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TREATMENT OF OBSTRUCTIVE SLEEP APNOEA

WITH ORAL APPLIANCES

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ABSTRACT

Treatment of Obstructive Sleep Apnoea with Oral Appliances

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The use of oral appliances for the treatment of obstructive sleep apnoea (OSA) has become an established treatment choice. To date, follow-up data on the effects of such treatment on the facial skeleton, pharynx and occlusion have been limited.

A study has subsequently been designed to address these issues. One hundred consecutively treated medically referred patients (87 males, 13 females, mean age 49 years, SD 8.5), were reviewed cephalometrically in six month intervals (6-30 months) following treatment with mandibular advancement therapy. Reference points and planes were digitized with a reflex metrograph and their means converted to linear and angular measurements. The mean mandibular advancement was 6.8mm (SD 1.8). No relationship was found between occlusal changes, degree of mandibular advancement, skeletal classification, duration of treatment, age or sex (ANOVA). When all patients were compared (N100) occlusal changes related to a reduction in overbite (-1.02mm $p<0.0001$) and overjet (1.06mm $p<0.0001$) this was associated with a retroclination of the upper anteriors (-1.9° $p<0.0001$) and a proclination of the lower anteriors (2.8° $p<0.001$). A reduction in maxillary arch length was also found (-0.47mm $p<0.006$). Skeletal differences included small statistically significant changes in SNA° ($p<0.023$), ANB° ($p<0.013$) and maxillary length ($p<0.002$). A change in the vertical position of the mandibular condyle was highly significant ($p<0.0001$). When the changes over time were determined, an increase in face height and reduction in overbite and overjet were evident at 6 months associated with a change in condylar position. Over-eruption of the maxillary

first premolars and mandibular first molars, along with a proclination of the lower incisors were only evident at 24 months. Occlusal changes tended to be progressive with on-going treatment with the greatest changes occurring at the final review period (30 months). Significant positive correlations were found also between the amount of anterior opening by the appliances and changes in overbite at 24 and 30 months.

Although changes were not evident in either hypopharyngeal width or hyoid bone position, long-term mandibular advancement does, however, have a demonstratable effect on both the oropharynx and velopharynx. Following 12 months of treatment, posterior airway space (PAS) increased from 10.7 to 12.0mm ($p<0.009$). Statistically significant changes in the velopharynx were observed as early as six months with a reduction in length of the soft palate of 1.5mm ($p<0.0001$). These changes were considered to be due to the loss of pharyngeal odema following the elimination of habitual snoring. A change in natural head position (NHP) from an extended to a more upright position was also significant .

Cephalometric differentiation between patients with mild or moderate OSA and those diagnosed with non-apneic snoring have been limited. Of this sample (N100), 58 patients were referred for the treatment of mild to moderate OSA and 42 for the treatment of non-apnoeic snoring. No statistically significant differences were observed between the apnoeic and non-apnoeic patients in either their skeletal or cranial base dimensions. Nasopharyngeal depth was, however, reduced in the apnoeic group.

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INTRODUCTION

The relationship between the fields of Dentistry and Sleep Medicine was in many respects established in 1995 following a review by the American Sleep Disorders Association (ASDA) which not only summarized the conclusive evidence of the efficacy of oral appliance therapy for the treatment of snoring and obstructive sleep apnoea (OSA) but also established practice parameters for their use. While the 1995 review found no reason for alarm, it concluded that follow-up data on the long-term effects of oral appliance therapy was indeed limited and recommended a systematic review of treated patients to determine the incidence and severity of occlusal changes and other related side effects. The research which forms the basis of this thesis is a commentary of the authors involvement in the field of Dentistry and Sleep Medicine over a fifteen year period (1988-2003) and follows the time-course of the development of oral appliances for the treatment of OSA and the evaluation of the long-term effects of these devices on the upper facial skeleton, pharynx and occlusion by way of a prospective longitudinal study carried out in a private orthodontic practice.

Obstructive sleep apnoea is a disorder in which recurrent closure of the upper airway occurs during sleep. Arousal from sleep is required to reopen the obstructed airway. These frequent arousals are the primary cause of excessive daytime sleepiness (Johns et al, 1998). The main nocturnal feature of OSA is snoring, reported in 97% of patients (Guilleminault et al 1978, Whyte et al 1989). This snoring tends to be loud and intermittent, often associated with breathing pauses and nocturnal choking. OSA has been defined as the cessation of airflow for greater than 10 seconds despite continuous respiratory effort, occurring at least five times per hour and associated with a drop in oxygen saturation levels of 4 per cent or more (Strollo and Rodgers 1996). Apnoea severity is classified as mild, moderate or severe, depending on the Apnoea-Hypopnoea index (AHI). The minimal diagnostic criteria for OSA has been defined as an AHI score

of 5 or greater with daytime hypersomnolence (Young et al 1993). Based on epidemiological evidence (Engleman et al 1997, Young et al 1997) the American Academy of Sleep Medicine Task Force (1999) defined mild OSA as an AHI of 5-15 events per hour, moderate OSA 15-30 events per hour and severe OSA greater than 30 events per hour. OSA is a recently recognised disorder and consequently only limited data is available on its long-term consequences. Untreated OSA is thought to be associated with an increased risk of cardiovascular morbidity including myocardial infarctions, ischaemic heart disease and cardiac arrhythmia (Ferguson and Fleetham1995). Patients with OSA also suffer from impaired cognitive function (Cheshire et al 1992) along with depression and a decrease in sexual functioning (Whyte et al 1989). The onset of hypertension has also been linked to the prior presence of sleep-disordered breathing (Peppard et al 2000). Another consequence concerned with increased morbidity is excessive daytime sleepiness (EDS) which is thought to be associated with increases in vehicle and work accidents (Findley 1996). Epidemiological studies have estimated that 2-4% of middle-aged men and 1-2% of middle-aged women are affected by OSA (Young 1993, Bearpark et al 1995).

Studies also demonstrated that obesity, classified as a Body Mass Index (BMI) greater than 30 kg/m^2 (Revicki and Israel 1986) was probably the most important risk factor associated with upper airway obstruction during sleep. Others have found greater neck circumferences in sleep apnoeics compared to weight and age-matched non-apnoeic snorers and normals (Hoffstein and Mateika 1992) and that neck circumference rather than general obesity demonstrated the best relationship with OSA severity (Davies and Stradling 1990). As men tend to deposit fat more centrally around the abdomen and the neck than woman, this appears to be one of the possible reasons why men are twice as likely to suffer from OSA (Young et al 1993). Upper airway resistance has also been

found to increase in middle-age men but not in women (White et al 1985) which is a probable cause why OSA tends to occur in middle-age.

Although obesity appears to be one of the most significant factors in relation to the severity of OSA, there is a subgroup of OSA patients who present with a low BMI that it has been hypothesized craniofacial abnormalities have put them "at risk" for upper airway closure (Mathur and Douglas 1995).

Abnormal craniofacial morphology demonstrated by cephalometric analysis has been extensively reported in patients with OSA (Riley et al 1983, Lowe et al 1986, Bacon et al 1990, Tangugsorn et al 1995, Ono et al 1996, Pracharktam et al 1996, Pae et al 1997, Özebek et al 1998). The features regarding bony elements include decreased sagittal dimensions of the cranial base, retrognathism of the maxilla and mandible, mandibular micrognathia, increased anterior lower facial height, reduced antero-posterior size of the bony pharynx, a low position of the hyoid bone and deviation of head posture. Aberrations in uvulo-glossopharyngeal morphology include thick and long soft palates, a reduced oropharyngeal width and large sized tongue. (Tangugsorn et al 1995). The position of the hyoid bone relative to the mandibular plane (H-MP) has been reported as one of the best cephalometric diagnostic predictors for the differentiation of apnoeic to non-apnoeic patients (Pracharktam et al 1996) as its position relative to the mandible appears to have a direct effect on respiration (Mayer and Meier-Ewart 1995).

Many consider that gross changes in tongue position can be assessed by analysing changes in hyoid bone position. Cephalometrically, hyoid bone position has been found to be more inferior in patients with OSA at a level of (C4-C6) in contrast to non-apnoeic controls at a level of (C3-C4). (Jamieson et al 1986, Partinen et al 1988, Mayer and Meier-Ewart 1995).

Developmentally, the hyoid bone is usually positioned between the third and fourth vertebrae and closely follows cervical vertebrae growth. Between 6-12 years of age there is no gender difference, however, between 12-18 years the position of the hyoid bone becomes lower in males than females. (Durzo and Brodie 1962). Between 22-42 years, the hyoid bone becomes even more inferior in males than females (Kollias and Krogstad 1999). These changes were also associated with an increase in soft-palate length and tongue size with a narrowing of the oropharynx with increasing age. (Johnston and Richardson 1999). According to Tourné (1991) this latent descent of the hyoid bone especially in males with increasing age, is a compensatory mechanism to maintain upper airway patency as a result of an increase in tongue bulk. In consideration of these findings, soft-tissue morphology and maturation changes are the most likely factors in the increasing prevalence of OSA with age. (Johnston and Richardson 1999).

Treatment of OSA can be classified into conservative, surgical and non-surgical. Conservative regimes include weight loss, avoidance of evening alcohol and antidepressants and avoiding sleeping in the supine position. Such therapies may help in a few cases, however, permanent loss of significant excess body weight by behavioural means alone is rarely successful. (Browman et al 1984).

Various drug treatments have been tried over the years as possible OSA therapies. Protriptyline, a tricyclic antidepressant has been found in some studies to reduce the frequency of apnoeas and hypopnoeas (Clarke et al 1979) but not in all studies (Whyte et al 1988). Patients have, however, reported troublesome side effects with the antidepressant (Whyte et al 1988). Modafinil is a new drug for the treatment or narcolepsy and appears to be a novel wake-promoting agent, which can be used to treat the daytime consequences of OSA. Arnulf et al (1997) found that modafinil improved objective sleepiness and long-

term memory with no adverse effects, drug therapies are, however, only regarded as adjunct therapy to other procedures.

The original surgical treatment for OSA was to perform a tracheostomy, with the aim of bypassing the obstructed airway. (Hill et al 1978). Tracheostomies have, however, been superseded by other surgical treatments being more acceptable to patients. In 1996, the American Sleep Disorders Association listed the following surgical procedures for the treatment of OSA: Nasal septal reconstruction, uvulopalatopharyngoplasty (UPPP), laser midline glossectomy, lingualplasty, inferior sagittal mandibular osteotomy and genioglossal advancement with hyoid myotomy and suspension (GAHM), maxillomandibular osteotomy and advancement, (MMA) and tracheostomy (Sher et al 1996).

The surgical procedure of UPPP was introduced in 1981 (Fujita et al) and in the early 1980's was a common procedure, however, this procedure has since fallen out of favour as the efficacy of this treatment for snoring and OSA has been reported as having only a 40% success rate (Sher et al 1996).

Maxillomandibular advancement is considered to be the most effective surgical treatment of OSA excluding tracheostomy. The technique includes a standard Le Fort I osteotomy in combination with bilateral sagittal split ramus osteotomies for the simultaneous advancement of the maxilla and mandible. (Prinsell 1999) Despite successful short term results of MMA in the management of OSA, little is known of the long-term clinical outcomes. Li et al (2000) demonstrated that oropharyngeal width improved after MMA but relapsed 34% at long-term follow-up. These authors concluded that greater advancement of the maxillomandibular complex tended to achieve better long-term clinical results.

The non-surgical treatment of OSA includes the use of nasal-continuous positive airway pressure (CPAP) (Sullivan et al 1981). This mechanical device delivers a stream of ambient air into a sealed nasal mask that the patient wears while sleeping. The positive pressure prevents upper airway occlusion by widening the lateral parts of the airway in particular and prevents the soft-palate and tongue from falling back against the posterior pharyngeal wall (Schwab et al 1996). CPAP therapy eliminates apnoeas, hypopnoeas, oxygen desaturation, improves sleep stage patterns and reduces daytime sleepiness (Issa and Sullivan 1986). CPAP is regarded as the first treatment alternative in patients with moderate to severe disease (McArdle et al 1999). Engleman et al (1999) found that CPAP also improved day-time sleepiness, cognitive function and mental depression in patients with mild apnoea. An improvement in long-term survival has also been reported (He et al 1988). The compliance of CPAP use after 3 years is said to be over 80% in patients with severe disease, but below 40% in patients with mild disease (McArdle et al 1999). Covert monitoring has shown, however, that the average usage of CPAP is less than 50% of the night (Kribbs et al 1993). Not all patients will tolerate this form of therapy as CPAP is an obtrusive device with patient compliance compromised due to mask discomfort, pump noise and nasal dryness.

The term "oral appliance therapy" was introduced to encompass all appliances placed in the mouth to modify upper airway anatomy and function during sleep for the relief of upper airway obstruction. The concept underlying oral appliance therapy is not new, as the relationship of the tongue and mandible to airway patency has been well known for years. The first use of an intraoral device was reported in 1934 by Pierre Robin, however, the first reports of the use of such devices to treat OSA were not until the 1980s (Cartwright and Samelson 1982, George 1987), The varieties of oral appliances can be categorised into four functional classifications; mandibular advancement devices (MADs),

tongue retaining devices (TRDs), soft palate lifters and tongue posture trainers (Rodgers 2000). Current practice suggests a discontinuance of soft palate lifters and tongue posture trainers with mandibular advancement devices being the most popular with clinicians. (Schmidt-Nowara 1999).

Cephalometric studies have demonstrated an increase in the retroglossal and retropalatal segments of the upper airway with mandibular advancement therapy. (Mayer and Meier-Ewart 1995, Lowe et al 1996). Endoscopic measurements have also demonstrated a change in the upper airway size and shape with mandibular advancement (Isono et al 1995). A direct effect on airway size may not be the only benefit of mandibular advancement, Lowe et al (1990) have shown that oral appliance therapy can produce increased EMG activity in genioglossus which is thought to translate to a greater resistance to collapse of the upper airway. In general it has been shown that oral appliance therapy surpasses that of CPAP for patient preference. In randomised cross-over controlled trials of oral appliances and CPAP, the majority of patients preferred oral appliance therapy over CPAP. (Ferguson et al 1996, Clark et al 1996). The 1995 Practice Parameters developed by the American Sleep Disorders Association stated that therapy with oral appliances is indicated as a first-line approach for simple snoring and mild OSA and as an alternative in more severe cases when CPAP was not tolerated and surgery not indicated.

Treatment of OSA patients with mandibular advancement therapy has been shown to improve sleep stage patterns by reducing the percentage of Stage 1 and Stage 2 sleep and increasing the percentage of Stages 3 and 4 and rapid-eye movement sleep (Petitjean et al 2000). The frequency of sleep arousals also decreases during treatment with mandibular advancement (Pancer et al 1999). A decrease in snoring between 73% and 100% has been reported in patients following treatment with mandibular advancement therapy (Marklin and Franklin 1996, Menn et al 1996, Ferguson et al 1997). Daytime sleepiness is also

reported to be reduced or eliminated with oral appliance use with associated improvement in cognitive functions. (Nambu et al 1997, Arai et al 1998.) A higher success rate with these devices has been reported in patients with mild to moderate apnoea compared with patients with more severe disease. (O'Sullivan et al 1995; Marklund et al 1998).

Recent studies have reported that long-term wear of mandibular advancement appliances were accompanied by small changes in the position of the mandible, reductions in both overbite and overjet, and a forward shift of the mandibular molars (Bondemark, 1999, Pantin et al 1999, Marklund et al 2001). Significant dental side effects in individual cases have also been reported (Panula and Keski-Nisula 2000, Rose et al 2001). The molar changes can be questioned because some of the observations were derived from study cases and, consequently, may be confounded by a downward and forward posturing of the mandible. While there may be some disagreement about the site(s) of the dental and occlusal changes due to mandibular advancement appliances there is less information about their onset.

A longitudinal, observational study was designed to determine in adults with sleep disorders the extent of the dental and occlusal changes following the use of a mandibular advancement splint (Robertson 1997) and secondly to determine the time-course of these changes.

One hundred adult subjects (87 males, 13 females) diagnosed with obstructive sleep apnoea (OSA) and/or asymptomatic snoring were treated with non-adjustable mandibular advancement appliance. At the outset each subject was randomly assigned to a group and reviewed 6, 12, 18, 24 or 30 months after placement of the device. There were 20 subjects in each group. Craniofacial changes were measured on lateral cephalometric radiographs taken at the initial and review appointments. When the changes in all subjects were examined the SNA, ANB angles, ANS – PNS length and face height increased and the

mandibular first molars and the maxillary first premolars had over-erupted. Significant retroclination of the maxillary incisors and proclination of the mandibular incisors were accompanied by reductions in maxillary arch length, overbite and overjet.

When the changes over time were determined the mandibular symphysis was significantly lower at all review periods. An increase in face height, and reductions in overbite and overjet were evident at 6 months, and over-eruption of the maxillary first premolars and mandibular first molars, and proclination of the lower incisors were found at 24 months. Significant positive correlations were also found between the amount of anterior opening by the appliances and changes in the overbite at 24 and 30 months.

The appliance used produced small, unpredictable changes in the occlusion that tended to occur after 24 months wear. It is postulated that the changes in overbite might be lessened by keeping the bite-opening to a minimum. (Robertson 2001, Robertson et al 2003.)

The effect of long-term mandibular advancement therapy on the pharynx and hyoid bone was also investigated. Following 12 months of treatment, the posterior airway space (PAS) a measure of oropharyngeal width increased from 10.7 to 12.0mm ($p<0.009$). Statistically significant changes in the soft palate were observed as early as six months with a reduction in length of 1.5mm ($p<0.0001$) and a reduction in thickness of 0.6mm. These changes were considered to be due to the loss of pharyngeal odema following the elimination of habitual snoring. No changes were observed, however, in the width of the hypopharynx or hyoid bone position following long-term mandibular advancement (Robertson 2000).

The relationship between natural head position (NHP) and OSA is well documented. Patients with OSA have been found to exhibit an extended and forward NHP when compared to non-apnoeic controls (Özbek et al 1998). In this study (Robertson 2002)

apnoeics had a greater cranial extension (NSL-vert) 101° compared to asymptomatic snorers (NSL-vert 97°). Following treatment with mandibular advancement a change in NHP from 99.7° to 93.0° was statistically significant ($p<0.001$). Although not confirmed by polysomnography, this change in head position from an extended to a more upright position may be indicative of improved upper airway function (Robertson 2002).

Tongue Retaining Devices (TRDs) have been objectively studied since the mid 1980s and have been shown to be effective in many cases. (Cartwright and Samelson 1982, Cartwright 1985). TRDs function by directly engaging the tongue and holding it in a forward position to open the upper airway during sleep. The major advantage of the TRD may be its ability to promote forward tongue position without having to engage the dentition which is a significant advantage in patients with compromised dentitions, or who are edentulous.

The Tongue Stabiliting Device (TSD) was developed by C.J. Robertson as an "off the shelf" product for the treatment of habitual snoring and mild to moderate OSA. Previously, fabrication of a TRD required the taking of dental impressions and insertion by a dental professional. Extension into the oral cavity by these devices often created difficulty with saliva control and restricted oral breathing.

A small pilot study was carried out to determine the efficacy of the TSD in terms of polysomnographic variables (Kingshott et al 2002). Six current users of the TSD were involved in this study. Each subject underwent two consecutive nights of polysomnography, one night with the TSD and one night without the device in place, in a randomised crossover design. The TSD significantly lowered the arousal frequency, the % stage 1 sleep and the frequency of snores/h slept. Trends were also found for reductions in the apnoea/hypopnoea frequency and oxygen desaturations. The results of this pilot study demonstrated that the TSD may be effective in lowering snoring severity and

microarousals, with trends for reducing the severity of sleep-disordered breathing. The TSD may therefore be an effective therapy in selected individuals with sleep-disordered breathing.

In conclusion, oral appliance therapy is now regarded as a first-line treatment for patients presenting with sleep-disordered breathing. To date, only limited data has been available on the long-term consequences of such treatment. The ensuing series of papers follows the development of a mandibular advancement appliance for the treatment of OSA and investigates the long-term effects of this device on the upper facial skeleton, pharynx and occlusion by way of a prospective longitudinal study carried out in a private orthodontic practice. The development and trialing of an alternative form of treatment (TSD) is also presented.

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Obstructive sleep apnoea. Part I: diagnosis, aetiology, and current treatment

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The term "apnoea" is derived from the Greek word "apnoia" meaning "want of breath". Apnoea has occurred if breathing stops for at least 10 seconds as detected by airflow at the nostrils and mouth. There are three forms of apnoea – central, obstructive, and mixed[1]. Central apnoea is cessation of airflow associated with complete cessation of all respiratory movements. The pathogenesis of central sleep apnoea is complex and usually involves a neurological disease, but the condition can occur without identifiable cause; fortunately its occurrence is rare[2]. Obstructive apnoea is associated with obstruction of the upper airway and is characterised by loss of airflow while respiratory movements remain normal; it is usually accompanied by loud snoring. This form is the most common of all apnoeas[3]. The clinical features of this disorder were clearly described by Charles Dickens in *The Pickwick Papers*, in the portrayal of Joe who had persistent hyper-somnolence[4]. Mixed apnoea typically has an initial central apnoea component followed by an obstructive component[1].

OBSTRUCTIVE SLEEP APNOEA

Obstructive sleep apnoea can be summarised as the cessation of airflow for greater than 10 seconds despite continuous respiratory effort, occurring at least five times per hour and associated with a drop in oxygen saturation levels of 4 percent or more.

Obstructive sleep hypopnoea has occurred if there is a decrease of 30-50 percent in airflow for 10 seconds or more, 15 or more episodes per hour of sleep, associated with a decrease of 4 percent or more of oxygen saturation levels[5]. Patients with "upper airway resistance" have no significant decrease in airflow, yet have 15 or more episodes of arousal per hour of sleep, but without significant decrease in oxygen saturation levels[5].

The prevalence of obstructive sleep apnoea varies according to the population surveyed[6]. Current figures range from 1 to 9 percent of the adult population, and a male to female ratio of 15:1 pre-menopausal[7,8]. Habitual snoring is the most common symptom of obstructive sleep apnoea[9]. Heavy snoring produces pharyngeal oedema which may in turn narrow the airway[10]. It can affect all age groups, especially children in association with large tonsils and adenoids[11]. Management of the latter often involves only tonsillectomy and adenoidectomy[12].

Snoring, a noise produced by vibration of the soft palate and the anterior and posterior pillars of fauces, is usually associated with obstruction of the upper airway. If the degree of obstruction is slight, the snoring will be regular and respiration is not compromised[1]. The type of snoring associated with obstructive sleep apnoea appears to be more of a pharyngeal nature[13]. A loud noise usually marks the onset of respiration following a silent period. This noise is associated with a clearance of the obstruction by respiratory drive; it can occur hundreds of times per night, causing frequent arousals[14].

DIAGNOSIS

Obstructive sleep apnoea is the most common type of respiratory disturbance documented by all-night polysomography[3]. A polysomographic recording includes electroencephalogram (EEG), electromyogram (EMG), electro-oculogram (EOG), electrocardiogram (ECG), thoracic respiratory movements, oro-nasal airflow, and oxygen saturation using pulse oximetry[1].

The clinical signs and symptoms of obstructive sleep apnoea are hyper-somnolence or excessive daytime sleepiness, headaches, nocturnal dysrhythmias, polycythaemia pulmonary hypertension, and right-sided heart failure, the latter condition a result of severe states of nocturnal hypoxaemia and pulmonary hypertension. Patients complain of lack of energy, depression, and personality changes. Testosterone levels are also diminished[15-18].

Hyper-somnolence

Hyper-somnolence is caused by sleep fragmentation[19]. The normal sleep cycle consists of four stages of non-rapid eye movement (non-REM) sleep and a final stage of rapid eye movement (REM) sleep. In a normal 8-hour sleep period, REM sleep occupies approximately $1^1/_2$ to 2 hours for a rested sleep. In patients with obstructive sleep apnoea, the most prolonged episodes of apnoea and significant oxygen desaturations tend to occur in Stage 4 non-REM and REM sleep[13].

