.9 and after therapy it was reduced to 7.1±2.0. The median obstructive apnoea-hypopnoea index decreased, after 6 months of therapy with an oral appliance, from 7.88 to 3.66 in all patients. The polysomnographic data are summarized in Table 2.

**Table 2** - Polysomnography results at baseline and after treatment of a group of children with obstructive sleep apnoea (OSA) with a modified monobloc appliance.

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Before</th>
<th>After</th>
<th>p</th>
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<tbody>
<tr>
<td></td>
<td>Mean±SD</td>
<td>Min</td>
<td>Max</td>
</tr>
<tr>
<td>AHI*</td>
<td>7.88±1.81</td>
<td>5.60</td>
<td>10.80</td>
</tr>
<tr>
<td>Arousal index</td>
<td>5.48±2.19</td>
<td>2.40</td>
<td>6.70</td>
</tr>
<tr>
<td>SaO2(%)**</td>
<td>97.4±0.66</td>
<td>96.0</td>
<td>98.00</td>
</tr>
</tbody>
</table>

AHI* = number of apnoeas and hypopneas per hours of sleep
SaO2(** = minimum arterial oxygen saturation
An example of the changes in lateral facial profile is shown in Figures 2a, b.

Statistical evaluation. A non parametric test was used because the study variables were not normally distributed. Statistical evaluation of the results showed that the MM was significantly more effective, when comparing polysomnography results, post treatment versus baseline readings, with respect to apnoea-hypopnoea index (AHI), but not to minimum oxygen saturation (min SaO₂). In addition the arousal index was also unaffected.

Discussion

The condition of sleep apnoea in children is distressing for both child and parent [Brouillette et al., 1982]. In a recent review on the subject, Guilleminault and Quo [2001] noted that OSA is often missed in general clinical practice. This is because the symptoms are insidious and it is not until they are marked that treatment is sought [Rosen et al., 1992]. Paediatric dentists and orthodontists have the greatest opportunity to see these young individuals and can formulate and implement treatment plans.

In a recent comprehensive review of the literature, covering 20 years, Lindman and Bondemark [2001] noted that there had been numerous reports and studies on various approaches to treatment of OSA. These authors concluded that there was not yet enough scientific evidence for a clinician to decide which appliance is more or less likely to improve symptoms for any given patient. However, on the evidence reviewed by Lindman and Bondemark, dental (orthodontic) appliances may have a place in the treatment of habitual snoring/apnoea in mild and moderate cases of OSA.

Functional appliances, such as the modified monobloc, are one solution, although not all authors agree that they are ideal. However, if appropriately used with a goal of enlarging the upper airway, they may help to prevent any possible surgical correction later. The question of OSA and orthodontic problems is an interesting one, as it has been suggested that orthodontic treatment of itself, such as the use of cervical headgear [Pirila-Parkkinen et al., 1999], may contribute to the occurrence of sleep apnoea. This may occur when there is a strong predisposition, such as when mandibular retrognathia to the development of upper airway occlusion already exists. Nevertheless, alternative approaches to treatment of OSA by orthodontic appliances are not all satisfactory.

The treatment of OSA depends upon the severity of symptoms, magnitude of clinical complications and aetiology of the upper airway obstruction and is directed toward improving the airflow by various surgical and non-surgical methods [Bernhold and Bondemark, 1998; Lowe et al., 2000]. There are a number of generally agreed specific indications for the use of oral appliances, which have been collated from the American Sleep Disorders Association and Sleep Research Society [ASDA, 1995; Levy et al., 1996]. These are:

- subjects with mild OSA who do not respond to or are not appropriate candidates for treatment with behavioural measures, such as weight loss or sleep position change;
- subjects with moderate to severe OSA who cannot tolerate or refuse treatment with nasal continuous positive air pressure (nCPAP);
- subjects who refuse or are not candidates for tonsillectomy and adenoidectomy, craniofacial operations or tracheostomy. The present study attempts to investigate the effects of a dental appliance (modified monobloc) treatment in children with OSA based on polysomnographic variables.

A good oral appliance designed for children favours bone and dentoalveolar changes during therapy, increases mandibular length and has effects on the vertical growth of the jaw, so it increases the intermaxillary space in which the tongue rests. It may have myofunctional rehabilitation in order to bring the tongue upward and forward. This appliance is routinely used in the Department of Orthodontics, University of Rome “Tor Vergata”, in the treatment of OSA.
The MM has been associated with cases of bruxism in children, although this was not seen in this study. However, the number of children treated was small. The only problem recorded by the parents was that of initial discomfort and salivation. But, as noted, these problems rapidly declined once the children had become used to wearing the appliance.

The rationale for selecting the MM appliance in OSA children was to increase the intermaxillary space, in which the tongue rests. In the present investigation, the MM significantly reduced the AHI score in children with sleep apnoea, whilst the minimum oxygen saturation remained unchanged. Furthermore, the MM reduced daytime sleepiness and subjectively improved the sleep quality.

In this study the use of a functional appliance seems to have been effective in controlling OSA. It is recognized that this approach is not the only one that can be used. Alternatives, such as nCAPD, have been tried with some success [Waters et al., 1995], but home cardiorespiratory monitoring devices less so [Poels et al., 2003]. However, various orthodontic approaches, such as palatal and transverse expansion, uprighting of lingually tipped teeth, and functional appliances [Denbar, 1998; Guilleminault and Quo, 2001; Villa et al., 2002; Cozza et al., 2002] have also met with success.

Lindman and Bondemark [2001] noted that case-studies reports in the literature raise questions on validity of studies as few prospective researches used appropriate control groups. In general these authors concluded that prospective OSA patients must be diagnosed, before beginning treatment with oral devices, with sufficient baseline data to establish the effectiveness of the treatment. This was the case in the present study. The question of an appropriate control group is a difficult one. It is not so easy these days to have a group of affected patients who are denied treatment so as to comprise an untreated control group. Unaffected children would not be a suitable control group. As scientific evidence of treatment outcomes and cost-effectiveness is now needed it is suggested that more well controlled prospective studies comparing the various approaches to the treatment of OSA are needed. Only by such research can an evidence based treatment protocol for OSA in children be developed.

Conclusions

The results of this study showed that the modified monobloc appliances may be an effective therapeutic alternative in children with mild to moderate OSA.

References