Patients with obstructive sleep apnoea tend to sleep for a shorter time, have an increased Stage 1 non-REM sleep, but decreased Stage 3 and 4 non-REM sleep. Morning headaches and nausea may be the result of nocturnal CO_2 retention[20].

Patency of the upper airway

The patency of the upper airway is a result of many inter-related anatomic and physiological factors[21]. During inspiration, a negative intra-pharyngeal pressure develops, but collapse of the airway is prevented by the action of the pharyngeal abductor and dilator muscles. These muscles are activated rhythmically during daytime respiration, but become hypotonic during sleep, and airway stability becomes dependent on pharyngeal size and compliance of the pharyngeal tissue[22].

AETIOLOGY OF OBSTRUCTIVE SLEEP APNOEA

The aetiology of obstructive sleep apnoea is complex[23]. The site(s) of upper airway obstruction may vary. This variance makes treatment selection and prognosis difficult. In obese individuals, the loss of airway patency is often attributed to the accumulation of peri-oropharyngeal fat.

Obesity may also contribute to a lack of control of the oral and pharyngeal musculature[24].

A simple mechanism may not apply to all patients. Electromyographic studies have revealed that genioglossus has a phasic burst during inspiration movements, producing a slight advancement of the tongue, maintaining airway patency during inspiration[26]. In individuals with obstructive sleep apnoea, genioglossus function may be impaired, allowing the prolapse of the tongue against the posterior pharyngeal wall with inspiratory effort during sleep[26]. Evidence also suggests that a general hypotonia of the pharyngeal dilating muscles of the upper airway can also be involved in allowing an obstruction of the upper airway[27].

Computed tomographic studies by Lowe *et al.*[28], have shown that the upper airway is narrower in patients with obstructive sleep apnoea. They also observed that obese patients had larger tongues and soft palates, and suggested a link between obesity and the abnormal upper airway observed in patients with obstructive sleep apnoea. Fat deposition may therefore be contributing to an enlargement of the soft palate and tongue in obese patients with obstructive sleep apnoea. Lowe *et al.* concluded that weight reduction may decrease the severity of apnoea in part by reducing the size of the tongue and soft palate. Sleep apnoea has, however, been identified as common to some families, and is not explained by obesity alone[29].

Cephalometric parameters

Lateral cephalometry shows that patients with obstructive sleep apnoea can have alterations in craniofacial structure. Riley *et al.*[30] found that the length of the soft palate, the position of the hyoid bone, and the posterior airway space were significant cephalometric parameters in patients with the condition. Soft palate length was measured from the posterior nasal spine (PNS) to the top of the soft palate contour (PNS-P). The mean distance was found to be 37 ± 3 mm.

Hyoid bone position was defined as the vertical distance of the hyoid bone (H), from the mandibular plane (MP), a plane constructed from gnathion (Gn) through gonion (Go) to the hyoid bone (MP-H). The mean distance was 15 ± 3 mm. Posterior airway space (PAS) was the width of the airway measured by a line constructed from B-point (the point of greatest concavity on the anterior surface of the mandibular symphysis) through Go to the posterior pharyngeal wall. The mean distance was 11 ± 1 mm. Partinen *et al.*[20] found that, when the MP-H distance was greater than 24 mm and the PAS width equal to or less than 5 mm, the respiratory disturbance index (RDI – defined as the total number of apnoeas and hypopnoeas per hour of sleep), was clearly influenced. Partinen *et al.* concluded that MP-H distance and PAS width were risk factors for an elevated RDI.

The hyoid bone has also been found to be more inferiorly positioned in patients with obstructive sleep apnoea, being at the level of cervical vertebrae C4-C6, compared with being at C3-C4 in controls[31]. Lowe *et al.*[22] found that patients with obstructive sleep apnoea often have posteriorly positioned maxillae and mandibles, steep mandibular planes, high upper and lower facial heights, and anterior open-bites, all associated usually with a long tongue. It can therefore be concluded that both anatomical and physiological variables may adversely affect airway size[32].

TREATMENT

Current treatment for patients with obstructive sleep apnoea can be categorised into behavioural, medical, surgical, and dental. The goals of treatment are to establish normal nocturnal oxygen saturation levels and to eliminate snoring and disruption of sleep due to upper airway resistance.

Behavioural treatment

Behavioural treatment involves the abstinence from alcohol and sedatives in the early evening[33]. The intake of alcohol selectively reduces the muscle tone of the upper airway and increases the frequency of abnormal breathing during sleep[34]. Alcohol also prolongs apnoea by delaying arousal[35]. Weight reduction in patients without anatomical risk factors can often eliminate obstructive sleep apnoea[36]. Behavioural methods also include training patients to sleep in a lateral position if upper airway obstruction is present only during sleep in the supine position.

Medical treatment

Medical treatment involves the use of continuous positive airway pressure (CPAP). This treatment is regarded by most as the first line of treatment in patients with moderate to severe obstructive sleep apnoea. This generally means patients with more than 20 episodes of apnoea or hypopnea per hour with associated oxygen desaturations[37]. However, patient compliance with CPAP is approximately 50 percent[37]. Many patients therefore go untreated. Side effects reported by patients include irritation related to the nasal mask, nasal congestion, occasional rhinorrhea, and feelings of claustrophobia.

Surgical treatment

Surgical methods to eliminate obstructive sleep apnoea include reduction of the inferior turbinate bones, adenoidectomy (nasopharyngeal involvement), uvulopalatopharyngoplasty (UPPP), tonsillectomy (oropharyngeal involvement), genioglossal and hyoid advancement (hypopharyngeal involvement), bimaxillary advancement, and tracheostomy. UPPP is curative in less than 50 percent of patients[38,39]. Laser-assisted UPPP has recently been introduced as an outpatient treatment for snoring. UPPP enlarges the oropharynx and reduces the collapsibility of the upper airway. Patients with a narrow oropharynx relative to tongue size have a good response to UPPP[40].

The use of fibre-optic nasopharyngoscopy as a preliminary procedure to establish the site(s) of obstruction prior to surgery is gathering wider acceptance. The effectiveness of this procedure, however, compared with pharyngeal pressure measurements supplemented with oxygen saturation and oro-nasal airflow recordings, is currently being debated[41].

Surgical treatment of obstructive sleep apnoea by maxillo-mandibular advancement should be restricted to patients with a retrognathic dolichofacial type combined with pharyngeal narrowing. The maxilla and mandible must both be advanced at least 10 mm to ensure success[42]. In severe instances of obstructive sleep apnoea that are life-threatening, the ultimate treatment is tracheostomy[43]. This procedure completely by-passes the upper airway and thus all upper airway obstruction. The procedure is, however, usually used as the last option in the treatment of obstructive sleep apnoea.

Dental appliances

Dental appliances in the treatment of obstructive sleep apnoea are of three classes. One type attempts to lift the soft palate with a distal extension from a palatal plate. This has not found wide acceptance due to gagging and the

uncertainty of maintaining hypopharyngeal width during sleep[44]. The second class of appliances is designed to act directly on the tongue by holding it forward by means of negative pressure from an anterior suction bulb or proprioceptive reminder. The tongue retaining device (TRD)[45] is the most successful of these appliances, especially in the elimination of snoring. The third group of appliances re-positions the mandible in a more protrusive position and has general acceptance as being the most effective design in the elimination of both snoring and obstructive sleep apnoea[44].

The mechanism by which these appliances work appears simple. Mandibular-advancement splints prevent the tongue collapsing against the posterior pharyngeal wall nocturnally. This is achieved by mechanical means in that the origin and insertion of genioglossus is at the hyoid bone and mandibular symphyseal region respectively. Thus, by advancing the mandible, the tongue is held in a more anterior position nocturnally. Elevation of the hyoid bone in an antero-superior direction is therefore the desired radiographic modification. A second consideration given by Lowe *et al.*[46] is that, in man, voluntary passive opening of the mandible produces definite enhancement of genioglossus EMG through activation of receptors located in the temporomandibular joint. Because the contraction of the genioglossus opens the airway, airway obstruction may be prevented.

An overview of the 13 current appliances with the United States Food and Drug Administration approval is given by Lowe[44]. Despite considerable variation in the design of these appliances, the desired effects are remarkably consistent. Snoring is reduced and often eliminated in almost all patients who use oral appliances[47]. Obstructive sleep apnoea improves in the majority of patients. Limited follow-up data indicate that oral discomfort is a common but tolerable side effect and that dental and mandibular complications appear to be uncommon[47].

Theoretical complications of long-term use of mandibular advancement splints include temporomandibular joint dysfunction. However, intermittent forward positions of the mandible have yet to be shown to produce irreversible TMJ dysfunction as a consequence[48].

Practice parameters for oral appliances: Practice parameters for the treatment of snoring and obstructive sleep apnoea with oral appliances, based on a review of the relevant scientific literature, have been formulated by the Standards of the Practice Committee of the American Sleep Disorder Association. In summary, the presence or absence of obstructive sleep apnoea must be determined before initiating treatment with oral appliances to identify those patients at risk due to complications of sleep apnoea and to provide a baseline to establish the effectiveness of subsequent treatment[49].

In brief, there is no doubt that dental appliances are simple, reversible, quiet, and cost effective, and may be indicated in patients who are unable to tolerate continuous positive airway pressure (CPAP) or who are poor surgical risks.

The design and use of a new type of appliance is described in Part II of this article.

SUMMARY

Obstructive sleep apnoea is a multi-factorial condition associated with high morbidity and mortality. Its prevalence is highest in middle-aged males with a predisposition to obesity. Certain facial types have been identified as being at risk. Cephalometric parameters have now been established to identify those patients who are anatomically compromised. Treatment modalities are dependent on the correct diagnosis of the site(s) of obstruction. Overnight polysomographic testing is the only definitive measure to quantify the presence and severity of obstructive sleep apnoea. Treatment options include behavioural, medical, surgical, and the use of oral appliances.

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articles

Obstructive sleep apnoea. Part II: treatment with a customised dental appliance

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SUMMARY

The use of dental appliances in the treatment of patients with snoring and obstructive sleep apnoea is an important treatment modality for those patients not severe enough for continuous positive airway pressure (CPAP) or who cannot tolerate this form of treatment. A mandibular advancement splint has been specifically designed to eliminate snoring and obstructive sleep apnoea. The appliance's design parameters included ease of insertion, comfort, and maximum effectiveness. Customised appliances have been designed for dentate, semi-dentate, and edentulous patients.

To date, over 100 appliances have been used with a symptomatic improvement in snoring and well-being in over 80 percent of patients.

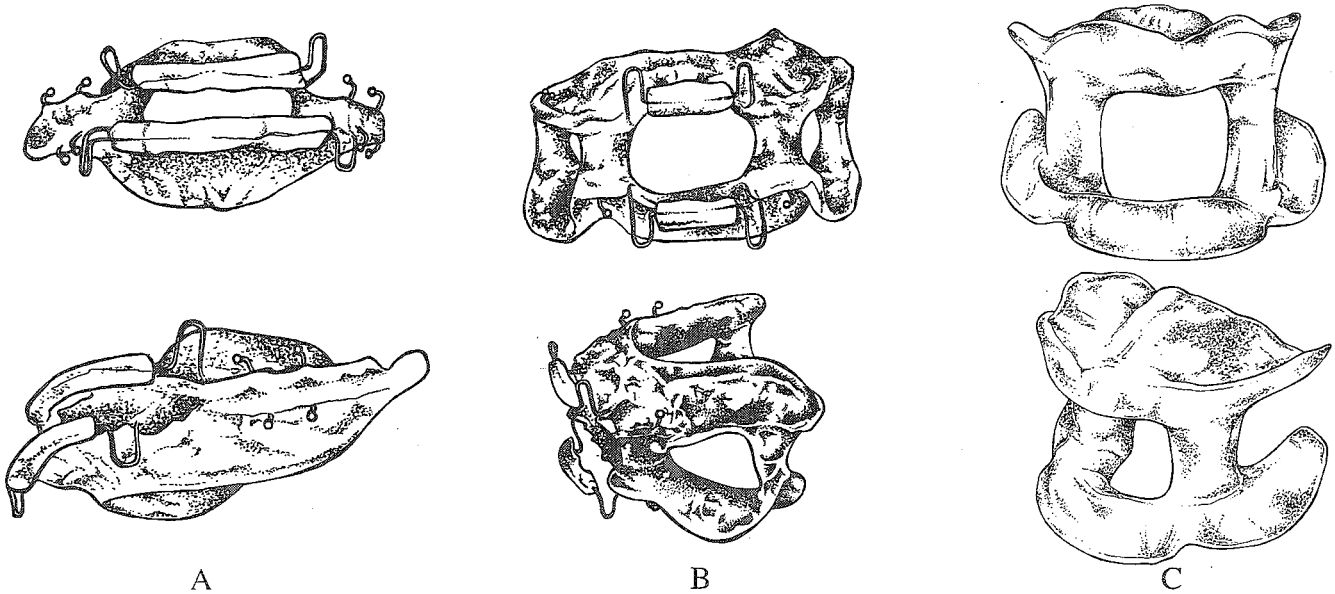
Dental appliances for the treatment of snoring and obstructive sleep apnoea are simple, cost effective, and reversible.

INTRODUCTION

In Part I of this paper[1] the diagnosis and aetiology of obstructive sleep apnoea were described, and current methods of treatment, including the use of dental appliances, were reviewed. The first reported use of an appliance to treat airway obstruction was in 1934, when Pierre Robin[2] described a mono-block functional appliance to move the mandible forward to treat "glossoptosis" (tongue obstruction). In 1984 Meier-Ewert, Schafer, and Klob[3] described a mandibular repositioning appliance with 3-5 mm of mandibular advancement to reduce obstructive sleep apnoea. In 1985, George[4] designed the nocturnal airway patency appliance (NAPA) from a modified functional appliance. Other modifications of existing functional appliances followed, namely the Equaliser, Herbst, Jasper, and mandibular repositioner[5]. Nearly all the dental appliances reviewed by Lowe[5] are modifications of existing orthodontic appliances. This paper describes the design and use of an appliance intended specifically to eliminate snoring and obstructive sleep apnoea, and summarises its use in 100 patients. A survey of these patients by Lamont[6], using the Epworth Sleepiness Scale, concluded that 87 percent of those patients surveyed had either stopped, or had a marked reduction in snoring. Nearly all patients found better quality of sleep, and day-time hyper-somnolence was subsequently less. Although these data should be interpreted somewhat cautiously as polysomographic testing was not performed before and after insertion of the appliances, these findings are consistent with the findings of other studies with polysomographic testing[7].

DESIGN OF THE APPLIANCE

The design of the appliance differs from other appliances in several aspects. Firstly, the appliance is fabricated in two components encompassing the upper and lower anterior



A B C

FIG. 1 – Anterior and lateral views of A, mandibular advancement splint designed for a dentate patient; B, mandibular advancement splint designed for a semi-dentate patient; and C, mandibular advancement splint designed for an edentulous patient.

teeth; the components are "spring-loaded" to allow for ease of insertion, yet are very retentive. Secondly, the design allows for modification to accommodate dentate, semi-dentate, and edentulous patients. Ease of adjustment, especially in enabling further mandibular advancement, is a critical factor. As the appliance is made of acrylic and has stainless-steel ball-hooks for posterior retention, it is durable, and repairs are minimal.

A posterior bite-plane with full occlusal coverage of both the upper and the lower arches prevents unwanted tooth movement. The thickness of this component is dependant on the degree of vertical opening required.

Figure 1A illustrates the anterior and lateral aspects of a mandibular advancement splint for dentate patients; Figure 1B illustrates a design for a semi-edentulous patient; and Figure IC illustrates a design for an edentulous patient. In the semi-dentate patient, the upper portion of the appliance can be substituted for a denture base, with extending wings to the lower aspect of the appliance.

MANDIBULAR ADVANCEMENT

The use of mandibular advancement splints to treat obstructive sleep apnoea is a prime consideration when the site of obstruction is at the hypo-pharyngeal level. This can usually be verified by fibre-optic nasopharyngoscopy in conjunction with lateral cephalometry.

The amount of mandibular advancement needed varies according to the individual. To avoid temporomandibular joint discomfort, the bite registration is not recorded at the maximum protrusive position, but at a position which could be termed the "most relaxed protrusive position" – usually 75 percent of the maximum protrusive position. Patients tolerate the protrusive position well if it is accompanied by vertical opening, the degree of which often relates to the amount of overbite that is present. In dentate patients, the vertical separation allows for airspace between the teeth.

FACIAL PATTERN CONSIDERATIONS

In patients with a Skeletal II facial pattern, mandibular advancement can be considerable. The female patient shown in Figure 2 had a Skeletal II facial pattern, and easily accommodated a mandibular advancement of approximately 1 cm. Wear facets on anterior teeth indicated that this position was not beyond her normal protrusive range. Radiographically, elevation of the hyoid bone in an antero-superior direction was evident, and this was accompanied by a 50 percent increase in hypopharyngeal width. Snoring was eliminated.

In patients with a Skeletal I facial pattern, the degree of mandibular advancement is less, usually 4-5 mm, but this does not appear to affect the efficiency of the appliance. Figure 3 shows the radiographs of a 55-year-old insulin-dependent

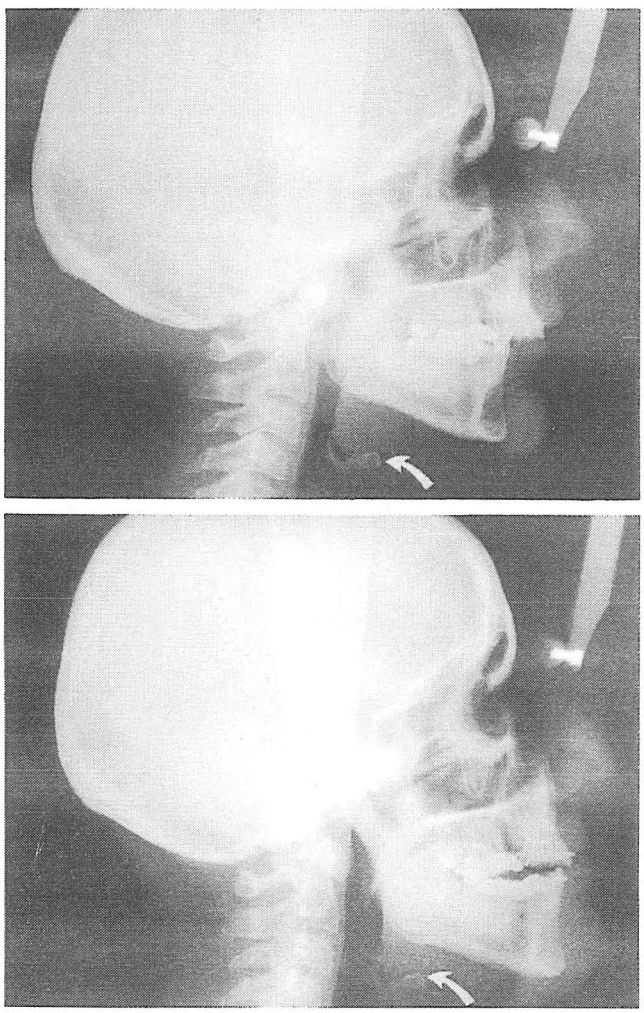


FIG. 2. – Lateral cephalograms of a patient with a Skeletal II facial pattern. Upper, before, and lower, after insertion of the appliance. With the appliance in place, the hypopharyngeal width is increased and the hyoid bone (arrowed) is elevated.

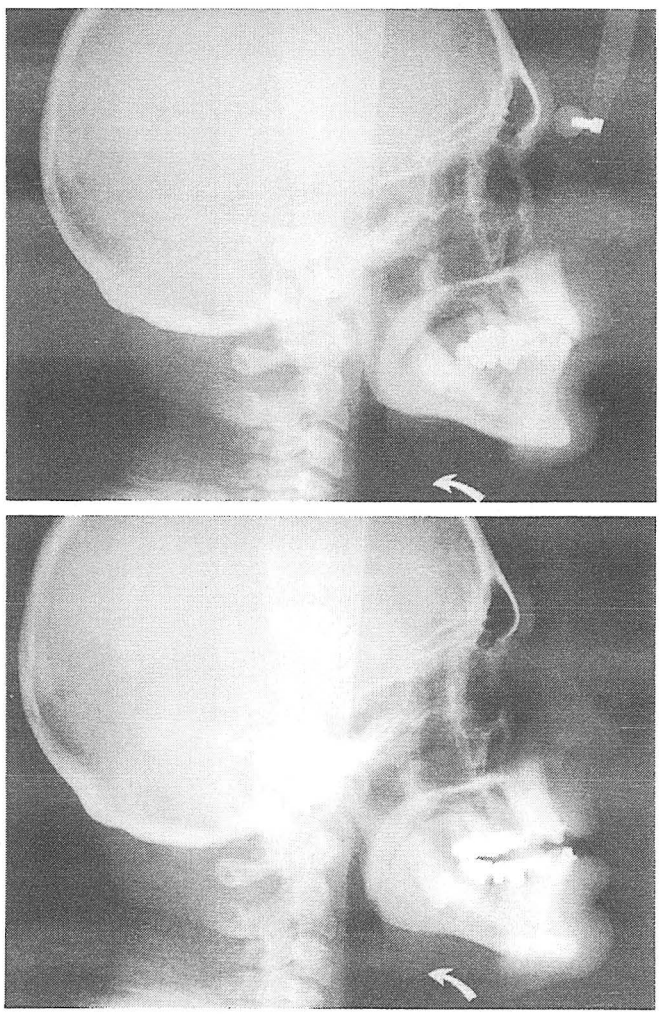


FIG. 3. – Lateral cephalograms of a patient with a Skeletal I facial pattern. Upper, before, and lower, after insertion of the appliance. With the appliance in place, the hypopharyngeal width is increased and the hyoid bone (arrowed) is elevated.

diabetic man with severe ischaemic heart disease who was referred by an ear, nose, and throat surgeon for snoring and problems with nocturnal angina. Following insertion of the mandibular advancement appliance the patient experienced loss of daytime hyper-somnolence, and nocturnal angina ceased. An overnight sleep study showed that, with the appliance inserted, the baseline oxygen saturation levels did not drop below 90 percent, no obstructive sleep apnoea occurred, and sleep was restful. Nocturnal angina re-occurred when the appliance was not used.

LIMITATIONS OF FACIAL PATTERN

In some facial types, the degree of mandibular advancement is limited. For example, patients with a Class II division II malocclusion are anatomically limited in the amount of mandibular advancement that can be achieved; this may influence the effectiveness of the appliance. The 45-year-old male shown in Figure 4 had a Class II division II malocclusion; he snored, had daytime hyper-somnolence, and morning headaches. The mandibular advancement splint resulted in an increase in the hypopharyngeal width and elevation of the hyoid bone.

In such patients the amount of vertical opening to achieve an anterior protrusion of the mandible is far greater than for other kinds of facial types, and this often has a detrimental affect on the elevation of the hyoid bone. In this patient, however, the hyoid bone was still elevated and an increase in hypopharyngeal width occurred. An overnight sleep study showed that the oxygen saturation levels did not fall below 90 percent for any significant period. Daytime symptoms were relieved.

TREATMENT OF EDENTULOUS PATIENTS

The limiting factor in the treatment of edentulous patients with obstructive sleep apnoea is the lack of retention in the mandible. The use of implants for retention would be the treatment of choice, but often the cost and the delay in the insertion of the appliance excludes this form of treatment.

The appliance design is different from that used in either dentate or semi-dentate patients. Firstly, denture bite blocks are fabricated and the protrusive position is taken – this is less than that for dentate patients. In order to maintain stability, a vertical component greater than the free-way space is necessary. Retention of the appliance is aided by

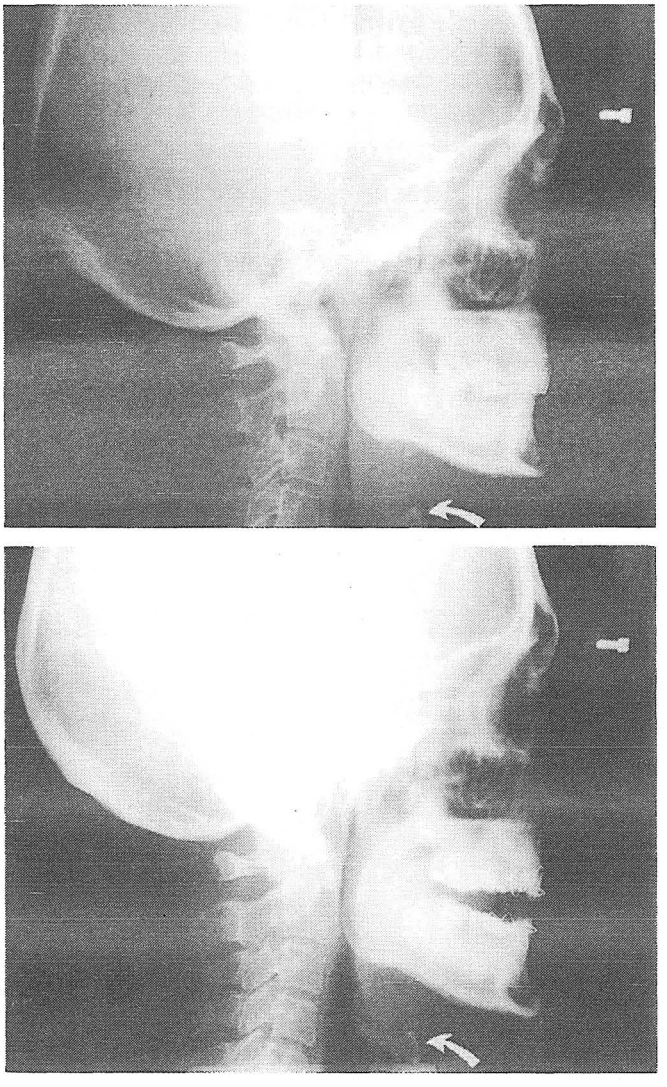


FIG. 4. – Lateral cephalograms of a patient with a Class II division II malocclusion. Upper, before appliance insertion; the hypopharyngeal width is minimal and the distance from the mandibular plane to the hyoid bone is long. Following the insertion of the appliance, lower, the hypopharyngeal width is increased and the hyoid bone (arrowed) is elevated.

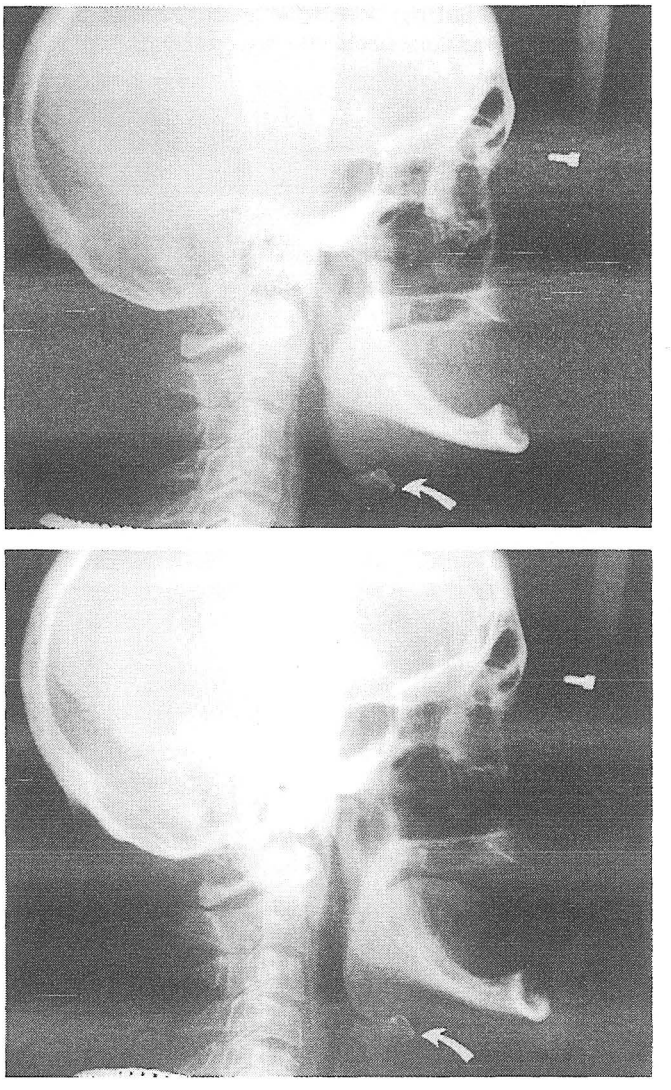


FIG. 5. – Lateral cephalograms of an edentulous patient. Upper, before, and lower, after insertion of the appliance. Elevation of the hyoid bone (arrowed) has occurred.

extensions onto the ramus and by fully utilising the extent of the labial sulcus.

This approach has been successful even in patients with little lower alveolar ridge. For example, a mandibular advancement splint was constructed for the 59-year-old man shown in Figure 5; he had severe obstructive sleep apnoea and ischaemic heart disease. An overnight sleep study revealed that, with the appliance inserted, the baseline oxygen saturation levels did not drop below 90 percent. When the splint was removed, however, his pattern of respiration was associated with episodes of hypopnoea and reduction in oxygen saturation. Radiographically, elevation of the hyoid bone in an antero-superior direction was achieved, and increase in the hypopharyngeal width also occurred.

TREATMENT OF CHILDREN

The mandibular advancement splint is not restricted to adult patients. The 11-year-old male whose lateral cephalograms are shown in Figure 6, was referred by an ear, nose, and throat surgeon for severe snoring, present since age 3 months. Nasopharyngoscopy showed that he had an obstruction at the hypopharyngeal level. A mandibular advancement splint was inserted. Radiographically there was a significant increase in hypopharyngeal width, and elevation of the hyoid bone. The appliance was used for 4 months and was then discontinued as snoring was not occurring without the appliance. This has been substantiated by subsequent reviews.

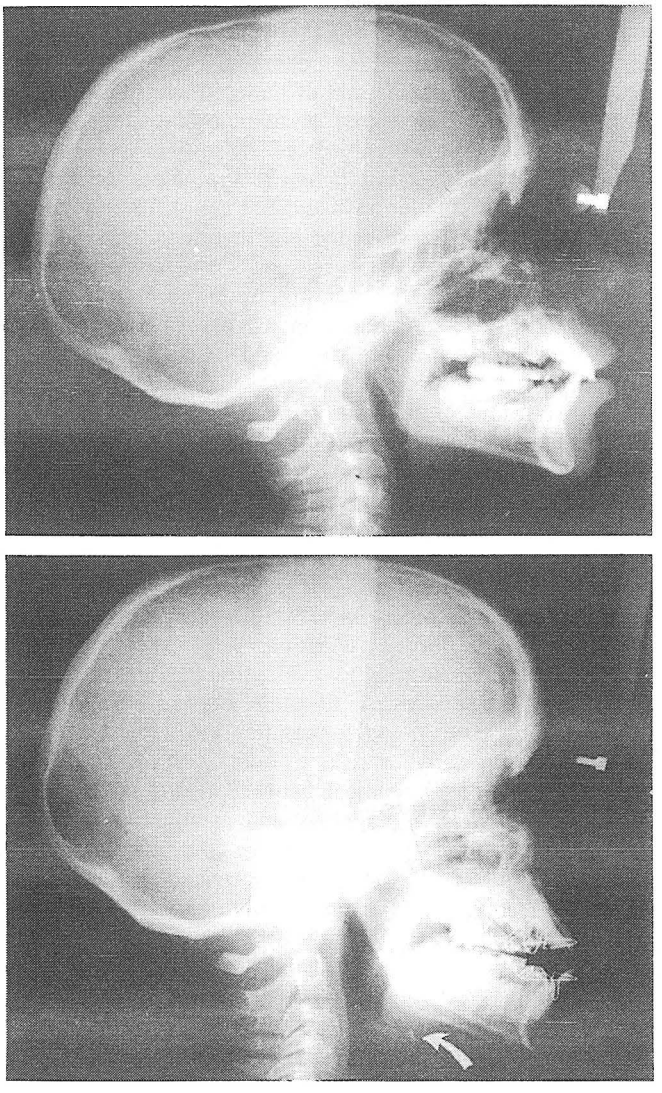


FIG. 6. – Lateral cephalograms of an 11-year-old male. Upper, before, and lower, after insertion of the appliance. A marked increase in hypopharyngeal width has occurred; the hyoid bone is visible in the lower cephalogram only (arrowed).

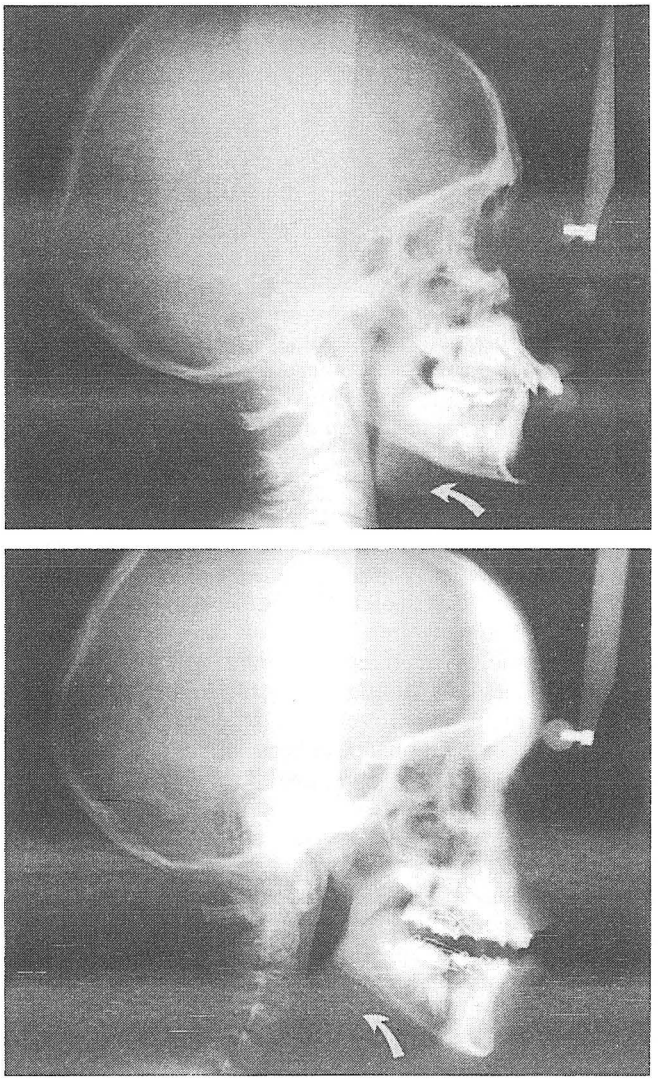


FIG. 7. – Lateral cephalograms of a 10-year-old male with a Skeletal II facial pattern with severe occlusion of the hypopharynx. Above, before, and below, following insertion of the appliance appliance. An increase in hypopharyngeal width is evident; the hyoid bone is arrowed.

The 10-year-old male shown in Figure 7 was referred by an ear, nose, and throat surgeon following nasopharyngoscopy. When the boy's mouth was wide open, the epiglottis and the base of the tongue were completely collapsed against the posterior pharyngeal wall. A lateral head film indicated severe constriction at the hypopharyngeal level. Symptomatic effects of this condition included lapses of concentration, headaches, and difficulty with learning. Night time gasping was also evident.

A mandibular advancement splint was inserted. The lateral cephalogram with the appliance *in situ* indicated an increase in hypopharyngeal width. Although not dramatic, this was adequate to eliminate snoring and subsequent sleep apnoea. Daytime hyper-somnolence disappeared, and concentration at school improved.

DISCUSSION

Nearly all dental appliances that advance the mandible to eliminate snoring and obstructive sleep apnoea have been

modifications of existing functional appliances[5]. However, the appliance described here has been designed specifically for the elimination of snoring and obstructive sleep apnoea. The parameters of appliance design included patient comfort and compliance, ease of insertion and removal, and ease of modification. In the design of the appliance, consideration was given to the often eccentric mandibular movement into the protrusive position. The use of a spring-loaded anterior component enables ease of insertion and removal, yet the appliance is still very retentive.

A review of the literature reveals a lack of information on why mandibular advancement splints are successful in some instances and not in others. Failure in some patients may be related to non-achievement of the degree of mandibular advancement required to eliminate obstructive sleep apnoea. The degree of mandibular advancement is, however, often limited by the facial pattern of some patients. For example, patients with a Class II division II malocclusion can tolerate only minimal protrusive movement; conversely, patients with a Class II division I malocclusion can tolerate significant mandibular protrusion. Patients often exhibit a "threshold protrusive distance" which is required to eliminate obstructive sleep apnoea. To achieve additional protrusion in patients with limited protrusive movement, following a month's nocturnal wear the appliance is horizontally sectioned; both halves are keyed and a thin wax-bite registration is taken with the halves *in situ* in a more protrusive position. The appliance is then mounted in an articulator and the two halves are joined by self-curing acrylic in this more anterior position. Often only minimal advancement of 1-2 mm is sufficient to achieve the "threshold protrusive position".

The use of lateral cephalometry is an important adjunct in the treatment of patients with obstructive sleep apnoea with a mandibular advancement splint. The desired radiographic modification is an antero-superior elevation of the hyoid bone following insertion of the appliance. However, several patients had very little elevation of the hyoid bone after their appliances were inserted, but snoring and obstructive sleep apnoea were eliminated. An explanation for success in these patients was the maintenance of existing hypopharyngeal width in preventing the mandible from rotating down and back during sleep, with subsequent hypopharyngeal occlusion. If this is indeed the case, mandibular protrusion may not be the only critical objective – rather it is the prevention of mandibular downward and backward rotation during sleep.

Although the mandibular advancement splint was designed to increase hypopharyngeal width, a constant clinical observation in almost all patients was a noticeable reduction of nasal resistance following insertion of the appliance. Respiration was notably slower and deeper. It can be assumed, therefore, that the appliance also has an effect on the nasopharynx, possibly by reducing resistance in the upper airway. Radiographically, the uvula was often found to be displaced antero-superiorly from the posterior pharyngeal wall and tongue. If the uvula was the cause of snoring, this repositioning would, no doubt, lead to the elimination of snoring in many patients.

A review of the literature reveals little evidence of long-term temporomandibular joint (TMJ) problems relating to intermittent use of the appliance. Any initial discomfort is usually transitory[8]. TMJ discomfort usually relates to a myostatic contraction of lateral pterygoid which appears to be the masticatory muscle most affected by use of the appliance[4]. A simple daily isometric exercise of clenching teeth immediately following removal of the appliance, five times for a 5-second hold initiates a reverse stretch reflex that relaxes lateral pterygoid. If continuing TMJ dysfunction is experienced, the appliance should be repositioned in a less protrusive position. Unilateral TMJ discomfort usually indicates that the initial bite registration was asymmetric; the bite registration should therefore be retaken. According to Lowe[5], appliances should be used for sleep for life; however, many patients have reported that snoring was no longer evident after several months of use of the appliance. In these patients it can be assumed that a reduction in pharyngeal oedema and weight loss may be the contributing factors in creating a better airway.

Although use of the appliance in children has been limited, snoring and obstructive sleep apnoea have been eliminated in several children following a relatively short period of use of a mandibular appliance, normal growth undoubtedly being an important factor. In the more severe instances, however, appliance therapy is considered to be more long-term, the duration of which is dependent upon symptomatic relief. Use of a mandibular advancement splint in children is indicated when obstructive sleep apnoea is occurring and is unrelated to either tonsillar or adenoidal hypertrophy.

In the construction of a mandibular advancement splint for edentulous patients, strict attention must be given to ensure that the vertical height of the appliance exceeds the free-way space. The degree of mandibular advancement should be limited to two-thirds of the maximal protrusive distance. Although not as efficient as appliances for dentate patients, the mandibular advancement splint for edentulous patients is still effective in the elimination of snoring and obstructive sleep apnoea.

In those patients for whom appliance therapy was unsuccessful, non-compliance in using the appliance and the severity of obstructive sleep apnoea were usually the most prevalent factors. Appliance therapy is only indicated in the treatment of patients who have mild to moderate obstructive sleep apnoea. Polysomographic testing is, therefore, mandatory when obstructive sleep apnoea is suspected.

In the treatment of severe obstructive sleep apnoea, continuous positive airway pressure (CPAP) is the treatment of choice[9]. Dental appliances do, however, provide another treatment modality for a group of patients who, to date, have no other treatment options. Cephalometric radiographic evaluation before and after insertion of the appliance is essential to establish appliance treatment parameters and to identify those patients who may be "anatomically" at-risk.

A multidisciplinary approach between dentist and doctor is important to reach a diagnosis and establish a plan for appropriate therapy, as obstructive sleep apnoea is a condition which has long-term morbidity and mortality. The key undoubtedly is early diagnosis and treatment. The use of lateral cephalometry in the early detection of those patients who are anatomically at-risk cannot be over-emphasised.

ACKNOWLEDGMENTS

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reports

Treatment of obstructive sleep apnoea in edentulous patients – design of a combination appliance: a case study

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SUMMARY

This report describes the fabrication of a prosthesis to prevent obstructive sleep apnoea in edentulous patients. The objective of treatment in a 62-year-old man was to establish a comfortable protrusive and vertical position of the mandible that minimised hypopharyngeal obstruction nocturnally. An appliance was designed incorporating two concepts in the elimination of obstructive sleep apnoea: mandibular advancement, which maintains hypopharyngeal width nocturnally; and advancement of the tongue with the aid of a device holding the tongue in a protrusive position by vacuum pressure. This combination appliance offers a treatment modality to a large group of otherwise forgotten patients.

Treatment of obstructive sleep apnoea with oral appliances in dentate patients has been well documented, and practice parameters have been established[1]. Unfortunately, information on the treatment of edentulous patients with obstructive sleep apnoea is sparse. Apart from tongue-retaining devices[2], use of oral appliances in this group of patients has been limited.

Meyer and Knudson[3] described a mandibular advancement splint made of acrylic bases that held the mandible in a protrusive position nocturnally. Difficulty was experienced, however, in the treatment of edentulous patients with atrophic mandibles and poor retention of dentures. Use of intra-osseous implants in such patients was recommended.

This report describes an appliance designed to advance the mandible and hold the tongue in a protrusive position nocturnally to eliminate obstructive sleep apnoea.

CASE REPORT

A 62-year-old edentulous male, who had severe obstructive sleep apnoea associated with simultaneous ischaemic heart disease with cardiomyopathy, was referred by a respiratory physician for the construction of a mandibular advancement splint. This patient was unable to tolerate continuous positive airway pressure (CPAP). Because of the severity of cardiomyopathy, a tracheostomy was considered to be the next treatment option.

A mandibular advancement appliance was constructed, consisting of acrylic denture bases jointed by acrylic posts. The mandible was held in a protrusive position that was two-thirds of the patient's maximal protrusive position. The appliance had a vertical height 5 mm greater than the free-way space. This dimension was essential to ensure that dislodgment did not occur nocturnally. To aid maximal protrusion of the tongue, a tongue-retaining device was constructed for attachment to the acrylic posts. This tongue retainer was made of silicone and was vacuum formed. It was able to be removed by the patient without removal of the acrylic splint (Figures 1 and 2).

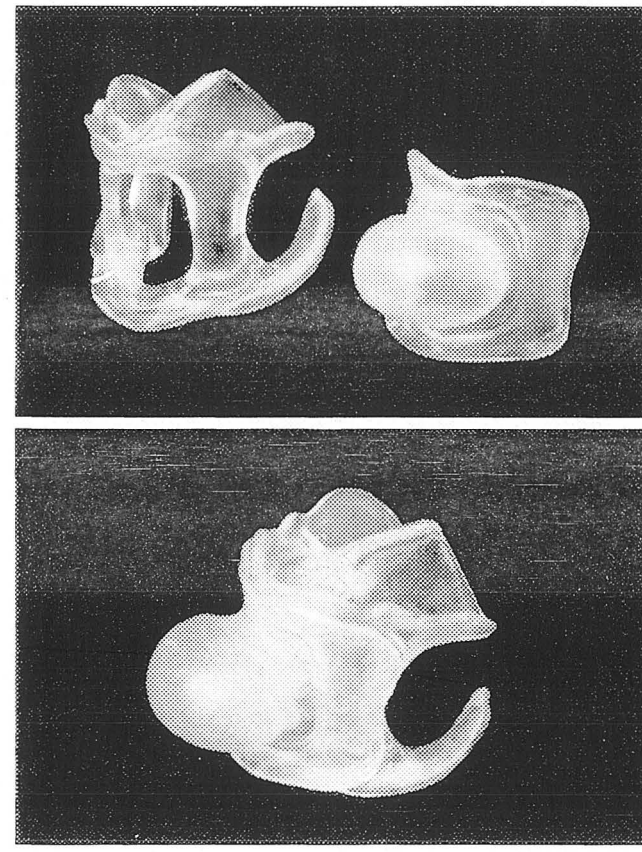

FIG 1 – Upper, mandibular advancement splint and the tongue retainer unattached; lower, mandibular advancement splint with tongue retainer attached.

A lateral headfilm with the appliance *in situ* illustrates the degree of protrusion of the tongue and an elevation of the hyoid bone (Figure 3). Although not able to be quantified, the degree of suction in the tongue-retainer was considerable. Discomfort of the tongue was, however, tolerable. The appliance did not dislodge nocturnally. A subsequent overnight sleep study established a marked reduction in apnoeas and hypopnoeas. The severity of hypoxaemia was not as significant as at a previous sleep study. Although this patient was still at risk, the cardiomyopathy could be controlled medically. A tracheostomy was not considered an option at that time. At a 6-month review, the patient was coping well with the appliance, and his cardiomyopathy was stabilised.

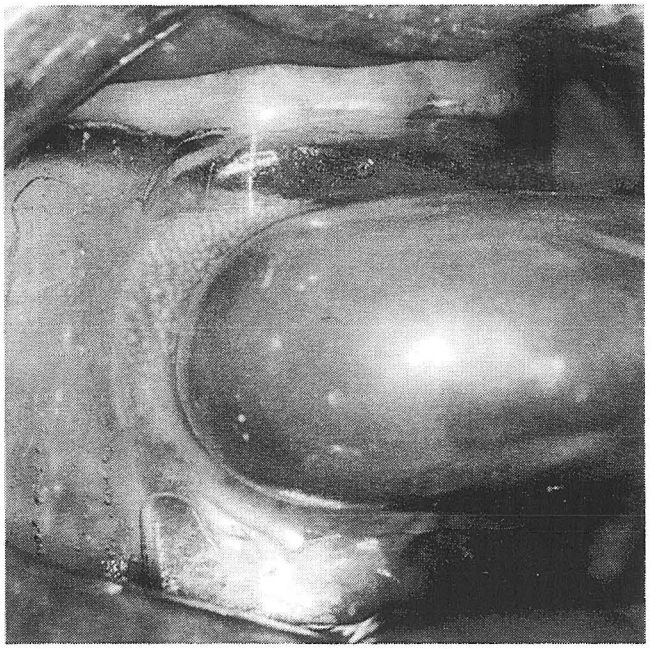

FIG 2 – Mandibular advancement splint with tongue retainer *in situ*.

The combination of a mandibular advancement splint and tongue-retaining device in edentulous patients is only indicated where nasal respiration is considered adequate. Use of a nasal expander is recommended in those patients with nasal restriction.

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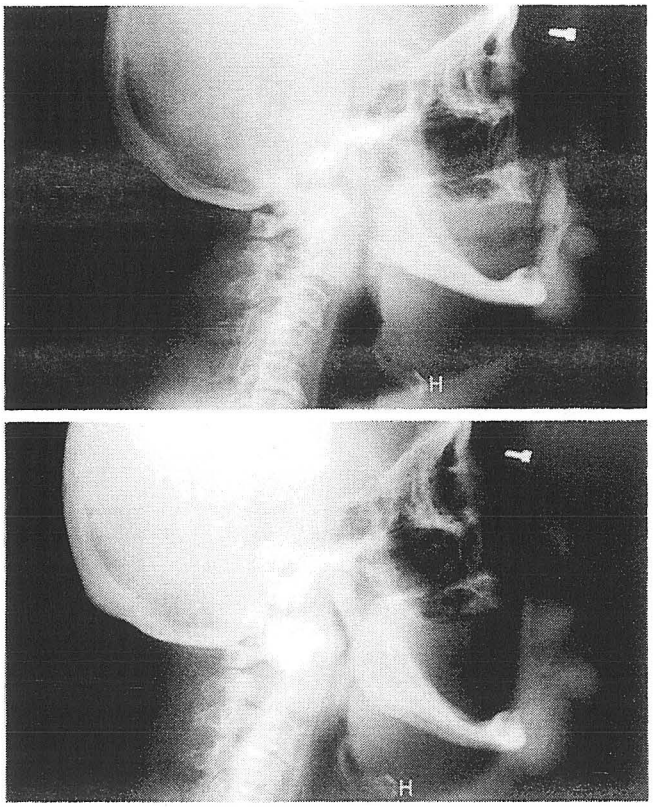

FIG 3 – Upper, lateral cephalogram without mandibular splint and tongue retainer; H shows the position of the hyoid bone position. Lower, lateral cephalogram with mandibular splint and tongue retainer in place; the hyoid bone, H, is now elevated relative to the lower border of the mandible, denoting a more stable antero-superior tongue position.

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Original Article

The effect of long-term mandibular advancement on the hyoid bone and pharynx as it relates to the treatment of obstructive sleep apnoea

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A cephalometric analysis was carried out to determine the effects of long-term mandibular advancement on the hard and soft tissues of the upper airway and, in particular, the relationship of the hyoid bone to both the cranium and the cervical spine, following mandibular advancement. One hundred consecutively-treated patients (87 males and 13 females; mean age: 49.26 years; SD: 8.56; range: 33–74 years) diagnosed with obstructive sleep apnoea and/or habitual snoring were reviewed at 6-month intervals over 6 to 30 months of treatment with a mandibular advancement splint. Significant changes to both the oropharynx and velopharynx were observed. At 12 months, the posterior airway space had increased from 10.71 mm to 11.99 mm (mean difference: 1.28 mm). At 6 months, significant changes had occurred in the soft palate length and thickness: a mean reduction in length of 1.46 mm ($p < 0.0001$) and in thickness of 0.57 mm. No changes were observed in the hypopharynx: the position of the hyoid bone remained unchanged in relation to both the cranium and cervical spine in all linear and angular measurements.
The author concludes that mandibular advancement with oral appliances should be considered as a treatment for life.

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Introduction

The use of mandibular advancement in the treatment of snoring and obstructive sleep apnoea is well established. Little is known, however, of the long-term effects of mandibular advancement on the structures of the upper airway and, in particular, the relationship of the hyoid bone to both the cranium and cervical spine following such treatment.

The hyoid bone is the bony origin of the root of the tongue and the site of insertion of the muscles of the hyoid bio-dynamic system that regulates respiration, mastication, deglutition and phonation.[1] Described by Durzo and Brodie[2] as a "floating bone", the hyoid bone is suspended posteriorly and bilaterally from the cranial base to an anterior attachment near the midline of the mandible, its horns delimiting the laryngeal part of the pharynx within which the superior opening of the larynx is included.[1]

Developmentally in man, the hyoid bone has assumed additional functions over those demanded in other species.[3] With man's change to an upright position, the larynx and trachea are no longer held away from the upper respiratory tract by gravity. As a consequence, patency of the upper airway can only be maintained by those muscles that lie anterior to the hyoid bone. To enhance breathing efficiency, the suprahyoid and infrahyoid muscle groups pull the hyoid bone forward to maintain oropharyngeal airway patency during the inspiratory phase of respiration.[4,5]

Durzo and Brodie,[2] and Bench[5] have shown that the hyoid bone becomes more inferiorly positioned during growth and that in adulthood it is usually situated at the level of the fourth cervical vertebra.[2,5] Kollias and Krogstad,[6] in a longitudinal cephalometric investigation of the position

of the hyoid bone in patients between 22 and 42 years of age, observed that over a 20-year period it assumed a more inferior position in males than in females; however, the horizontal position in relation to the third cervical vertebra was found to be stable for both genders. Tallgren and Solow[7] considered that this stable hyo-cervical relationship was probably due to the close anatomical and functional relationship of the hyoid bone to the laryngeal cartilages. Other investigators have suggested that this stable position reflects the functional demand for maintenance of upper airway patency.[8,9] Tourné[10] considered that the descent of the hyoid bone in older age groups was a functional compensation for an increase in tongue bulk. Tallgren and Solow[7] found that changes in hyoid bone position were related to changes in the position of the mandible and concluded that it was co-ordinated with both facial morphology and head and cervical posture. The relationship of the hyoid bone to natural head position was recently reported by Özbek *et al.*,[11] who found that a lower hyoid bone position in relation to the mandibular plane was statistically related to a cranio-cervical extension.

Cephalometric analyses have been used by many investigators to identify morphological parameters characteristic of obstructive sleep apnoea.[12–17] The position of the hyoid bone in patients with documented obstructive sleep apnoea has been found to be more inferior in relation to the vertical distance to the mandibular plane at a level of C4-C6, than in non-apnoeic controls at a level of C3-C4.[14,18–22]

Aberrations in uvulo-pharyngeal morphology based on cephalometric analyses have also been reported; the typical features in patients with obstructive sleep apnoea including thick and long soft palates, reduced posterior airway space (PAS), and a larger than normal tongue.[23-26] The dimensions of the pharynx are also reported to be considerably reduced, irrespective of whether or not patients were investigated in the upright or supine position.[27–28]

Pracharktan *et al.*,[29] in an assessment of a craniofacial index score (CIS) that was used to differentiate patients with obstructive sleep apnoea from non-apnoeic snorers, found that the following variables had the highest predictive values: hyoid bone to mandibular plane distance, soft palate length and Body Mass Index (kg/m^2). They concluded that a CIS constructed from cephalometric measurements could be used to differentiate patients.

Mandibular advancement has been established as a recognised method of treating patients presenting with mild to moderate obstructive sleep apnoea.[30–37] Isono *et al.*[38] observed endoscopically that the velopharynx was the most common site of obstruction in patients with obstructive sleep apnoea. Their research found that the anterior movement of the mandible widened the retro-palatal airway, as well as that of the base of the tongue, thereby stabilising the airway. They concluded that, as well as increasing the pharyngeal area, a decrease in the collapsibility of the velopharynx may be also due to an increase in palatoglossal arch tension resulting in an improvement of velopharyngeal airway patency. Various other authors have reported changes to the pharynx and the position of the hyoid bone in patients with mandibular advancement splints in situ.[31,36,39–41] Supine cephalometric investigations by Battagel *et al.*[40] and Johal and Battagel[31] observed an upward movement of the hyoid bone relative to the mandibular plane of 4.3 mm and 4.6 mm, respectively, in patients who had appliances *in situ*. These finding were comparable with a 3.0 mm decrease in the same dimension in upright patients, as reported by Bonham *et al.*[36]

Gale *et al.*,[41] using computerisd tomography, reported significant increases in the minimum pharyngeal cross-sectional area in 32 conscious, supine obstructive sleep apnoea patients with similar mandibular advancement. They concluded, however, that there was a wide individual variation that could not be predicted from the degree of mandibular advancement. Mayer and Meier-Ewart[22] found that the position of the hyoid bone relative to the mandible was the only cephalometric parameter to show a direct correlation with respiration; the closer the hyoid bone was positioned to the mandible with mandibular advancement, the greater the improvement in respiration. In light of these findings, it is not unrealistic to assume that any adaptation of the hyoid apparatus to intermittent mandibular advancement may be of benefit to upper airway patency. No studies to the author's knowledge have been undertaken to investigate the effects of long-term mandibular advancement on the structures of the upper airway.

The present investigation is a cephalometric analysis of the effects of long-term mandibular advancement on the hard and soft tissues of the upper airway and, in particular, the relationship of the hyoid bone to both the cranium and the cervical spine, following mandibular advancement.

Materials and methods

In this study, one hundred consecutively-treated dentate patients, medically referred, were treated using mandibular advancement splints for obstructive sleep apnoea and/or habitual snoring. Patients were required to wear a non-adjustable mandibular advancement splint[42] for a minimum of five to six hours per night. Mandibular advancement was established at 75 per cent of maximum protrusion, measured from incisal edge to incisal edge, as determined by the George-gauge[43] (range 3–14 mm). The sample comprised eighty-seven males and thirteen females (mean age: 49.25 years; SD: 8.56 years; range: 33-74 years).

Cephalograms

A cephalogram was taken for all patients at their initial consultation. Following insertion of mandibular advancement splints, review cephalograms were taken at 6-month intervals, over a period of from 6 to 30 months, without the appliance *in situ*. Only one review cephalogram was taken for each patient. Prior to splint insertion, patients were randomly assigned to a 6-month review period, each sample size consisting of twenty patients. Cephalograms were taken with the subjects seated, with maximal intercuspation of the teeth, the lips in light contact, and in natural head position as described by Moorrees and Kean.[44] Bilateral ear rods were inserted into the external auditory meatus to stabilise the head during exposure. All cephalograms were taken by the same operator. Rare earth intensifying screens were used to obtain maximum detail. Exposure was adjusted for each patient, the average being 85 KVP and 15 MA, with a 1.0 second exposure.

Cephalometric measurements

Reference points and planes (Figure 1) were based on anatomically stable structures in the cranial base, maxilla, mandible, pharynx and cervical spine. These landmarks were identified and transferred to mylar film. The co-ordinates of each point on the tracings were digitised twice with a reflex metrograph, and the mean values converted to linear and angular measurements. The student's *t*-test for paired data was used to compare all measurements before (T1) and after (T2) mandibular advancement. A value of $p < 0.05$ was considered as significant. To determine the investigative error, double determinations were taken of ten randomly-selected cephalograms and compared with Dahlberg's formula.[45] The reliability of the cephlometric measurement in this study was found to be comparable to that reported by others.[46,47] The smallest linear error was 0.25 mm (H-MP) and the greatest error, 0.71 mm (apw_4–ppw_4). This latter measurement determined lower pharyngeal depth and was difficult to obtain because of the soft tissue definition. The error in angular measurement was comparable to other studies,[46,47] the largest error being 0.97 degrees for the angle H axis/vert.

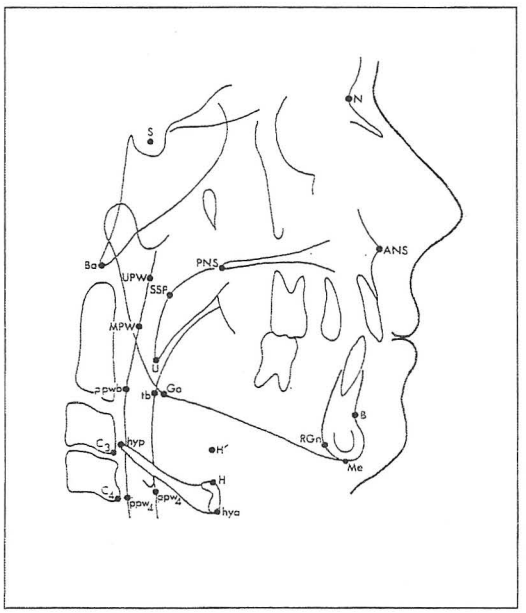

Figure 1. Cephalometric reference points and planes.

Because no lead wire was used during exposure to indicate the true vertical on the cephalogram, the right-hand border of the cephalogram was used as the vertical reference line.[7]

Hyoid measurements vertical (mm)

H–MP	Distance along a perpendicular from H to the mandibular plane (Go-Me)
H–H′	Distance between H and a perpendicular to the line C3-RGn
H–Pal P	Distance along a perpendicular from H to the palatal plane (ANS– PNS)
H–SN	Distance along a perpendicular from H to the anterior cranial base (S–N)

Cephalometric reference points

ANS Anterior nasal spine: the tip of the anterior nasal spine

apw_4 The anterior pharyngeal wall along the line intersecting C4 and H

B B point: the most posterior point on the anterior contour of the lower alveolar process

Ba Basion: the most postero-superior point on the anterior margin of the foramen magnum

C_3 The most inferior anterior point on the third cervical vertebrae

C_4 The most inferior anterior point on the fourth cervical vertebrae

Go The most posterior inferior point on the angle of the mandible

H Hyoidale: the most antero-superior point on the body of the hyoid bone

H′ The intersection point between the perpendicular from H to the line connecting C_3 and RGn

hya The most anterior point of the body of the hyoid bone

hyp The most posterior point of the greater horn of the hyoid bone

Me Menton: the most inferior point on the lower border of the mandibular symphysis

MPW The middle pharyngeal wall. The point on the posterior pharyngeal wall identified by an extension of the functional occlusal plane (FOP)

N Nasion: the most anterior point of the fronto-nasal suture

PAS Posterior airway space. linear distance between tb and ppwb, determined by an extension of the line B through Go to the posterior pharyngeal wall

PNS Posterior nasal spine: the most posterior point of the bony hard palate

ppwb The intersection point of a line from B through Go to the posterior pharyngeal wall

ppw_4 The posterior pharyngeal wall along the line intersecting C_4 and H

RGn Retrognathian: the most inferior posterior point on the mandibular symphysis

S Sella: the centre of sella turcica

SSP The most prominent point on the superior soft palate

tb The intersection point of a line from the point B through Go and the base of the tongue

U The tip of the uvula

UPW The upper pharyngeal wall: the point on the posterior pharyngeal wall identified by an extension of the palatal plane (ANS-PNS) to the upper pharyngeal wall

Reference planes

MP Mandibular plane: the line connecting Go and Me

Pal P Palatal plane: the line connecting ANS-PNS

FOP Functional occlusal plane: the average occlusal plane of the buccal teeth including the canines and first permanent molars.

H axis Long axis of the hyoid bone: the line connecting hya and hyp

True Vert Right border of cephalogram

Angular (°)

H axis/MP The angle between the long axis of the hyoid bone and the mandibular plane

H axis/Pal P The angle between the long axis of the hyoid bone and the palatal plane

H axis/N-Ba The angle between the long axis of the hyoid bone and the line N–Ba

H axis/vert The angle between the long axis of the hyoid bone and the true vertical

Pharyngeal and soft palate measurements (mm)

PAS Posterior airway space: the linear distance between the tongue base (tb) and the posterior pharyngeal wall (ppwb) determined by an extension of the line B through Go to the posterior pharyngeal wall

UPW-PNS Upper pharyngeal depth: the distance between the posterior nasal spine (PNS) and the intersection point between the palatal plane and the posterior wall of the nasopharynx

MPW/U-PNS	Distance between the posterior pharyngeal wall (MPW) and the intersection of a perpendicular to the line U-PNS
$apw_4 - ppw_4$	Lower pharyngeal depth: the linear distance between the pharyngeal anterior and posterior walls defined by the intersection of the line C_4-H
Soft P length	Linear distance between the posterior nasal spine (PNS) and the tip of the uvula (U)
Soft P thickness	The maximum dimension of the soft palate between its oral and nasal surfaces. Linear distance between the point SSP and a perpendicular to the line PNS-U

Anteroposterior (mm)

C_3–H	Distance between C_3 and H
H–RGn	Distance between H and the mandibular symphysis (RGn)

Results

The mean values, standard deviations and mean differences of all patients for the hyoid bone, and for the soft tissue and pharyngeal measurements are given in Tables I and II, respectively. A comparison of the vertical position of the hyoid bone relative to the mandible, maxilla and cranial base (Table I) demonstrated no change following long-term mandibular advancement. A change in the antero-posterior position relative to the third cervical vertebra (C_3-H) was statistically significant ($p < 0.019$; mean difference: 0.52 mm); however, no change was found in the variable H–RGn (Table I). Although statistically significant, this minor change was not considered to be clinically relevant. The measurement C_3–H, using the 12-month data, was not statistically significant ($p < 0.467$; mean difference: 0.25 mm).

Analyses of the angular position of the long axis of the hyoid bone (H–axis) to the mandibular plane (H–axis/MP), the palatal plane, H–axis/Pal–P, the cranial base line (H–axis/N–Ba) and the true vertical (H–axis/vert) were made (Table I), and no change was observed relative to the overlying cranial structures.

An analysis of pharyngeal and soft palate dimensions for all patients is given in Table II. Posterior airway space increased from 10.99 mm to 11.79 mm (mean difference: 0.79 mm; $p < 0.002$), which was significant. A significant increase of PAS was present at 12 months (10.71 mm to 11.99 mm; mean difference: 1.28 mm; $p < 0.009$). A 6-month review found that this change was not statistically significant. Therefore, it can be concluded that an increase in PAS occurs, on average, between 6 and 12 months following mandibular advancement.

Changes to both the length and thickness of the soft palate were observed. In a comparison of all patients, soft palate length decreased from 43.49 mm to 42.02 mm (mean difference: 1.46 mm; $p < 0.0001$). The change was significant both at 12 months (mean difference: 1.40 mm; $p < 0.020$) and at six months (mean difference 1.35 mm; $p < 0.033$). Therefore, it can be concluded that a significant reduction in soft palate length occurs within the first six months of mandibular advancement. This occurrence is probably due to a loss of oedema associated with the elimination of habitual snoring. A reduction in soft palate thickness was observed in a comparison of all the patients (mean difference: 0.57 mm; $p < 0.002$). These changes were not statistically significant at the 12-month review. Although other studies have noted difficulties in landmark identification of soft-tissue structures,[48] the error in soft palate length (0.37 mm) and thickness (0.30 mm) was surprisingly low.[45]

The author attributes changes in the posterior airway space and in the length and thickness of the soft palate to the elimination of habitual snoring, and observed no changes in the position of the hyoid bone relative to its surrounding structures, following long-term mandibular advancement.

Discussion

Cephalometry is one of the more accepted techniques used to evaluate upper airway and craniofacial morphology during the awake period in patients with obstructive sleep apnoea.[49] It is inexpensive and correlates with other, less readily available techniques, such as computer tomography[50] or somofluroscopy.[51] Cephalometry allows a static evaluation of the pharynx, soft palate, tongue and the hyoid bone, relative to their surrounding cranio-cervical structures. Although the obvious limitations of any two-dimensional cephalometric study have been clearly recognised, many practitioners consider that gross changes in tongue position can be assessed by analysing changes in the position of the hyoid bone.[52,53] The position of the hyoid bone is,

Table I. Hyoid bone measurements, before (T1) and after (T2) mandibular advancement (6–30 months).

	(T1)		(T2)			
	Mean	SD	Mean	SD	Mean Diff	*p*
Linear (mm						
H–MP	24.84	5.27	24.78	5.59	-0.06	0.832
C_3–H	40.33	4.89	40.85	5.05	0.52	**0.019**
H–RGn	42.83	5.99	42.64	5.92	0.19	0.580
H–H′	11.49	5.11	11.46	5.38	0.03	0.925
H–PalP	76.50	6.92	77.15	7.14	0.65	0.079
H–SN	125.05	8.51	125.68	8.65	0.63	0.102
Angular (°)						
H axis/MP	8.96	6.13	9.08	6.07	0.12	0.802
H axis/Pal	32.67	8.37	33.51	7.87	0.84	0.147
H axis/N–Ba	60.72	8.43	61.75	8.32	1.03	0.081
H axis/vert	66.81	7.72	67.08	7.46	0.27	0.675

Table II. Pharyngeal and soft palate measurements before (T1) and after (T2) mandibular advancement.

	(T1)		(T2)			
	Mean	SD	Mean	SD	Mean Diff	*p*
Linear (mm)						
6 months						
Soft palate length	43.57	4.42	42.22	4.61	-1.35	**0.033**
PAS	10.77	3.67	11.64	3.70	0.87	0.188
12 months						
Soft palate length	43.75	5.06	42.35	4.48	-1.40	**0.020**
Soft palate thickness	8.56	2.34	7.98	2.39	-0.58	0.063
PAS	10.71	3.18	11.99	3.29	1.28	**0.009**
6–30 months						
PAS	10.99	3.15	11.79	3.19	0.79	**0.002**
UPW–PNS	26.12	3.47	25.93	3.28	-0.19	0.370
MPW/U–PNS	13.33	2.95	13.54	2.55	0.21	0.233
apw_4–ppw_4	16.87	4.09	17.42	3.74	0.54	0.091
Soft palate length	43.49	5.15	42.02	5.14	-1.46	**<0.0001**
Soft palate thickness	8.55	2.21	7.98	2.38	-0.57	**0.002**

therefore, of great clinical interest, as favourable changes to its position may lead to improved airway patency and function.[4,22,36,38]

Changes in the linear and angular measurements of the position of the hyoid bone in this study were similar to other studies, and demonstrated a lower hyoid-to-mandibular-plane distance (MP-H) in apnoeic patients (mean: 24.8 mm) than in non-apnoeic patients (mean: 15.4 mm).[13-19] No change was found relative to the facial structures, following long-term mandibular advancement. The only statistically significant change occurred in the distance C_3-H (mean difference: 0.52 mm); however, Stepovich[54] has stated that measurements of the hyoid bone of less than 2.0 mm may be considered within the realm of individual physiological variation between sequential cephalograms.

Previous studies of the relationship of the hyoid bone to the facial skeleton and cervical column have indicated that the relationship of the hyoid bone to the cervical column is more stable than its relationship to the cranium and mandible.[7,52] In this study, the long axis of the hyoid bone was closely aligned to the mandibular plane, which is in accordance with the findings of Adamidis and Spyropoulos.[55]

Haralabakis *et al.*[9] found that the hyoid axis formed significantly higher angles with the basion-nasion plane (Ba-N), as well as with the palatal plane (ANS-PNS), when their patients with anterior open bites were compared with those with normal occlusions. No difference was found when a comparison was made of the angle between the long axis of the hyoid bone and the mandibular plane. These authors concluded that the hyoid bone moved in close correlation with the pharynx, cervical spine and mandibular plane in patients with entirely different skeletal patterns.[9] The relative position of the hyoid bone appeared to be unchanged even after orthognathic surgery. Takagi *et al.*[56] found little change in the antero-posterior relation of the hyoid bone relative to the cervical vertebrae, following bilateral oesteotomy of the mandible. LaBanc and Epker[57] investigated the relationship between the tongue and the position of the hyoid bone following surgical mandibular advancement and, although changes in tongue posture were observed two years post-operatively, the hyoid bone had returned to its original pre-operative position.

Similar adaptive changes in the position of the hyoid bone were reported by Graber,[58] who observed a greater than normal descent of the hyoid bone after three years of chin-cup therapy for the correction of a Class III malocclusion. Graber concluded that both stability and patency of the pharyngeal airway were primary factors in hyoid bone positioning. Recent longitudinal normative growth studies of the hyoid bone and the pharynx have led to a better understanding of the prevalence of obstructive sleep with increasing age[59] and its gender predisposition.[60] Kollias and Krogstad[6] cephalometrically evaluated the position of the hyoid bone and related structures in patients between 22 and 42 years of age and noted that, although the antero-posterior position remained unchanged during this period, in both genders it assumed a more inferior position with age. The descent was significantly more pronounced in males than in females.

Uvulo-glossopharyngeal changes with age were also reported by Kollias and Krogstad.[61] The overall significant gender differences over a 20-year period were that males showed a more caudally extended tongue mass, with a greater reduction in the sagittal dimension of the minimal pharyngeal airway space. These changes were coupled with a greater increase in the sagittal area of both the soft palate and the tongue. Johnston and Richardson,[62] over a 32-year period also observed an increase in soft palate length and a narrowing of the oropharynax with increasing age. According to Tourné,[10] this latent descent of the hyoid bone, especially in males with increasing age, is a compensatory mechanism to maintain upper airway patency as a result of an increase in tongue bulk. In the light of these findings, soft tissue morphology and maturation changes are the most likely factors in the increasing prevalence of obstructive sleep apnoea with age.[62]

Although an upward elevation of the hyoid bone occurs, on average, with mandibular advancement,[31,36] with associated increases in pharyngeal width,[39-41] these changes are transitory in nature and exist only when an oral appliance is *in situ*. The findings of this present study indicate that long term intermittent mandibular advancement does not have any adaptive effect on either the position of the hyoid bone or the hypopharynx.

In this study, changes were evident when the upper pharyngeal dimensions were considered. Like the findings of Isono *et al.*,[38] mandibular advancement with oral appliances appeared to have the most notable effect on the velopharynx and oropharynx. It should be noted that posterior airway space (a measurement of lower oropharyngeal width), increased significantly from a mean width of 10.71 mm to 11.99 mm (mean

difference: 1.28 mm). These findings are of considerable clinical importance as evidence suggests that any increase in pharyngeal width denotes a significant change in upper airway patency.[36,63,64] This increase in width was thought to be related to a loss of pharyngeal oedema following the elimination of habitual snoring, and possibly due to an increased tonus of the pharyngeal musculature.

Pressure measurement studies have shown that the soft palate region is an obstruction site of central importance in apnoeic patients. Shepard and Thawley[65] found that 56 per cent of apnoeic patients had collapse confined to the velopharynx and retro-palatal segment of the upper airway. Skatvedt[66] found that 70 per cent of apnoeic patients had varying degrees of residual obstruction in the nasopharynx at the level of the soft palate, even after uvulopalatopharynoplasty. Change in the length of the soft palate following long-term use of mandibular advancement is, therefore, of considerable clinical importance. These changes were statistically significant at six months following appliance insertion, and were thought to be attributed to a loss of oedema following elimination of habitual snoring.

Conclusion

This study revealed that long-term mandibular advancement had no adaptive effect on the hypopharynx, whereas, as indicated by significant changes that occurred in both the posterior airway space and the length and thickness of the soft palate, it did have a demonstrable effect on the oropharyngeal and velopharyngeal regions of the upper airway. These findings confirm the author's agreement with Lowe;[64] namely, that mandibular advancement with oral appliances should be considered as a treatment for life as, to date, no treatment appears to be effective in inducing a permanent change in the hypopharynx.

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SLEEP-RELATED BREATHING DISORDERS

Dental and Skeletal Changes Associated with Long-term Mandibular Advancement

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Study Objectives: Little is known of the possible dental or skeletal side effects following the use of mandibular advancement in the treatment of obstructive sleep apnea. A study has subsequently been designed to investigate these issues.

Design: 100 consecutively treated medically referred patients were reviewed cephalometrically in 6-month intervals (6—30 months) following mandibular advancement therapy.

Setting: Orthodontic Private Practice.

Patients: 87 males, 13 females (mean age 49 years, SD 8.5, range 33-74 years).

Interventions: N/A

Measurements and Results: Reference points and planes in the cranial base, maxilla, and mandible were digitized with a reflex metrograph and their means converted to linear and angular measurements. Significant changes following mandibular advancement were observed in lower face height, vertical condylar position, incisor angulation, overbite, and overjet. Skeletal changes were attributed to a vertical repositioning of the mandibular condyle relative to the cranial base and were present at the first review period (6 months). Dental changes occurred later with treatment with the most significant changes occurring at the final review period (30 months) which resulted in a 4.9° proclination of the mandibular incisors and a reduction in overbite of 1.82mm.

Conclusion: The data suggests that long-term use of mandibular advancement can cause dental and skeletal changes which may be progressive over time. As many consider mandibular advancement a treatment for life, it is strongly recommended that all patients be fully informed of the potential for such changes prior to treatment and undergo mandatory dental reviews with long-term mandibular advancement.

Key words: Mandibular advancement; cephalometric; obstructive sleep apnea; dental side effects; longitudinal

INTRODUCTION

THE USE OF ORAL APPLIANCES FOR THE TREATMENT OF OBSTRUCTIVE SLEEP APNEA IS NOW WELL ESTABLISHED. A review of current appliances is given by Schmidt-Nowara et al.[1] along with practice parameters for their use.[2] Despite considerable variation in the design of these appliances it is now recognized that the generic effect of maintaining the mandible in a protrusive position nocturnally is of benefit in the elimination of mild to moderate obstructive sleep apnea.[3,4] Until recently investigations of the possible dental side effects of long-term use of such devices have not been forthcoming. Limited follow-up data indicate that oral discomfort is a common but tolerable side effect and that dental and occlusal complications appear to be uncommon.[1] Theoretical complications of long-term use of mandibular advancement splints include occlusal changes and temporomandibular joint dysfunction (TMD); however, until now a quantitative investigation of these issues has not been formally carried out. Pantin et al[5] reviewed 106 patients treated over a five-year period with mandibular advancement for obstructive sleep apnea. An analysis of dental study models of patients' occlusions prior to and following appliance wear was carried out along with a clinical examination of the temporomandibular joint.

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Although 81% of their sample reported some side effects, namely excessive salivation (30%), xerostomia (23%), temporomandibular joint pain (TMD) (26%), and myofacial pain (25%), these side effects were mostly minor and decreased with continuing use of the appliance. Occlusal changes resulting in a decrease of overjet of 1—3mm were observed in 14% of their sample. Overall, 10 patients in the sample of Pantin et al.[5] discontinued appliance wear due to adverse dental side effects. Pancer et al.[6] evaluated 121 patients following mandibular advancement treatment (65 of whom had worn the appliance for more than one year) and found that 45% of their patients complained of some side effects, the most frequent being excessive salivation (32%), coupled with teeth and jaw discomfort (26%). Occlusal changes were not investigated by Pancer et al.[6] As cephalometric analyses were not undertaken by either Pantin et al.[5] nor Pancer et al.[6] it is difficult to establish the cause of any occlusal changes and whether such changes would continue with ongoing treatment. Although the use of cephalometrics in the treatment of obstructive sleep apnea is not new, it has usually been confined to a diagnostic role in the determination of the differentiation of non-apneic to apneic patients.[7-10] Cephalometric analyses of the effects of long-term mandibular advancement on the facial skeletal and dentition is however, limited. In a pilot study, Bondemark[11] investigated cephalometrically the effects of two years' treatment with mandibular advancement in 30 patients diagnosed with habitual snoring and or obstructive sleep apnea. Overall, Bondemark[11] observed that a small forward and downward change in mandibular position was accomplished by a minor increase in mandibular length in relationship to a decrease in overjet of 0.4mm and a 0.1mm decrease in overbite. A change

in mandibular posture was observed in 17 patients, while 13 patients showed no change. Bondemark[11] concluded that this change in mandibular position might be the result of a condylar and/or glenoid fossa remodeling or a condylar positional change within the fossa as a compensatory reaction to mandibular advancement therapy. As many consider that the treatment of obstructive sleep apnea with oral devices will be lifelong, further investigations are necessary to establish whether changes in the occlusion are progressive with continuing treatment. A long-term cephalometric study has subsequently been undertaken to investigate these issues.

METHODS

Sample

One-hundred consecutively treated medically referred patients using mandibular advancement splints for the treatment of obstructive sleep apnea and or habitual snoring were included in this study. Only patients who stated that they were wearing the appliance seven nights per week (for a minimum of five to six hours per night) were included in this study. A total of 114 patients were initially contacted, 13 patients were excluded from the study as they were not wearing the appliance on a regular basis, one patient declined to be in the study. Overall, 87 males and 13 females were included in this study, mean age 49 years (SD 8.5), range 33—74 years. The mean age for males was 49 years (SD 8.3) and females 51 years (SD 10.2).

The mandibular advancement splint[12] used in this study was non-adjustable and had full occlusal coverage. Mandibular advancement was established at 75% of maximum protrusion as determined by the George-gauge,[13] (range 3—14mm).

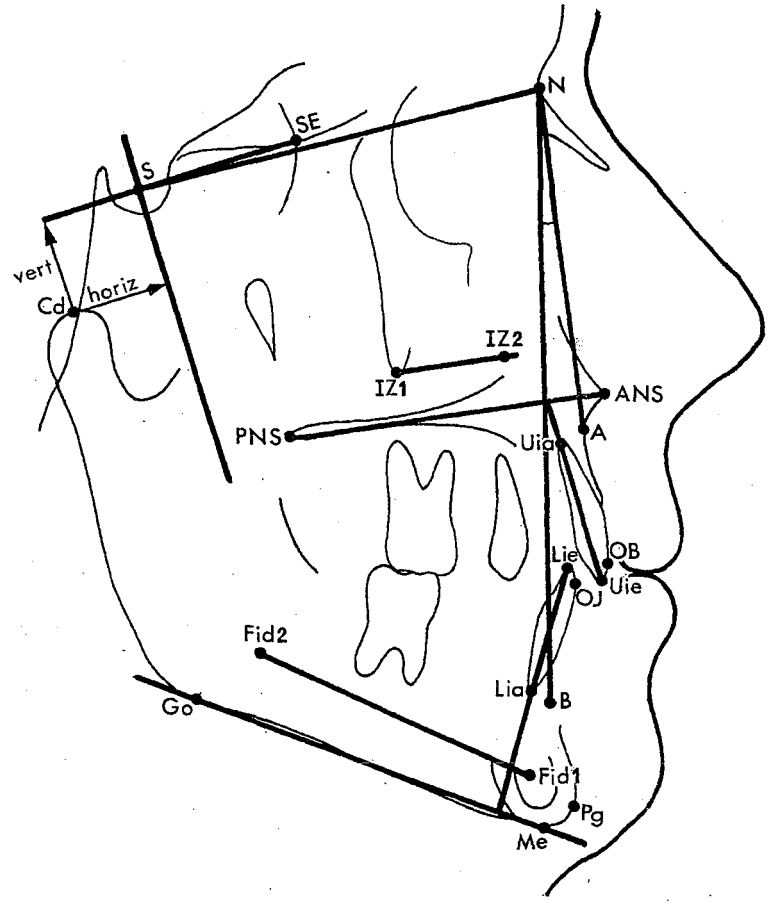

Figure 1—Sagittal skeletal reference points and planes

A—Innermost point on the anterior contour of maxilla; ANS—Tip of the anterior nasal spine in the midsagittal plane; B—The innermost point on the contour of the mandibular alveolar process; Cd—(Condylion) most superior point on the condylar head; Fid1—Fiducial point 1. A point on a natural structure within the mandibular symphysis; Fid2—Fiducial point 2. A point on a natural structure of the posterior body of the mandible; Go—(Gonion) bisector of the mandibular ramal plane and the mandibular plane, on the mandibular border; IZ1—Inferior zygoma (1). The lower most point on the average of the right and left outlines of the zygoma; IZ2—Inferior zygoma (2). The constructed point two centimetres rostral to IZ1 along the line parallel to the floor of the nose through IZ1; Lia—Lower incisor apex; Lie—Lower incisor edge Me—(Menton) most inferior point of the mandibular symphysis; N—(Nasion) anterior point of the intersection between the nasal and frontal bones; OB—(Overbite) vertical overlap between the tips of the upper and lower incisors; OJ—(Overjet) horizontal distance between the labial surface of the most prominent upper incisor and the lower incisor behind this point; Pg—(Pogonion) most anterior point on the anterior symphysis; PNS—(Posterior nasal spine) most posterior point on the bony hard palate in the midsagittal plane S—(Sella) midpoint of the concavity of sella turcica; SE—The point of intersection between the greater wings of the sphenoid and the anterior cranial base; Uia—Upper incisor apex; Uie—Upper incisor edge; MP—(Mandibular plane) Me-Go ; PP—(Palatal plane) ANS-PNS; NSL—(Cranial base reference line) S-N

Table 1—Long-term mandibular advancement before and after treatment (all patients)

Skeletal	Before (T_1)		After (T_2)			
	Mean	SD	Mean	SD	Mean Diff	P
Anteroposterior						
SNA (°)	81.38	3.74	81.70	3.53	0.32	**0.023**
SNB (°)	78.41	3.80	78.42	3.59	0.01	0.927
ANB (°)	3.22	2.10	3.50	2.09	0.29	**0.013**
Maxillary						
ANS- PNS (mm)	54.40	4.09	55.36	3.81	0.95	**0.002**
Mandibular						
Cd - Pg (mm)	125.35	7.19	125.18	7.01	-0.17	0.362
Cd - Go (mm)	67.04	6.32	66.74	6.15	-0.31	0.116
Cd - Vert (mm)	17.61	4.05	18.43	3.85	0.82	**0.0001**
Cd - horiz (mm)	14.79	3.56	14.88	3.55	0.09	0.607
Go - Pg (mm)	80.68	5.14	80.60	5.19	-0.08	0.606

Table 2—Long-term mandibular advancement before and after treatment (all patients)

	Before (T_1)	SD	After (T_2)	SD	Mean Diff	P
Vertical						
N - Me (mm)	130.89	7.81	131.66	7.72	0.76	**0.0001**
N/ANS - PNS (mm)	56.94	3.52	57.05	3.56	0.11	0.356
Me/ANS - PNS (mm)	72.96	5.93	73.62	5.90	0.66	**0.0001**
S -Go (mm)	87.20	7.42	87.70	7.41	0.50	**0.0001**
S/ANS - PNS (mm)	47.99	3.76	47.94	3.84	-0.05	0.560
Go/ANS - PNS (mm)	38.75	6.24	39.31	6.29	0.56	**0.001**
Dentoalveolar						
U1/ANS - PNS (°)	105.91	10.30	104.03	10.03	-1.88	**0.0001**
U1/IZ1 - IZ2 (°)	104.75	8.79	103.14	9.10	-1.58	**0.001**
L1/Me - Go (°)	86.03	10.47	88.85	7.77	2.81	**0.001**
L1/Fid1 - Fid 2 (°)	83.81	10.87	86.53	8.21	2.71	**0.001**
Overbite (mm)	4.09	2.62	3.07	2.10	-1.02	**0.0001**
Overjet (mm)	4.25	2.23	3.19	1.76	-1.06	**0.0001**

Cephalograms

A cephalogram was taken for all patients at their initial consultation. Following insertion of mandibular advancement splints, review cephalograms were taken at six month intervals, over a period of from 6 to 30 months, without the appliance in situ. Only one review cephalogram was taken for each patient. Prior to insertion of the appliance, patients were randomly assigned to a six month review period, (each sample period consisting of twenty patients). Cephalograms were taken with the subjects seated, with maximal intercuspation of the teeth, the lips in light contact, and a natural head position as described by Moorrees and Kean.[14] All cephalograms were taken by the same operator. Rare earth intensifying screens were used to obtain maximum detail. Exposure was adjusted for each patient, but generally 85 KVP and 15 MA was used at one-second exposures.

Cephalometric Measurements

Reference points and planes (Figure 1) were based on anatomically stable structures in the cranial base, maxilla, and mandible. These landmarks were identified and transferred to mylar film by the author (CJR) who was blinded to both the subjects name and whether it was a pre or post treatment cephalogram. The co-ordinates of each point on the tracings were then digitized twice with a reflex metrograph[15] and the mean values converted to linear and angular measurements.

To determine the overall effect of mandibular advancement all patients (n100) were initially included in the pre-treatment group (T1) and were compared to the post-treatment group (T2) using the t-test for paired data. Further analysis of the data was then undertaken comparing each of the review periods (6—30 months) before (T1) and after treatment (T2) in relationship to time. To determine the error in the investigative method, double determinations were taken of 10 randomly selected cephalograms and compared with Dahlberg's formula.[16] The error in the linear measurements fell within the range 0.21mm—0.50mm, and in the angular measurements 0.2°—0.87°. There were no statistically significant differences between the duplicate measurements.

RESULTS

Overall, 39% of the total sample presented with a skeletal I facial pattern, 47% had a skeletal II facial pattern and 14% a skeletal III facial pattern (Appendix 1). An assessment of all patients before (T_1) and after (T_2) treatment with mandibular

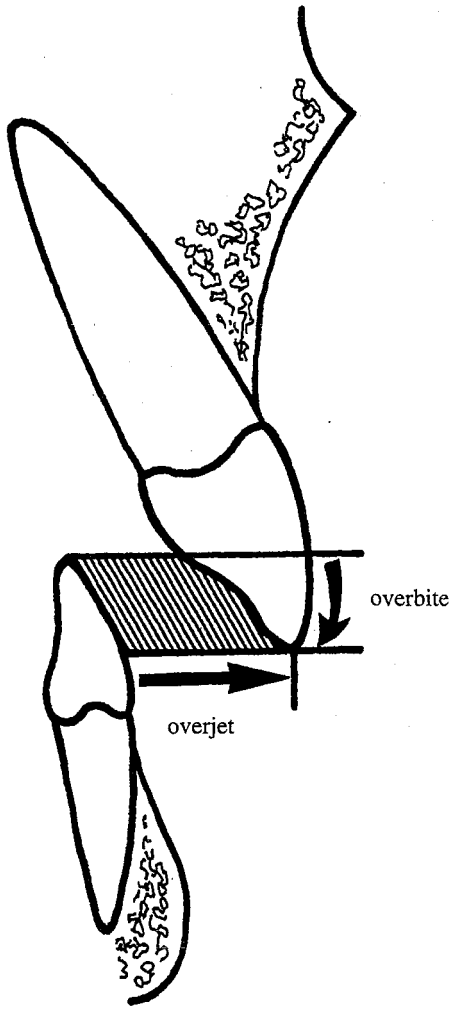

Figure 2—Overbite and overjet

advancement is given in Tables 1 and 2.

Although minor increases in SNA angulation and ANB difference (appendix 1) were noted (0.32° and 0.29° respectively, which were statistically significant) these changes were not considered to be of clinical importance. An increase in palatal length (ANS-PNS) was also observed ($p<0.002$). This linear measurement did, however, have the largest mean error (±0.5mm). Identification of ANS is by nature difficult owing to the size and density of the anterior nasal spine.

A vertical change in condylar position (Cd-vert) was, however, highly significant ($p<0.0001$) (mean difference 0.82mm). No such change was found in condylar horizontal (Cd-horiz), nor any other mandibular skeletal measurement. Changes in vertical face height following long term mandibular advancement were, however, observed and are given in Table 2. The anterior dimensions N-Me, (total anterior face height) and Me/ANS-PNS (lower anterior face height) were both highly statistically significant ($p<0.0001$). Upper anterior face height (N/ANS-PNS) remained relatively unchanged. Statistically significant changes to total posterior face height (S-Go) ($p<0.0001$) and lower posterior face height (Go/ANS-PNS) ($p<0.001$) were likewise observed (Table 2). No change was found in the distance S/ANS-PNS (upper posterior face height). It is therefore considered that lower facial height increases related to a vertical displacement of the mandibular condyle.

Most statistically significant changes occurred in the dento-alveolar measurements (Table 2). The angulation of the upper incisor teeth to the palatal plane (Ui/ANS-PNS) was retroclined by 1.88°. The angulation of the lower anterior teeth relative to the mandibular plane (Li/Me-Go) was proclined by 2.81°. Both overbite and overjet (appendix 1) were reduced by –1.02mm and –1.06mm, respectively. Overall, statistically significant changes occurred in vertical condylar position, lower facial height, maxillary and mandibular incisor angulation, overbite and overjet.

Following the comparison of all patients before (T_1) and after (T_2) mandibular advancement, a further analysis of the data was undertaken to determine at which time interval initial dental and skeletal changes occurred. Prior to this analysis, all groups (6—30 months) underwent a one-way analysis of variance followed by Duncan's multiple range test to determine if initial differences existed between any groups prior to treatment with mandibular advancement. No differences were found.

Statistically significant changes in relationship to time following treatment with mandibular advancement are given in Table 3. At the first review period (six months) statistically significant changes were observed in condylar vertical position (Cd-vert) ($p<0.012$) total anterior (N-Me) and posterior (S-Go) face heights, overbite, and overjet. At the 12 month review period the only observed significant treatment change was a 1.9° retroclination of the maxillary incisors relative to the palatal plane (Ui/ANS-PNS). Following 18 months of treatment, statistically significant changes were found in the following skeletal measurements; vertical condylar position (Cd-vert) (<p0.043) total anterior (N-Me) and lower anterior (Me-ANS) face heights, and in the dento-alveolar measurements a retroclination of 2.3° of the maxillary incisors relative to the palatal plane (Ui/ANS-PNS) and a reduction in overbite and overjet of 0.9mm and 1.06mm respectively.

After 24 months of treatment, changes were observed in vertical condylar position, (Cd-vert) and in total and lower anterior face heights (N-Me and Me-ANS), respectively. Also at the 24-month review period, significantly greater changes were observed in the dento-alveolar variables overbite and overjet when compared to previous review periods, with reductions of 1.36mm and 1.26mm respectively (Table 3). Changes to the angulation of the mandibular incisors relative to the mandibular plane (Li/Go-Me) were first observed at this review period with a proclination of these teeth of 2.2°.

At the final review period (30 months) the greatest reduction in overbite was observed (–1.82mm). This was associated with a marked proclination of the mandibular incisors relative to the mandibular (Go-Me) and constructed planes (Fid_1-Fid_2) of 4.9° and 4.3° respectively.

DISCUSSION

The use of mandibular advancement in the treatment of snoring and obstructive sleep apnea is well established. Little is known, however, of the long-term effects of mandibular advancement on the structures of the facial skeleton, and in particular, the effect on the dentition following such treatment. Cephalometry has been used extensively in the fields of orthodontics and anthropology to record craniofacial form. Recently it has also

Table 3—Significant treatment changes in relationship to time

	(T_1)		(T_2)			
	Mean	**SD**	**Mean**	**SD**	**Mean Diff**	**P**
6 months						
ANS-PNS (mm)	53.39	3.71	54.70	3.02	1.31	**0.015**
Cd-vert (mm)	16.82	3.46	17.89	3.53	1.07	**0.012**
N-Me (mm)	131.02	5.06	132.05	4.93	1.03	**0.0001**
S-Go (mm)	86.88	7.46	87.89	7.47	1.01	**0.001**
Go/ANS-PNS (mm)	38.59	6.12	39.84	5.86	1.25	**0.001**
Overbite (mm)	3.35	1.98	2.74	1.95	-0.61	**0.037**
Overjet (mm)	3.67	1.45	2.80	1.61	-0.87	**0.001**
12 months						
U1/ANS°-PNS°	107.90	7.05	106.00	7.80	-1.9°	**0.035**
18 months						
Cd-Vert (mm)	17.48	3.47	18.41	3.63	0.93	**0.043**
N-Me (mm)	131.63	5.36	132.33	5.43	0.70	**0.033**
Me-ANS (mm)	74.10	4.04	74.66	4.06	0.56	**0.048**
U1/ANS-PNS (°)	107.50	9.37	105.19	9.18	-2.31°	**0.004**
Overbite (mm)	3.83	2.45	2.93	1.74	-0.90	**0.008**
Overjet (mm)	4.36	1.97	3.30	1.62	-1.06	**0.0001**
24 months						
SNA (°)	81.25	4.48	81.95	4.09	0.70	**0.024**
Cd-Vert (mm)	18.04	3.59	19.07	3.24	1.03	**0.007**
N-Me (mm)	130.47	8.36	131.75	8.64	1.28	**0.0001**
Me-ANS (mm)	72.00	5.57	73.05	5.65	1.05	**0.007**
Overbite (mm)	4.50	2.31	3.14	2.21	-1.36	**0.0001**
Overjet (mm)	4.79	3.49	3.53	2.52	-1.26	**0.001**
L1/Me-Go (°)	86.47	9.00	88.66	8.43	2.19°	**0.001**
L1/Fid1-Fid2 (°)	84.56	10.66	86.71	9.87	2.15	**0.002**
30 months						
Me/ANS-PNS (mm)	71.84	6.24	72.68	6.37	0.84	**0.015**
Overbite (mm)	4.43	2.6	2.61	2.27	-1.82	**0.0001**
Overjet (mm)	3.84	1.71	2.63	1.27	-1.21	**0.005**
L1/Me-Go (°)	84.31	9.26	89.24	9.05	4.93°	**0.0001**
L1/Fid1-Fid2 (°)	82.04	9.70	86.34	9.36	4.30°	**0.0001**

been used as an adjunctive procedure for assessing craniofacial patterns associated with obstructive sleep apnea.[7-10]

The results of this present study indicate that both skeletal and dento-alveolar changes can be associated with long-term mandibular advancement. Cephalometrically, dental changes related to a retroclination of the maxillary incisors, a proclination of the mandibular incisors with associated reductions in both overbite and overjet. Skeletal changes related to an increase in vertical face height which was thought to be attributed to a repositioning of the head of the mandibular condyle in the glenoid fossa.

To the author's knowledge only one other cephalometric study has been undertaken to assess the effects of long-term mandibular advancement on the upper facial skeleton and dentition. In a pilot study Bondemark[11] investigated the effects of two years' nocturnal treatment with mandibular advancement in 30 patients diagnosed with habitual snoring and or obstructive sleep apnea. No cephalometric differentiation was made between these groups. Bondemark[11] found that a small statistically significant forward and downward change in mandibular position was accomplished by an increase in mandibular length (0.4mm) with significant changes in both overbite and overjet. Bondemark[11] reported that none of his patients observed any permanent sense of altered occlusions and concluded that the change in mandibular position may be a result of a condylar and/or glenoid fossa remodeling or condylar position changes within the fossa as a compensatory reaction to the advancement of the mandible. In this present study, the position of the mandibular condyle was measured both vertically and horizontally relative to the cranial base reference plane S-SE (this dimension is considered to be unaffected by possible continuing maturation changes.)[18] Changes in condylar vertical position were initially observed at the first review period (six months), along with changes in vertical face height, overbite, and overjet. All of these changes were considered to be related to vertical repositioning of the condylar head (Cd-vert) relative to the cranial base. No changes were observed, however, in the horizontal position of the mandibular condyle (Cd-Horz). Changes in the angulation of the incisor teeth were not evident. From these results it can be concluded that initial skeletal changes were due to a vertical repositioning of the head of the mandibular condyle and not due to a remodelling of the glenoid fossa and/or condylar head owing to the early occur-

rence of these changes. Similar changes were observed at the 18-month review period (Table 3) with the addition of a 2.3° retroclination of the maxillary incisors, along with significant reductions in both overbite (- 0.90mm) and overjet (-1.06mm). Following 24 months of treatment, the first changes in mandibular incisor position were observed, with a proclination of these teeth of 2.2°. The most significant dental changes were, however, observed following 30 months of treatment with a 4.9° proclination of the mandibular incisors and a reduction in overbite of 1.82mm. From these results it can be concluded that changes in the angulation of the maxillary and mandibular incisors tended to occur with increasing length of treatment; conversely, skeletal changes (changes in face height etc) tended to occur soon after the onset of treatment and were most likely attributed to a repositioning of the head of the mandibular condyle within the glenoid fossa. Similarly to Pantin et al,[5] occlusal changes were the reason for discontinuance of treatment in one patient. This patient following 30 months of mandibular advancement had developed a 5mm posterior open-bite (loss of contact of opposing molar teeth) resulting in severe incisal wear due to a marked proclination of the mandibular incisors associated with a retroclination of the maxillary incisors. Transcranial temporomandibular joint radiographs of this patient showed a marked postero-superior increase in joint space between the head of the mandibular condyle and the glenoid fossa. No bony remodeling of the fossa was observed radiographically. Contact of the molar teeth was only re-established after mandibular advancement had been discontinued for nine months. Angulation changes of the incisor teeth were, however, permanent and required corrective orthodontic treatment. This patient was subsequently well controlled with the use of a tongue stabilizer (confirmed by polysomnography).

Although the type of mandibular advancement splint used in this study and the studies of Pantin et al[5] and Bondemark[11] were non-adjustable, most types of mandibular advancement splints share a common functional mechanism. The effect of oral appliance treatment is based on causing an anterior position of the mandible creating anatomic alterations of the oropharyngeal structures. As the majority of oral appliances in common usage today are fully adjustable, it could be argued that the results of this present study are not comparable to those who use fully adjustable devices. Pantin et al[5], however, considered that dental side effects were largely generic to mandibular repositioning and were not generally device specific. In this present study, changes to overbite were related to a vertical displacement of the mandible and not intrusion of the incisor teeth. Changes in overjet were, however, related to an retroclination of the maxillary incisor teeth and a proclination of the mandibular incisor teeth. These changes were thought to be attributed to the forces generated by mandibular repositioning relative to the dentition. Several orthodontic studies on growing individuals have shown some growth modification of the mandible with mandibular advancement.[19-21] In consideration of such changes, Bondemark[11] hypothesized that the treatment of patients with obstructive sleep apnea by forward mandibular repositioning may also have a similar effect in non-growing individuals with subsequent favorable growth changes in oropharyngeal dimensions. In this present study no changes attributed to growth were observed in any of the cephalometric variables measured. As reported elsewhere by the author,[22] any changes evident in the oropharynx or velopharynx following mandibular advancement were attributed to the elimination of habitual snoring resulting in a reduction of pharyngeal odema.

As the use of mandibular advancement in the treatment of obstructive sleep apnea is now a recognized form of treatment, this present study has demonstrated that changes to both the facial skeleton and dentition can occur with treatment over time. Although in most cases these changes go largely unnoticed by the patient, nonetheless prior disclosure to the patient of the potential for such changes along with regular dental reviews should be mandatory in the treatment of patients with mandibular advancement therapy.

APPENDIX 1

Cephalometric Definitions

Skeletal classification[17]—an assessment of the anteroposterior relationship of the maxilla and mandible in the sagittal plane relative to the cranial base line (S-N) defined by the difference (ANB° diff) between the angles SNA° (maxillary position) and SNB° (mandibular position). N being nasion (anterior point of the intersection between the junction of the nasal and frontal bones) and S (Sella); midpoint of the concavity of sella turcica. (A) being the inner most point on the anterior contour of the maxilla and (B) being the innermost point on the contour of the mandibular alveolar process. Skeletal I—normal anteroposterior relationship between maxilla and mandible as defined by cephalometric norms, (maxilla anterior to mandible, ANB diff 1-3°). Skeletal II—mandible postnormal to maxilla (ANB diff >3°) Skeletal III—mandible prenormal to maxilla (ANB diff <1°) Overbite (Fig 2)—the vertical distance in millimeters between the tips of the maxillary and mandibular incisors Overjet (Fig 2) horizontal distance in millimeters between the labial surface of the most prominent maxillary incisor and the mandibular incisor behind this point

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Cranial base considerations between apnoeics and non-apnoeic snorers, and associated effects of long-term mandibular advancement on condylar and natural head position

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SUMMARY One hundred consecutively medically referred patients (58 apnoeic and 42 asymptomatic snorers) were reviewed cephalometrically at six-monthly intervals (6–30 months) following treatment for obstructive sleep apnoea (OSA) and/or habitual snoring by mandibular advancement. Eighty-seven males and 13 females (mean age 49 years, SD 8.5, range 33–74) were included in this study. Reference points and planes in the cranial base, nasopharynx, and mandibular condyle were digitized with a Reflex Metrograph and their means converted to linear and angular measurements.

No statistically significant differences were observed between the apnoeic and non-apnoeic groups in either their skeletal or cranial base measurements. All linear cranial base dimensions were, however, reduced in the apnoeic group, with the exception of the distance (S–SE). Following mandibular advancement, statistically significant changes were observed in vertical condylar position (Cd–vert) with changes occurring at 6 ($P < 0.012$), 18 ($P < 0.043$), and 24 months ($P < 0.007$). No changes in horizontal condylar position (Cd–horiz) were found. Significant changes were observed in natural head position (NHP) with a reduction from an extended (NSL–vert 99.7 degrees) to a more upright NHP (NSL–vert 93.0, $P < 0.001$).

Introduction

Abnormal craniofacial morphology has been extensively reported in patients presenting with obstructive sleep apnoea (OSA). Cephalometric radiographs have shown a number of skeletal and soft tissue abnormalities of the upper airway that predispose patients to pharyngeal occlusion, which are reported to be related to the severity of OSA. In particular, these include a decreased anterior cranial base length (S–N; Bacon *et al.*, 1990; Andersson and Brattström, 1991; Zucconi *et al.*, 1993), an acute cranial base angle (S–N–Ba degrees) (Steinberg and Fraser, 1995), and a decreased bony pharyngeal aperture and small oropharyngeal airway (Lowe *et al.*, 1986; Bacon *et al.*, 1990; Tangugsorn *et al.*, 1995; Ono *et al.*, 1996; Pracharktam *et al.*, 1996; Pae *et al.*, 1997). Although normal in all other respects, non-apnoeic snorers have also been shown to have smaller bony pharyngeal apertures (Zucconi *et al.*, 1992; Johns *et al.*, 1998). Conversely, there are reports that indicate that cephalograms of apnoeic patients and those of habitual snorers do not differ (Hochban and Brandenburg, 1994; Cistulli, 1996). Limited data suggest that ethnicity can play an important role in the cephalometric differentiation of patients with and without OSA (Sakakibara *et al.*, 1999; Coltman *et al.*, 2000; Liu *et al.*, 2000), and that comparisons between apnoeic and non-apnoeic patients can only be made when ethnicity has been considered, as differences in cranial base dimensions can

influence cephalometric values (Sassouni, 1962). The relationship between natural head position (NHP) and OSA has been demonstrated (Solow *et al.*, 1993; Tangugsorn *et al.*, 1995). OSA patients were found to exhibit an extended and forward NHP when compared with controls. Minor changes in NHP were initially caused by a cranial extension (NSL–vert; Woodside and Linder-Aronsen, 1979; Hellsing, 1989; Solow *et al.*, 1996). These findings were not, however, substantiated by Özbek *et al.* (1998), who concluded that this difference was due to the inclusion of non-apnoeic snorers in their control group. To-date, only one cephalometric study has been undertaken to examine the effect of long-term mandibular advancement on the facial skeleton.

Bondemark (1999) cephalometrically investigated the effects of two years' treatment with a mandibular advancement appliance in 30 patients diagnosed with habitual snoring and/or OSA. That author found that a small forward and downward change in mandibular position was accomplished by a minor increase in mandibular length associated with a decrease in overjet of 0.4 mm and a 0.1-mm decrease in overbite. A change in mandibular posture was observed in 17 patients, whilst 13 subjects showed no change. Bondemark (1999) concluded that this change in mandibular position may be the result of condylar and/or glenoid fossa remodelling or a condylar positional change within the fossa as a compensatory reaction to mandibular advancement therapy. To the author's knowledge no cephalometric studies have been undertaken to determine the effect of long-term mandibular advancement on the upper facial skeleton and in particular its effect on condylar position and NHP. This investigation was therefore undertaken to investigate these issues.

Subjects and methods

Sample

One hundred consecutively treated medically referred patients using a mandibular advancement appliance for OSA and/or habitual snoring were included in this study. Fifty-eight patients were referred from the Tom McKendrick Sleep Laboratory, Dunedin Hospital, for the treatment of mild to moderate OSA, six patients in this group were non-compliant with nasal continuous positive airway pressure. The remaining 42 subjects were referred for the treatment of non-apnoeic snoring. Only patients who stated that they wore the appliance seven nights per week (for a minimum of 5–6 hours per night) were included. A total of 114 patients were initially contacted; 13 subjects were excluded as they did not wear the appliance on a regular basis, one patient declined to be in the study. Overall, 87 males and 13 females were included, mean age 49 years (SD 8.5), range 33–74 years. The mean age for males was 48 years (SD 8.3) and for females 51 years (SD 10.2).

The mandibular advancement splint used was non-adjustable and had full occlusal coverage (Robertson, 1997). Mandibular advancement was established at 75 per cent of maximum protrusion as determined by the George-gauge (range 3–14 mm; George, 1992).

Cephalograms

A cephalogram was taken for all patients at their initial consultation. Following insertion of the mandibular advancement splint, review cephalograms were taken without the appliance *in situ*, at six-monthly intervals from 6 to 30 months. Only one review cephalogram was taken for each patient. Prior to insertion of the appliances all patients were randomly assigned to a six-month review period (each period consisting of 20 patients). Cephalograms were taken with the subjects seated, with maximal intercuspation of the teeth, the lips in light contact and in NHP as described by Moorrees and Kean (1958). All cephalograms were taken in the afternoon by the same operator. Rare earth intensifying screens were used to obtain maximum detail. Exposure was adjusted for each patient, but generally 85 KVP and 15 MA was used at 1.0-second exposures. Prior to exposure all patients were clinically examined by the author to check for occlusal discrepancies between centric occlusion (CO) and centric relation (CR).

Cephalometric measurements

Reference points and planes (Figure 1) were identified and transferred to mylar film. The co-ordinates of each point on the tracings were recorded twice with a Reflex Metrograph (Scott, 1981) and the mean values converted to linear and angular measurements.

The *t*-test for paired data was used to compare all measurements between the before (T1) and after treatment (T2) groups; a value of $P < 0.05$ was regarded as significant. To determine the error of the method, double determinations were taken of 10 randomly selected cephalograms and compared using Dahlberg's (1940) formula. The error in the linear measurements fell within the range 0.20–0.47 mm, and in the angular measurements 0.21–0.79 degrees. There were no statistically significant differences between the duplicate measurements.

Because no lead wire was used during exposure to indicate the true vertical on the cephalogram, the right-hand border of the cephalogram was used as the vertical reference line (Tallgren and Solow, 1987). The head position in relation to the vertical was expressed by the angle (NSL–vert).

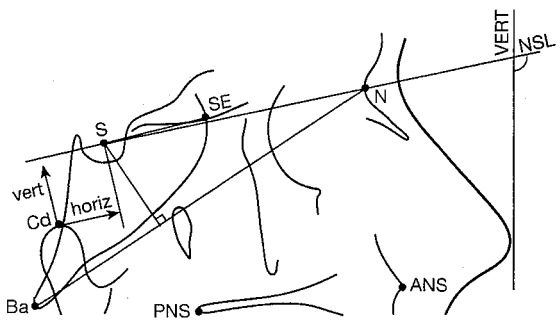

Figure 1 Reference points and planes used in this study. Ba (basion), lowest point on the anterior margin of foramen magnum; Cd (condylion), most superior point of the condylar head; N (nasion), anterior point of the intersection between the nasal and frontal bones; PNS (posterior nasal spine), most posterior point of the bony hard palate in the mid-sagittal plane; S (sella), mid-point of the concavity of the sella turcica; SE (spheno-ethmoidal registration point), the point of intersection between the greater wings of the sphenoid and the anterior cranial base; Cd vert, the vertical distance between the mandibular condyle and an extension of the line S–SE; Cd horiz, the horizontal distance between Cd to a line perpendicular to the line S–SE through S; S–SE, cranial base reference plane; NSL, nasion–sella line (N–S); VERT, true vertical, right hand border of cephalogram; S perp N–Ba (sella vertical position), the perpendicular distance between sella and N–Ba.

Results

After adjusting for age and sex (ANOVA), no statistically significant differences were found between the apnoeic and non-apnoeic snoring groups in relationship to their skeletal classifications (Table 1). Female patients in both groups had, however, statistically significantly smaller overall linear skeletal measurements when compared with their male counterparts. No differences were observed when angular measurements were considered.

Overall, 39 per cent of the total sample presented with a Skeletal I facial pattern, 47 per cent Skeletal II, and 14 per cent Skeletal III (Table 1).

Although most cranial base linear dimensions were reduced in the apnoeic group (Table 2) no statistically significant differences were observed between patients with documented OSA and those presenting with asymptomatic snoring. The only linear measurement not reduced in the apnoeic group was the distance (S–SE). Differences in linear measurements between apnoeic patients and non-apnoeic snorers, therefore, do not include that aspect of the anterior cranial base from sella to the intersection of the

Table 1 A comparison of the skeletal classifications of apnoeic and non-apnoeic snorers.

	Skeletal classification			Row total
	I	II	III	
Snorers	16	20	6	42
Apnoeics	23	27	8	58
Total	39	47	14	100

Chi-square	Value	DF	Significance
Pearsons	0.02533	2	0.98742

Skeletal classification (Tulley and Campbell, 1970).
Skeletal I (ANB difference 1–3 degrees).
Skeletal II (ANB difference >3 degrees).
Skeletal III (ANB difference <1 degrees).

Table 2 Cranial base and nasopharyngeal measurements of apnoeics and non-apnoeic snorers.

	Non-apnoeic snorers		Apnoeics		Mean difference	*P*
	Mean	SD	Mean	SD		
Linear (mm)						
N–Ba	112.45	5.39	111.48	5.47	0.97	0.382
S–N	75.47	3.85	74.84	3.72	0.63	0.409
S–Ba	50.50	3.54	49.66	3.83	0.83	0.269
N–Cd	91.46	4.75	90.42	5.14	1.04	0.307
S–SE	26.70	2.53	26.97	2.67	–0.26	0.615
S–Cd	24.02	3.83	22.88	3.20	1.14	0.109
Cd–vert	18.40	4.52	17.02	3.60	1.37	0.095
Cd–horiz	14.85	3.81	14.75	3.39	–0.10	0.891
S perp–N–Ba	27.50	3.29	26.78	3.67	0.73	0.316
Ba–PNS	45.88	4.16	44.56	4.73	1.32	0.152
Angular (degrees)						
N–S–Ba	125.5	5.6	126.3	6.3	–0.8	0.520
N–S–Cd	125.9	8.7	127.2	8.5	–1.2	0.481
NSL–vert	97.8	14.4	101.0	14.0	–3.2	0.275

sphenoid and jugum. No significant differences were observed between angular measurements (Table 2); however, apnoeics tended to have a greater cranial extension (NSL–vert 101.0 degrees) compared with non-apnoeic snorers (NSL–vert 97.8 degrees).

As no statistically significant differences were observed between the apnoeic and non-apnoeic groups, both groups were combined to investigate the effect of long-term mandibular advancement on the upper facial skeleton and in particular the relationship of the mandibular condyle to the cranial base. Significant differences were observed in condylar position relative to sella (S–Cd, $P < 0.001$; Table 3). Further analyses of condylar position were

Table 3 Cranial base and nasopharyngeal dimensions before (T1) and after (T2) treatment.

All patients ($n = 100$)	T1		T2		Mean difference	*P*
	Mean	SD	Mean	SD		
Linear (mm)						
N–Ba	111.88	5.43	111.95	5.48	–0.07	0.700
S–N	75.10	3.77	75.00	3.76	0.10	0.324
S–Ba	50.01	3.72	50.17	3.54	–0.16	0.356
N–Cd	90.85	4.99	90.94	4.92	–0.09	0.607
S–SE	26.86	2.60	26.83	2.51	0.03	0.702
S–Cd	23.35	3.50	23.97	3.48	–0.62	0.001*
Cd–vert	17.61	4.05	18.43	3.85	–0.82	0.000*
Cd–horiz	14.79	3.56	14.88	3.55	0.09	0.607
S perp–N–Ba	27.08	3.52	27.10	3.39	–0.02	0.836
Ba–PNS	45.10	4.53	45.04	4.18	0.06	0.763
Angular (degrees)						
N–S–Ba	125.9	6.0	126.0	5.9	0.1	0.832
N–S–Cd	126.6	8.6	125.6	8.2	1.0	0.026*
NSL–vert	99.7	14.2	93.0	18.0	6.7	0.001*

*Significant difference between groups.

made in the vertical (Cd–vert) and horizontal (Cd–horiz) planes relative to the cranial base reference line (S–SE). No statistically significant changes were found for the horizontal aspect (Cd–horiz), but significant changes were observed in the vertical aspect (Cd–vert, mean diff 0.82 mm, $P < 0.001$), when all patients were compared (Table 3). Likewise, a small but statistically significant change was observed in the angle N–S–Cd degrees ($P < 0.026$). This was thought to be related to a change in the vertical position of condylion. A statistically significant change in NHP was also observed, with a reduction of (NSL–vert) from 99.7 to 93.0 degrees ($P < 0.001$). This resulted in a change in head posture from an extended to a more upright NHP (Table 3).

Following comparison of all patients at T1 and T2, a one-way analysis of variance followed by Duncan's multiple range test was used to determine whether initial differences existed between any group prior to treatment with a mandibular advancement appliance. No differences were found. Further analysis of the data was then undertaken to determine at which time interval changes occurred.

Changes in condylar vertical position relative to length of treatment are given in Table 4. Condylar displacement was first observed at 6 ($P < 0.012$), at 18 ($P < 0.043$), and 24 months ($P < 0.007$). Although changes occurred at 12 and 30 months, these differences did not reach a statistical level of significance. Further evaluation of the data by analysis of covariance was then carried out to determine whether changes were progressive with continuing treatment. After adjusting for time no statistically significant differences were observed in condylar vertical position between any of the groups.

Conversely, continuing changes in NHP occurred between 12 and 30 months ($P < 0.019$), and 18 and 30 months ($P < 0.008$); however, as no other inter-group changes were found these findings were considered inconclusive.

Discussion

All patients in this study were referred for treatment of mild to moderate OSA and/or non-apnoeic snoring. Historically, the differentiation between these two groups has caused some debate especially at the lower end of the spectrum. The minimal diagnostic criteria for OSA has been defined as an apnoea-hypopnoea index (AHI) of 5 or greater with daytime hypersomnolence (Young *et al.*, 1993). Based on epidemiological evidence (Engleman *et al.*, 1997, 1999; Young *et al.*, 1997) the American Academy of Sleep Medicine Task Force (1999) defined mild OSA as an AHI of 5–15 events per hour, moderate OSA 15–30 events per hour, and severe OSA greater than 30 events per hour. Morbidity associated with OSA with AHI values as low as 5 has been reported (Peppard *et al.*, 2000), likewise simple snoring has been reported as being the beginning of sleep-disordered breathing with a significant risk for elevated blood pressure (Young *et al.*, 1996). The differentiation between patients with OSA and those with non-apnoeic snoring is undoubtedly difficult and is arbitrarily based on a diagnostic cut-off value (AHI > 5 <) with or without associated daytime symptoms. Nonetheless, the diagnostic criteria used

Table 4 Changes in vertical condylar position relative to length of treatment.

Months	Before treatment (T1)		After treatment (T2)		*P*
	Mean	SD	Mean	SD	
6	16.82	3.46	17.89	3.54	0.012*
12	17.83	5.49	18.10	5.51	0.582
18	17.48	3.47	18.41	3.64	0.043*
24	18.05	3.58	19.07	3.24	0.007*
30	18.21	4.30	18.92	3.29	0.198

*Significant difference between groups.

in this study to differentiate patients presenting with OSA and those with non-apnoeic snoring are in accordance with current international recommendations (American Academy of Sleep Medicine Task Force, 1999).

As well as polysomnography to evaluate patients with sleep-disordered breathing, cephalometry has shown a number of skeletal abnormalities of the cranial base and nasopharynx that many consider to be related to the onset and severity of OSA. Decreased anterior cranial base length (Bacon *et al.*, 1990; Tangugsorn *et al.*, 1995) and an acute cranial base angle (S–N–Ba; Jamieson *et al.*, 1986; Andersson and Brattström, 1991; Steinberg and Fraser, 1995), along with a reduced nasopharynx (Ba–PNS; Zucconi *et al.*, 1992; Ono *et al.*, 1996; Pae *et al.*, 1997) have been reported. Conversely, no such differences in these dimensions have been reported in the differentiation of mild apnoeics to non-apnoeic snorers. In agreement with the findings of Johns *et al.* (1998), although trends suggest a shortening of cranial base dimensions, no statistically significant differences were found between mild apnoeics and non-apnoeic snorers in the present study.

A cautionary note should be given, however, to the interpretation of some of these findings of cranial base discrepancies between patients with documented OSA and those without reported by others. It is important that cephalometric ethnic differences are considered.

In a cephalometric comparison of Chinese and Caucasian patients with OSA matched for age, gender, and skeletal patterns, Liu *et al.* (2000) reported that the Chinese groups revealed more underlying craniofacial skeletal discrepancies with significantly smaller maxillae and mandibles, and in particular steeper and shorter anterior cranial bases. These authors concluded that such cephalometric differences may indicate the need to consider ethnicity when planning treatment of OSA. It is therefore important that inter-racial differences are taken into account when cranial base measurements are used in the diagnosis of patients with OSA. In the present study, all patients were of Caucasian origin. Another cause of concern relates in part to the variability of nasion. For example, in a comparison of Caucasian and Chinese males, nasion is on average higher in Chinese by 4 mm (Sassouni, 1962). Likewise, nasion is higher in Negroes by 2 mm when compared with Caucasians (Sassouni, 1962). Binder (1979) found that an antero-posterior displacement of nasion of 5 mm or more resulted in a 2.5-degree deviation in the ANB angle. It can be concluded, therefore, that the relative position of nasion can influence any cephalometric measurement incorporating this landmark if ethnicity is not accounted for. In lieu of these findings, the cranial base dimension (S–SE) was used as the major cranial base reference line to compare condylar position before (T1) and after (T2) mandibular advancement.

This dimension (S–SE) is considered to be unchanged from adolescence (Ford, 1958) and is regarded as one of the most stable landmarks in the facial skeleton (Johnston, 1996). Of note (S–SE) was the only cranial base linear dimension that was not reduced when apnoeic patients were compared with asymptomatic snorers. Overall, although all cranial base and nasopharyngeal dimensions were reduced in the apnoeic group, these were not statistically significant. These findings may be related to the severity of OSA in the symptomatic group, as only patients with documented mild to moderate OSA were referred for management of their condition with mandibular advancement. Steinberg and Fraser (1995) investigated the cranial base length and flexure in adult patients with documented OSA with asymptomatic snorers and observed that the cranial base flexure was significantly more acute in the apnoeic group ($P < 0.001$). No cranial base length differences were observed. Steinberg and Fraser (1995) considered that an acute cranial base angle played a role in the development of OSA by the anterior repositioning of the posterior pharyngeal wall with a consequential decrease in pharyngeal airway dimension. These authors concluded that cranial base length abnormalities were not common in patients with airway problems and, as a consequence, did not play a role in the aetiology of OSA.

In the present study no angulation differences in cranial base flexure were observed between the non-apnoeic and apnoeic groups (Table 2).

In agreement with the findings of Johns *et al.* (1998), however, differences were observed in nasopharyngeal depth (BA–PNS) and, although not statistically significant, this dimension recorded one of the greatest mean differences (1.32 mm) between the apnoeic and non-apnoeic groups. As many (Bacon *et al.*, 1990; Tangugsorn *et al.*, 1995; Ono *et al.*, 1996; Pracharktam *et al.*, 1996; Pae *et al.*, 1997; Özbek *et al.*, 1998; Johnston and Richardson, 1999) consider a reduced bony nasopharyngeal aperture to be significant in the patency of the upper airway, a difference in this dimension may well be a major factor in the differentiation between apnoeic and non-apnoeic snoring patients.

Following treatment with a mandibular advancement appliance statistically significant changes were found in condylar vertical position (Table 4) at 6 ($P < 0.012$), 18 ($P < 0.043$), and 24 months ($P < 0.007$). Although changes in condylar position occurred at 12 and 30 months, these changes did not reach a level of statistical significance. After adjusting for time by analysis of covariance, changes in condylar vertical position were not progressive with ongoing treatment. These findings may indeed be related to the fact that a non-adjustable mandibular advancement appliance was used in this study.

Studies investigating the effect of long-term mandibular advancement on the facial skeleton have been limited. Bondemark (1999) observed a forward and downward change in mandibular posture after two years' nocturnal treatment with a mandibular advancement appliance. He speculated that this may be the result of condylar or glenoid fossa remodelling as a compensatory reaction or, indeed, a functional adaptation in mandibular position. In the present study, as statistically significant changes in condylar vertical position were observed as early as 6 months with mandibular advancement, these changes are unlikely to be related to glenoid fossa or condylar remodelling, but repositional changes of the head of the mandibular condyle.

As reported previously (Robertson, 2001), in this present study treatment was discontinued for one patient due to adverse occlusal changes. This patient after 30 months of mandibular advancement treatment had developed a 5-mm posterior open bite resulting in severe incisal wear due to a marked proclination of the lower incisors and a retroclination of the upper incisors. Transcranial temporomandibular radiographs of this patient showed a marked postero-superior increase in joint space between the head of the mandibular condyle and the glenoid fossa. No bony remodelling of the fossa was observed radiographically. Posterior contact of the molars was re-established, but only after mandibular advancement therapy had been discontinued for 9 months. Angulation changes of the incisors were, however, permanent and required corrective orthodontic treatment. The patient was subsequently well controlled with the use of a tongue stabilizer. In light of the reversibility of these changes it can be assumed that in this case occlusal changes were related to condylar repositioning, and not to growth modification of the condylar head or glenoid fossa.

Previous studies of the interactions between airway adequacy and head posture have demonstrated that minor changes in NHP lead to a changed mode of breathing that is caused by cranial extension (NSL–vert; Hellsing, 1989; Solow *et al.*, 1993; Tangugsorn *et al.*, 1995; Özbek *et al.*, 1998). These findings are in accordance with the present study and, although not statistically significant, all apnoeic patients had an extended NHP when compared with non-apnoeic snorers (mean difference 3.2 degrees). Many investigators have, however, noted marked differences in NHP in so-called 'normals' (Solow and Tallgren, 1971; Foster *et al.*, 1981; Leitao and Nanda, 2000). However, Özbek and Koklu (1993) considered that alternations in the variable (NSL–vert) could be explained by the inter-individual variability of the vertical position of sella. In the present study, the vertical position of sella was investigated relative to the cranial base reference line Ba–N (S perp N–Ba; Table 2). No statistically significant differences were observed in sella vertical position between the apnoeic and non-apnoeic groups. To the author's knowledge, no study has been undertaken that has investigated changes in NHP associated with long-term mandibular advancement. A reduction from an extended NHP (NSL–vert 99.7 degrees) to a more upright 'normal' NHP (NSL–vert

93.0 degrees) may indeed be an indication of improved upper airway function. Owing to the individual variability of NHP, these changes were only significant, however, when the entire sample ($n = 100$) was compared (Table 3).

Conclusions

No statistically significant differences in cranial base or nasopharyngeal dimensions between patients with OSA and those with asymptomatic snoring were found in this investigation. This may in part be related to the severity of the patients referred for treatment with a mandibular advancement appliance. Changes observed in vertical condylar position (Cd–vert) following long-term mandibular advancement are thought to be related to changes in mandibular position reported by others (Bondemark, 1999). Although not confirmed by polysomnography, changes in NHP from an extended to a more upright position may be coincidental with improved upper airway function following long-term mandibular advancement.

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ORIGINAL ARTICLE

The Efficacy of a Novel Tongue-Stabilizing Device on Polysomnographic Variables in Sleep-Disordered Breathing: A Pilot Study

Ruth N. Kingshott, Ph.D.,[1] David R. Jones, R.P.S.G.T.,[1] D. Robin Taylor, M.D.,[1] and Christopher J. Robertson, M.D.S.[2]

ABSTRACT

The polysomnographic efficacy of a novel tongue-stabilizing device (TSD) in the treatment of snoring and sleep-disordered breathing (SDB) was evaluated in this pilot study. Six current users of the TSD with SDB underwent polysomnography with and without the TSD in situ in a randomized crossover design. The TSD significantly lowered the frequency of snores per hour slept (61- to 70-dB range) (no TSD: mean = 41/h slept ± 52 SD; TSD: 8/h slept ± 16 SD; P = 0.046) but did not alter snoring in the other decibel ranges (all Ps > 0.1). Trends were found for reductions in the frequency of apneas plus hypopneas (no TSD: 26/h slept ± 17/h slept; TSD: 15/h slept ± 13; P = 0.06) and oxygen desaturations of 4% or more (no TSD: 10/h slept ± 10; TSD: 5/h slept ± 5; P = 0.09). Significant improvements in microarousal frequency with the TSD were found (no TSD: 34/h slept ± 16; TSD: 22/h slept ± 14; P = 0.004). Significant reductions in percentage of Stage 1 sleep with the TSD were also demonstrated (no TSD: 10 ± 3%; TSD: 8 ± 2%; P = 0.03). The results of this small pilot study indicate that the TSD may be effective in reducing snoring severity and microarousals, with favorable trends for reducing SDB severity in selected individuals. Additional larger prospective studies are required to identify suitable candidates for TSD use in the treatment of snoring and SDB.

KEYWORDS: Oral appliances, tongue retainers, sleep-disordered breathing, snoring

Sleep and Breathing, volume 6, number 2, 2002. Address for correspondence and reprint requests: Christopher J. Robertson, M.D.S., Department of Oral Sciences and Orthodontics, School of Dentistry, University of Otago, P.O. Box 647, Dunedin, New Zealand. E-mail: c.robertson@clear.net.nz. [1]Respiratory Research Unit, Dunedin School of Medicine and [2]Department of Oral Sciences & Orthodontics, University of Otago, Dunedin, New Zealand. 1520-9512,p;2002,06,02,069,076,ftx,en;sbr00206x.

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Sleep-disordered breathing (SDB) ranges from snoring to severe obstructive sleep apnea (OSA).[1] Continuous positive airway pressure (CPAP) is the current treatment of choice for moderate to severe OSA.[2,3] Mandibular advancement splints have been used as an alternative first-line therapy for the management of SDB, in particular for snoring and mild sleep apnea. These intraoral devices hold the mandible in a forward position, thus potentially increasing upper airway dimensions. [4–6]

Less attention has been focused on a second type of oral appliance devised in the 1980s: tongue-retaining devices (TRDs).[7] These devices contain a plastic bulb into which the anterior part of the tongue is positioned. The bulb is depressed to create a negative suction pressure and hold the tongue in a forward position. TRDs have been shown to significantly reduce the frequency of breathing pauses and improve sleep quality.[4,7,8] The tongue protrusion created by a TRD increases oropharyngeal, hypopharyngeal, and velopharyngeal cross-sectional areas of the upper airway during awake states.[9] TRDs have also been shown to affect genioglossus muscle activity in a different manner in awake sleep apneics compared with controls.[10] Therefore, it is hypothesized that tongue protrusion alters the shape of the upper airway and is important in alleviating impaired upper airway function.

The tongue-stabilizing device (TSD) was developed by Christopher J. Robertson and manufactured in the United States (Great Lakes Orthodontics, Ltd., Tonawanda, New York) (Figs. 1 and 2). Based on the bulbous compartment of the TRD, this novel oral appliance incorporates a narrowed isthmus joined to the anterior bulbous compartment. The tip of the tongue is inserted into the bulbous compartment, which contains vertical external supports to hold the tongue in a forward position by negative pressure. TRDs incorporate some form of occlusal stop or grooving over the dentition and in doing so usually require the taking of dental impressions. In contrast to the TRD, the TSD only extends intraorally to incorporate the incisor teeth or, in edentulous patients, the alveolar ridge. The TSD is a nonadjustable universal device that is available in four

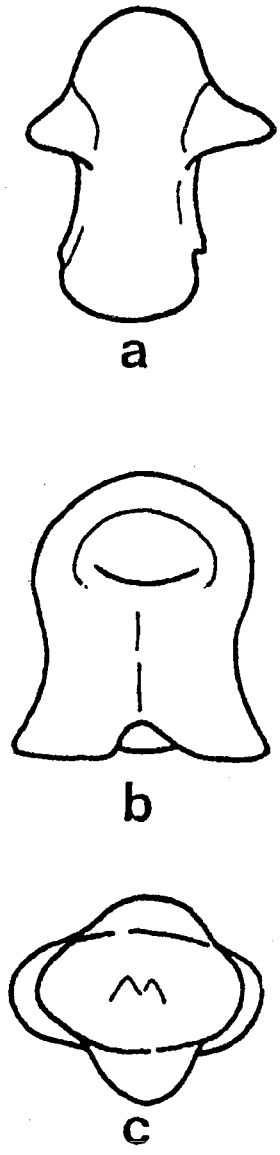

Figure 1 Tongue-stabilizing device: (A) lateral view; (B) superior view; (C) anterior view.

different sizes. In addition, the TSD allows for oral breathing, has no moving parts, and is small and simple to use. As such, it was designed as an inexpensive "off-the-shelf" product for health professionals involved in the treatment of snoring and SDB.

Currently, the TSD is prescribed for the treatment of self- or partner-reported snoring. Treatment success is based on subjective reduction in snoring and daytime dysfunction. The objective effi-

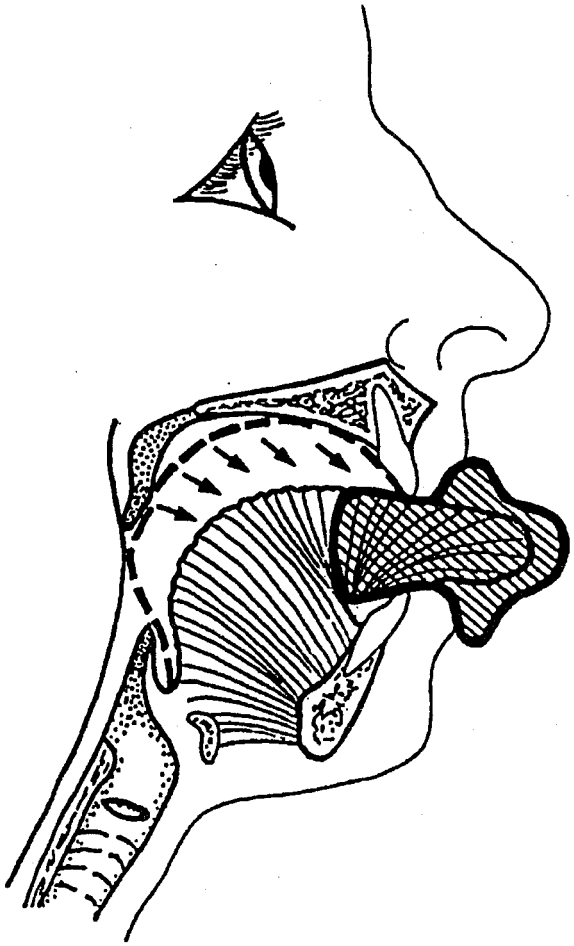

Figure 2 Tongue-stabilizing device in situ.

cacy of the TSD as a therapy for SDB is not known. The aim of the current pilot investigation was to examine the effect of the TSD on polysomnographic variables in patients with mild SDB who were already using the TSD.

METHODS

Patients

Suitable study patients had been medically referred from Sleep or Ear, Nose, and Throat Clinics to the University of Otago Orthodontic Outpatient Clinic for the treatment of snoring and had been fitted with TSDs. Inclusion criteria were as follows: current TSD users (> 3 h/night, self-reported), use of TSD for more than 2 months, and willingness to stop using TSD for one night. Exclusion criteria were self-reported symptoms of OSA, which would constitute a referral to the Sleep Laboratory for polysomnography; taking medication known to affect muscle activity; previous upper airway surgery; and ongoing treatment for SDB.

Eight patients had been issued with the TSD and fulfilled the study criteria. Six male patients (mean age: 51 years ± 4 SD; mean body mass index: 30 kg/m^2 ± 3) agreed to participate. The remaining two patients were unable to take part because of family and work commitments.

The Otago Ethics Committee approved the study, and each study participant provided written informed consent. The TSD has Food and Drug Administration approval for the treatment of snoring (FDA No. K993381).

Study Design

The pilot study was a single-center efficacy trial. Each patient attended the Sleep Laboratory on two consecutive nights: one night with the TSD in situ and one night without the TSD in situ. The order was randomized.

Overnight Polysomnography

On both study nights, patients attended the Sleep Laboratory for full diagnostic polysomnography.[11] Before their arrival, patients were instructed to abstain from caffeinated beverages and alcohol for at least 4 hours. Sleep was monitored by electroencephalography (EEG; C3-A2 and O2-A1), electrooculography, and submental electromyography (EMG). Thoracic and abdominal respiratory movements were measured by inductance plethysmography and arterial oxygen saturation using pulse oximetry. Electrocardiogram, body position, and right and left leg movements were also monitored. In

addition, an integrated sound meter (NL-05, Rion Co., Ltd., Tokyo, Japan) monitored sound levels. The calibrated sound meter was situated at the side of the bed; the microphone was located 4 cm out from the wall at the head of the bed and 20 cm above the pillow. All signals were recorded onto a computerized system (Compumedics S, Victoria, Australia) using a 16-channel polygraph configuration. On the study night with the TSD in situ, patients were instructed to use their device all night.

Off-Line Analysis Sleep stages were manually scored using standard Rechtschaffen and Kales scoring guidelines.[12] Sleep stage values were expressed as a percentage of sleep period time. An apnea episode was defined as a complete cessation of airflow for a minimum of 10 seconds and hypopnea as a 50% reduction in thoracoabdominal movement for a minimum of 10 seconds.[13] The total number of respiratory events was divided by total sleep time to give the apnea–hypopnea index (AHI) per hours slept. Microarousals were scored using the definition of a return to alpha or theta waves on the EEG for a minimum of 3 seconds during non-rapid eye movement sleep, with the addition of a concurrent minimum 3-second rise in submental EMG tone during rapid eye movement sleep.[14] Both spontaneous and respiratory event-related arousals were included in the microarousal frequency. Oxygen desaturations of 4% or more of baseline were calculated from each overnight study using an automatic desaturation detection algorithm (Compumedics S) and divided by total sleep time to give a desaturation index per hour slept. The snore parameters were as follows: The background baseline value was set at 40 dB in each bedroom, a minimum deviation of 5dB from the sound baseline was required before a snore was detected, and the minimum time between snores was 1 second. Each peak snoring sound was then automatically counted and placed into a range of decibel bins using an automated program (Compumedics S) and divided by total sleep time to give a snore index for each decibel range. Each record was anonymous so that the polysomnographer was unaware of whether the TSD was in situ or not.

Statistical Analyses

Paired data were analyzed using mixed two-way analysis of variance for repeated measures, with treatment as a within-subject factor and treatment order as a between-subject factor. Order effects were seen for percentage of Stage 2 sleep and percentage of slow wave sleep. These data were, therefore, analyzed as suggested by Hills and Armitage[15] using an unpaired *t* test on first-limb data only. Snore indexes in the ranges of 51 to 60 dB and 61 to 70 dB displayed substantial heterogeneity of variance and were compared using Wilcoxon rank-sum tests for paired differences. A probability value of less than 0.05 was accepted as statistically significant. All data were analyzed using SPSS version 10 for Windows.[16]

RESULTS

Participants

All participants reported wearing their TSD for the complete duration of the study night.

Efficacy Measures

Snoring Use of the TSD significantly decreased the snore frequency in the 61- to 70-dB range ($P = 0.046$). However, no significant improvements in snoring levels were seen in the other decibel ranges or in the overall frequency of snores per hour slept with the TSD in situ (Table 1; Fig. 3).

AHI A nonsignificant trend was seen for a reduction in AHI with the TSD (Table 2; Fig. 4). The mean reduction in the AHI with the TSD in situ was 11/h slept ± 10 SD.

Table 1 Treatment Differences in Sound Levels (N = 6)

Snoring Index	Without TSD†	With TSD†	P
45–50 dB	90 ± 52	115 ± 4	0.16
51–60 dB	196 ± 208	100 ± 55	0.17
61–70 dB	41 ± 52	8 ± 16	0.046
>70 dB	1.3 ± 3	0.7 ± 1	0.35
Total snore peaks	329 ± 282	223 ± 106	0.31

TSD, tongue-stabilizing device.
†Values represent mean ± standard deviation.

Oxygen Desaturations There was a nonsignificant trend for a reduction in the frequency of oxygen desaturations of 4% or more when the TSD was in situ (see Table 2).

Arousal Frequency A significant decrease was noted in the arousal frequency with the TSD in situ (see Table 2; Fig. 5). The mean reduction in the arousal frequency with the TSD in situ was 12/h slept ± 5 SD.

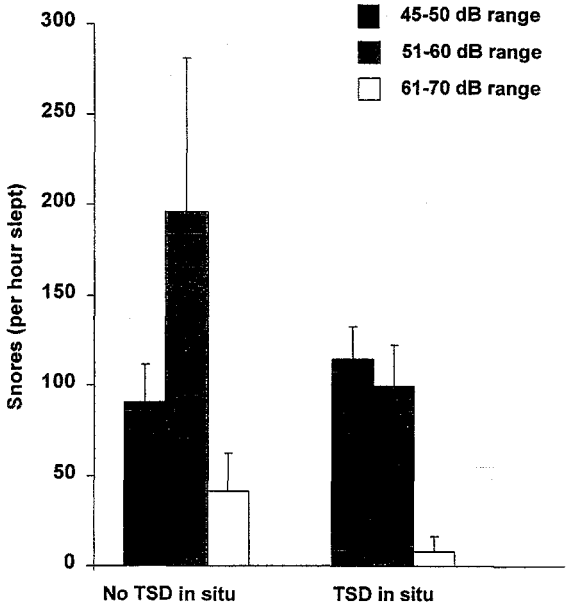

Figure 3 Comparison of snoring levels with and without the tongue-stabilizing device in situ.

Sleep Stages Use of the TSD significantly decreased the percentage of Stage 1 sleep (P = 0.03); however, the TSD had no significant effect on any of the other sleep stages, including stage wake (see Table 2). Total sleep time was not significantly different (P = 0.7) between the two study nights.

Treatment Outcome Of the six participants, three were recommended by the respiratory and sleep physician (D. Robin Taylor) to continue using the TSD as first-line treatment for SDB or snoring. The three remaining patients were recommended CPAP therapy, a mandibular advancement splint, and conservative therapy (alcohol avoidance and sleep position training), respectively, as first-line therapy.

DISCUSSION

This study examined the effect of a novel TSD on polysomnographic variables in the treatment of SDB. In this pilot study, the TSD significantly reduced snoring and sleep fragmentation. The TSD did not significantly lower the AHI or frequency of oxygen desaturations of 4% or more, although trends were found. Findings indicate that the TSD may be an effective therapy in selected individuals with SDB and snoring. Further study is required to determine whether these statistically significant improvements correspond with any clinical benefit and which factors can predict TSD treatment success.

A significant reduction in snoring in the 61- to 70-dB range was found in the current study, in agreement with subjective snoring reports from a previous study using a TRD.[17] Significant reductions in objective snoring sounds have been demonstrated in an efficacy study using a mandibular advancement splint.[18] The TSD was not as effective as the mandibular advancement splint in reducing snoring in our population sample; no significant changes in the other decibel ranges were seen. One possible explanation for the lack of a greater reduction in snoring

Table 2 Treatment Differences in Nocturnal Sleep and Respiratory Variables (N = 6)

Variable	Without TSD*	With TSD*	P
Apnea-hypopnea index†	26 ± 17	15 ± 13	0.06
≥4% oxygen desaturations†	10 ± 10	5 ± 5	0.09
Arousal frequency†	34 ± 16	22 ± 14	0.004
Total sleep time (minutes)	400 ± 55	410 ± 70	0.72
% Awake	15 ± 6	14 ± 5	0.32
% Stage 1 sleep	10 ± 3	8 ± 2	0.03
% Stage 2 sleep	48 ± 4	43 ± 4	0.15
% Slow wave sleep	11 ± 1	15 ± 5	0.17
% REM sleep	18 ± 6	20 ± 5	0.16

TSD, tongue-stabilizing device; REM, rapid eye movement.
*Values represent mean ± standard deviation.
†Per hour slept.

sounds in our study is that patients only snored intermittently; the snoring was frequently interspersed with hypopneas and occasional apneas. It is possible that the TSD reduced the AHI and replaced the silent airway obstruction with partial airway narrowing, thus increasing snoring sound.

Early studies demonstrated significant reductions in the apnea index with a TRD but were performed before the recognition of hypopneas.[7,19] A later study using a TRD did find significant improvements in AHI with a TRD in situ.[8] The current study demonstrated similar improvements in AHI with the TSD, although significance in our smaller sample was not reached. Cartwright[19] also found that patients with a higher AHI in the supine position responded well to a TRD compared with those with no positional component. Although body position was measured in the current study,

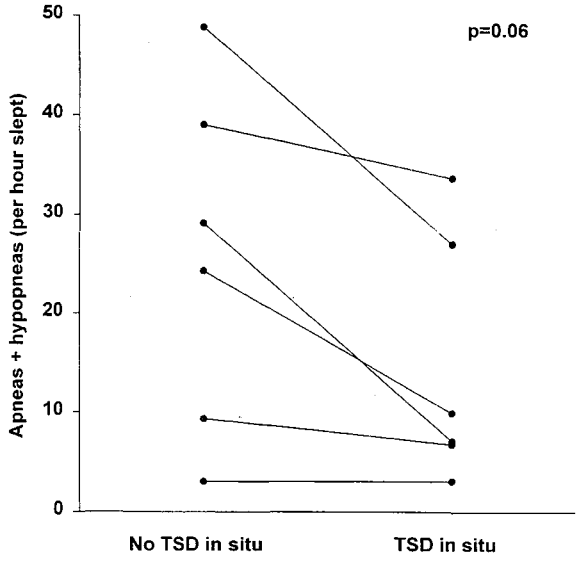

Figure 4 A comparison of the apnea–hypopnea frequency with and without the tongue-stabilizing device in situ.

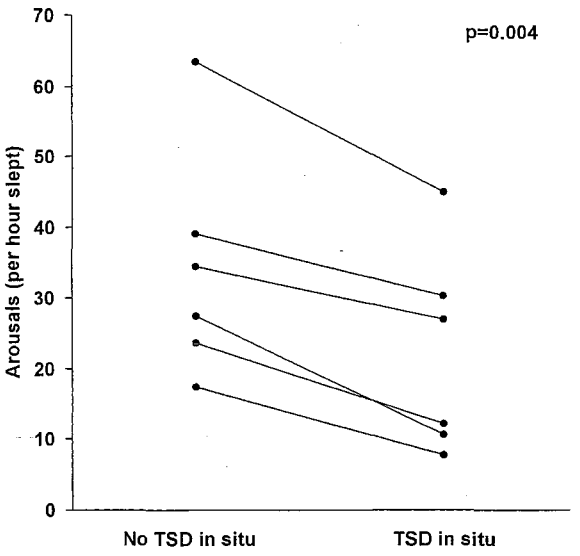

Figure 5 A comparison of the nocturnal arousal frequency with and without the tongue-stabilizing device in situ.

because of the small sample size, subdividing patients into positional and nonpositional SDB was not undertaken. Cartwright[19] also found a significant improvement in the minimum oxygen saturation with a TRD in place. A trend for a reduction in the frequency of oxygen desaturations of 4% or more with the TSD was found in the current study. The greater respiratory treatment success of a TRD could be attributable to the larger sample sizes and greater disease severity studied by Cartwright and Samelson.[7,19] However, it is worth noting that, although TRD studies have demonstrated significant reductions in apnea frequency, the on-treatment apnea indexes remain clinically high for these two studies 22.7 and 32.9/h slept, respectively).[7,19]

The current study found a significant improvement in the arousal frequency with the TSD. Early efficacy studies[7,19] of a TRD were performed before the routine scoring of microarousals; therefore, direct study comparisons cannot be made. However, in agreement with TRD studies,[7,19] the TSD significantly reduced the percentage of Stage 1 sleep. These results indicate that the TSD may reduce the sleep fragmentation associated with SDB.

Limitations to the current study include study power and study design. First, this is a pilot study with a limited sample size. The trends might have been statistically significant with a larger sample size. (Based on our data, if the effect we measured is the true effect, then 23 individuals would be required to detect a significant reduction in AHI and arousal frequency [at $P < 0.05$] with a power of 80%.) Second, the study design has its limitations. Participants were all current users of the TSD who stopped using their devices for one night of polysomnography for the study. This night acted as a "diagnostic" nontreatment night. However, the TSD had been worn consistently for at least 2 months before. Therefore, treatment carryover effects of prior TSD use may have reduced potentially significant differences with and without the TSD in situ. One night without treatment is unlikely to be sufficient to allow the return of upper airway edema. Third, the patients recruited were highly selected; thus, patient acceptability and subjective outcomes were not assessed in the current pilot. Fourth, the sample studied had a wide range of SDB severity. All were self-reported snorers without symptoms of sleep apnea. Yet results demonstrated that four of the six participants had a diagnostic AHI of more than 20/h slept. This highlights the importance of validated questionnaires, screening tools, or prior polysomnography to rule out moderate to severe OSA before fitting an orthodontic device. However, the primary purpose of this pilot was to assess the effect of the TSD on polysomnographic variables rather than predicting disease severity in nonsymptomatic snorers.

The results of this pilot study indicate that the TSD may be an effective therapy in selected individuals with snoring and SDB. Further work on the efficacy and acceptability of the device is required to identify suitable candidates for this simple form of treatment. In addition, the role of the TSD as an alternative, adjunct, or temporary therapy for snoring and SDB needs to be determined.

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FINANCIAL DISCLOSURE

Christopher J. Robertson developed the TSD and has a small commercial interest.

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ORIGINAL ARTICLE

The Efficacy of a Mandibular Advancement Splint in Relation to Cephalometric Variables

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ABSTRACT

The efficacy of a titratable mandibular advancement splint (MAS) for the management of obstructive sleep apnea (OSA) was investigated in relation to supine cephalometric variables. Fourteen adults with diagnosed OSA were recruited following an initial polysomnogram. Supine cephalographic radiographs were taken at baseline and subjects wore the MAS nightly for 6 to 8 weeks. The polysomnogram and cephalogram were repeated with the MAS at maximal titration. The MAS resulted in complete or partial treatment response in all subjects as measured by the improvement in mean apnea/hypopnea index (AHI) (baseline AHI 34 ± 22/hr, with MAS 10 ± 5/hr; $p = 0.001$). The perpendicular distance between the hyoid bone and the mandibular plane (HYML) measured in awake subjects decreased with the MAS (baseline HYML 25.3 ± 7.8 mm, with MAS 16.5 ± 9.6 mm; $p = 0.002$). Baseline HYML was the only cephalometric variable associated with a successful clinical outcome. It was strongly linked to improvements in AHI (adjusted $R^2 = 0.37$, $p = 0.012$) and arousals (adjusted $R^2 = 0.455$, $p = 0.005$). We conclude that the MAS is an effective therapy for OSA and baseline HYML is an important predictor of improvement. Improvements in AHI may be explained by the MAS maintaining the new or existing relationship of the hyoid and its surrounding structures, thus preventing obstruction in the upper airway during sleep.

KEYWORDS: Obstructive sleep apnea, cephalometry, mandibular splint

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Obstructive sleep apnea (OSA) includes repetitive periods of partial or complete airway obstruction during sleep and daytime symptoms such as hypersomnolence and decreased vigilance.[1] A wide spectrum of anthropometric, postural, pathophysiologic, and cervicocraniofacial features may contribute to OSA, some of which may be related to ethnic groupings.[2–7] Members of the Polynesian population, for example, have greater neck circumferences and more prognathic mandibles than Caucasians.[4] Continuous positive airway pressure (CPAP),[8] which provides a pneumatic splint for the upper airway, is the standard therapy and treatment of choice in moderate to severe OSA. However, the efficacy of oral appliances, used predominantly in the conservative management of mild to moderate OSA, has also been confirmed as an appropriate alternative treatment.[9–15]

Despite considerable variation in appliance design, the main objective of oral appliances is to draw the mandible forward and alter the dynamics of the upper airway by changing relationships between the pharyngeal space, hyoid bone, and tongue position.[10–18] The clinical benefits are remarkably consistent and approximately half of treated patients achieve an apnea hypopnea index (AHI) of fewer than 10 per hour slept (AHI < 10/hr)[15,19] with a device in situ.

Recently, new design features have been introduced in some oral devices in an attempt to reduce side effects such as discomfort from downward rotation of the mandible and a fixed jaw position. Oral splints, which enable the patient to gradually adjust the amount of anterior positioning of the mandible to achieve optimal position, have improved the biomechanical relationships between the mandible and the surrounding bony and soft-tissue structures.[14] Despite this, the optimal design and an objective means of predicting the therapeutic response to oral appliances have not been clarified.

The hyoid bone and its surrounding musculature play a key role in regulation of the pharyngeal airway, and a low position of the hyoid relative to the mandibular plane is a distinguishing feature of OSA.[20,21] In the upright posture, the cross-sectional area of the hypopharynx is smaller in OSA patients compared to snorers or normal subjects.[22,23] In the supine posture, awake OSA subjects maintain pharyngeal muscle tone in order to prevent the upper airway from collapsing further due to increased gravitational load on the tongue.[23] However, during sleep, loss of airway tone increases the risk of airway closure.

Investigators have used lateral cephalometry, fluoroscopy, and videoendoscopy to evaluate the effect of oral appliances on the upper airway dimensions of awake OSA subjects.[24–26] Anterosuperior elevation of the hyoid has been shown to occur with mandibular advancement.[17,25,27,28] However, studies have not standardized supine cephalometric measures using reliable bony landmarks, and in only one study using an oral device[26] have supine cephalometric measures been correlated with clinical outcomes. Perhaps for these reasons the role of cephalometry in predicting responses to treatment has not been fully evaluated.

The present study was designed to confirm the efficacy of a novel titratable mandibular advancement splint (MAS) in the management of OSA and to evaluate objective improvements in OSA in relation to supine cephalometric variables.

METHODS

Study Population

Sixteen subjects medically referred for suspected OSA underwent full-night polysomnography (PSG) at the Tom McKendrick Sleep Laboratory, Dunedin Hospital. The subjects were diagnosed with OSA and agreed to participate in the initial screening for the study.

Inclusion criteria: Subjects with mild to moderate sleep apnea (AHI 10 to 40/hr slept); subjects with moderate to severe OSA (AHI 30 to 80/hr) who were noncompliant with a trial of nasal continuous positive airway pressure (nCPAP).

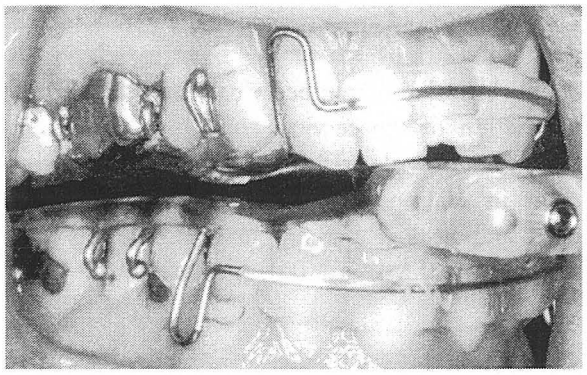

Figure 1 Titratable mandibular advancement splint (MAS) in situ.

Exclusion criteria: Subjects who were edentulous or had insufficient teeth on the maxillary and mandibular arches to support the MAS; had a medical history including severe cardiovascular, psychological, or neurological disorders affecting sleep; or had other coexisting sleep disorders.

Oral Appliance

The MAS (Fig. 1), made from a dental impression, was a modified Thornton anterior positioner appliance (TAP, Oral Appliance Technologies, Dallas, TX) of methyl methacrylic material with full upper and lower occlusal coverage. A stainless steel screw, hook, and bar arrangement connected the upper to the lower portion while still allowing for a small amount of lateral jaw movement and oral airflow. Clockwise turns of the screw enabled the mandibular section of the MAS to be titrated forward approximately 2 mm with each half turn to a maximum of 16 mm.

Subjects were instructed on how to fit and titrate the splint by rotating the screw clockwise, one half turn every 1 to 2 nights as tolerated. Subjects were also instructed in active exercises for the temporomandibular joint to assist with realignment of the bite on removal of the MAS in the mornings. The MAS was worn nightly for 6 to 8 weeks. End points for titration were partner-reported cessation of snoring and subjective improvement in daytime symptoms or maximal titration tolerance with subject-reported symptom improvement. Subjects were reviewed at the Orthodontic Clinic at least once during the trial period. They were also followed up by telephone during the trial to monitor for side effects or difficulties with the splint and seen again at the Orthodontic Clinic if further checks of the splint were deemed necessary. A telephone follow-up at 1 year was undertaken to ascertain the long-term compliance with the therapy and potential side effects.

Outcome Measures

Sleep habits, snore symptoms, exercise capacity, general health, and medications at presentation were recorded on a standardized questionnaire. The Epworth Sleepiness Scale (ESS) was used to measure the subject's daytime sleepiness,[29] and body height, weight, and neck circumference (at the fourth cervical vertebra) were measured at baseline and at the end of the trial. A self-reporting diary was used during treatment to record sleep habits, the period of time wearing the device at night, levels of sleepiness/alertness on waking, daytime hypersomnolence, and any other relevant symptoms.

Polysomnography

Each subject underwent full-night PSG prior to entering the study and had a second PSG with the splint in situ at the end of the study period. Data were recorded on a 16-channel Compumedics polysomnographic system (Compumedics, Victoria, Australia). OSA scoring criteria used were in accordance with standard definitions.[1] Apnea was defined as cessation of airflow lasting at least 10 seconds. Hypopnea was defined as a reduction in thoracoabdominal wall movement of greater than 50% of the baseline measurement for more than 10

seconds or as a reduction in thoracoabdominal movement with an accompanying oxygen desaturation of at least 3% and/or associated with arousal. Sleep was scored according to Rechtschaffen and Kales' guidelines.[30] Arousals were defined as a shift in electroencephalogram (EEG) frequency for a minimum of 3 seconds with a concurrent rise in electromyogram (EMG) in REM sleep.[31] All studies were manually scored in 30-second epochs for sleep stage, apnea type, and duration and arousals, by one scorer blinded to the study. These results were confirmed by a second independent scorer.

Treatment Outcomes

Treatment success was defined as an AHI less than or equal to 10/hr and resolution of symptoms; partial success as an AHI in the range of 10 to 15/hr, with improvement in symptoms; and treatment failure as an inability of the patient to continue to use the MAS.

Cephalometric Radiographs

Cephalograms were taken at baseline with each subject lying supine in the natural head position[32] using the orthoposition method.[33] A fluid level was used to register the head posture and care was taken to limit changes in cervical posture during the introduction of the cephalostats. A chain suspended from the film cassette registered the true vertical plane. Subjects were instructed to hold the teeth in occlusion and cephalograms were taken at the end of an expiration. The procedure was repeated for the cephalograms taken at the end of the self-titration period with the MAS in situ.

All lateral cephalometric landmarks were coordinated with true horizontal and vertical lines. Conventional bony landmarks (Fig. 2A) were marked on tracing film and digitized with a reflex metrograph. The landmarks and measures (Fig. 2B) were used to analyze pharyngeal relationships; postural relationships of the head in relation to craniovertical and craniocervical variables, and the inclination of the cervical column in relation to cervicohorizontal variables; and the hyoid bone position in relation to cephalometric variables of the head and neck. Reference lines and angles were measured to the nearest 0.01 mm or degree.

Statistical Analysis

Statistical analysis was carried out using the computer statistical package SPSS for Windows (SPSS Inc., Release 10.0). Variables were normally distributed. Paired t-tests were undertaken on all variables for the polysomnographic and cephalometric data. Stepwise linear regression analysis was undertaken to examine the relationships between changes in AHI and arousals and significant cephalometric variables including pharyngeal relationships, postural relationships of the head and neck, and relationships of the hyoid bone to the head and neck (Fig. 2). Because multiple comparisons were made, a significance level of 0.01 was used in the statistical analysis.

The study protocol was approved by the Otago Ethics Committee. All subjects gave written informed consent.

RESULTS

Study Population

Fourteen males and one female were enrolled in the study. One male subject failed to return for appointments, leaving 14 subjects who completed the study. New Zealand ethnic groupings included European (12), Maori (1), and Chinese (1). Other demographic and anthropometric variables at study entry are shown in Table 1. There was a significant relationship between body mass index (BMI) and

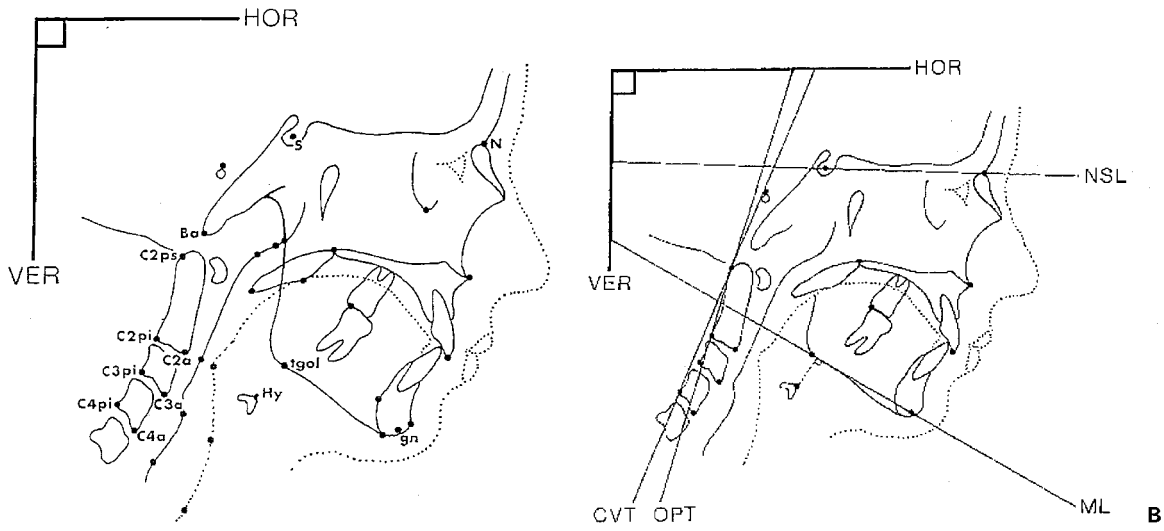

Figure 2 (A) Key landmarks identified on cephalometric radiographs and digitized with a reflex metrograph. Hy: the most anterior and superior point on the body of the hyoid bone; gn: the most anteroinferior point on the bony mandibular symphysis; tgol: the most lateral external point at the junction of the horizontal and ascending rami of the mandible; N: nasion, the most anterior point on the frontonasal suture; S: center of sella turcica, the center of the pituitary fossa of the sphenoid bone; Ba: basion, the most posteroinferior point on the anterior margin of the foramen magnum; c2a, c3a, c4a: most anteroinferior point on the corpus of the second, third, and fourth vertebral bodies, respectively; c2pi, c3pi, c4pi: most posteroinferior point on the corpus of the second, third, and fourth vertebral bodies. respectively; c2ps: most posterosuperior point on the corpus of the second cervical vertebra. (B) Key reference lines and angles measured on the cephalometric radiographs. HYML: the perpendicular distance from hyoid to mandibular plane (distance measured on the perpendicular line drawn from point Hy to the intersection of a line drawn between points gn and tgol). NSL: anterior cranial base line connecting the center of the sella turcica S and nasion N. Postural relationships of the head and craniovertical and craniocervical variables, and the inclination of the cervical column in relation to craniohorizontal variables. CVT: cervical vertebral tangent, posterior tangent on the odontoid process c2ps through c4pi; OPT: second cervical vertebral tangent on the odontoid process c2ps through c2pi. HYML was the only cephalometric measure to change significantly ($p = 0.001$) with the mandibular advancement splint (MAS) in situ; Hy was repositioned in an anterosuperior direction with the MAS in situ, thus decreasing the mean HYML distance.

neck circumference ($R^2 = 0.614$, $p = 0.001$). No significant change in body weight or neck circumference occurred during the study period.

Clinical Outcomes

The MAS resulted in complete or partial treatment response in all 14 subjects (Fig. 3). Treatment success (AHI < 10/hr) with the splint was achieved in 50% of the subjects (n = 7). The remaining subjects had AHIs in the sub-optimal range with improvement in symptoms. Four subjects (29%) had AHIs greater than 10 and less than 15/hr and for the remaining three subjects (21%), AHIs of 15.2/hr, 15.8/hr, and 15.9/hr were recorded.

Results of paired t-tests for polysomnographic data are presented in Table 2. Highly significant improvements were recorded for mean AHI

Table 1 Demographic Details of Study Participants

Measurement	Mean (SD) (n = 14)	Range
Age (years)	47.6 (10.9)	25–63
Body Mass Index ($kg.m^{-2}$)	29.3 (4.6)	18.5–35.6
Neck circumference (cm)	42.1 (4.6)	31.0–48.5

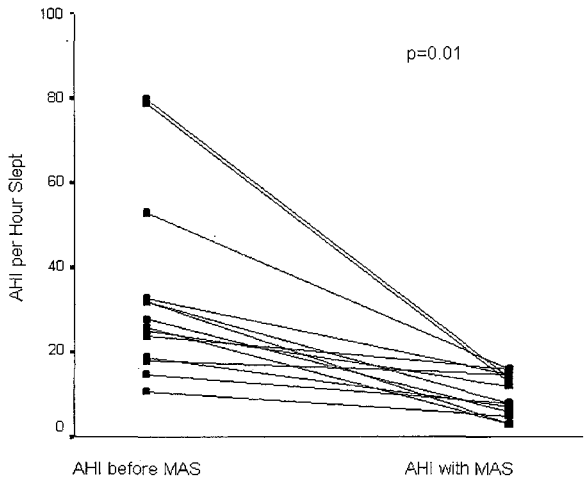

Figure 3 Changes in apnea hypopnea index (AHI) for individual subjects (n = 14) before and with the MAS.

(p = 0.001) (Fig. 3) and mean arousal frequency with the MAS. There was a significant improvement in the minimum oxygen desaturation with the MAS (p = 0.01). The proportion of time spent in REM sleep increased with the MAS, but there were no other significant changes in sleep architecture.

Cephalometric Outcomes

A comprehensive range of variables was measured from the supine cephalometric films taken before and with the MAS (n = 11), but for reasons of brevity only the significant results are reported. All cephalometric measures for the Maori and Chinese subjects fell within the mean ± SD for the European subjects. The only significant difference in any distances or angles measured for the hyoid bone position in relation to head and neck posture was a significant reduction in the perpendicular distance from the hyoid bone to the mandibular plane (HYML): without MAS, 25.3 ± 7.8 mm, with MAS 16.5 ± 9.6 mm; p = 0.002 (Fig. 2(B)). No significant differences were identified for pharyngeal relationships or for postural relationships of the head and craniovertical and craniocervical variables, or for the inclination of the cervical column in relation to craniohorizontal variables.

Linear regression analysis was used to examine the relationship between the changes in polysomnographic variables and supine cephalometric measurements at baseline. The decreases in both AHI and arousal index with the MAS were significantly related to the baseline HYML (adjusted $R^2 = 0.37$; p = 0.012, and adjusted $R^2 = 0.455$; p = 0.005, respectively). No other cephalometric measures were associated with improvement in polysomnographic variables.

Subjective Outcomes

The MAS was well tolerated by all subjects throughout the study period (Table 3). Thirteen subjects (93%) reported nightly use. The mean

Table 2 Mean (SD) and Range of Values for Polysomnographic Data Recorded Before and with the Mandibular Advancement Splint (MAS) in situ

	Before MAS	With MAS	p Value
Apneas + Hypopneas/hr slept	34 ± 22 (11–79)	10 ± 5 (3–16)	0.001
Arousal frequency/hr slept	37 ± 20 (15–77)	19 ± 7 (10–30)	0.001
% Minimum oxygen saturation	76 ± 6 (74–81)	82 ± 4 (80–85)	0.012
Total sleep time (min)	391 ± 64 (354–427)	389 ± 71 348–429	0.905
Sleep efficiency	81 ± 5 (70–89)	82 ± 9 (28–64)	0.75
% Stage 1*	9 ± 4 (3–18)	7 ± 2 (2–11)	0.09
% Stage 2*	59 ± 10 (46–83)	54 ± 8 (39–71)	0.09
% Slow wave sleep*	14 ± 7 (0–24)	18 ± 4 (12–25)	0.1
% REM*	17 ± 6 (5–24)	22 ± 7 (5–34)	0.03

*Sleep stage was calculated as a percentage (%) of total sleep time.

Table 3 Subjective Reporting by Patients at the Conclusion of the Trial

Question	Response (%)
Wore the MAS nightly for 5 or more hours	93%
Decreased snoring (partner report)	93%
Improved sleep quality	79%
Woke refreshed	50% always 50% sometimes
Improved well-being	79%
Increased saliva	7%
MAS loose/fell off	21% sometimes
Short-term tooth discomfort	28%
Temperomandibular joint pain	0%

nightly use was 5.5 ± 1.9/hr. Daytime sleepiness as measured by the ESS significantly improved with the MAS (without MAS, ESS 12 ± 5; with MAS, ESS 6 ± 4; $p = 0.0001$). An improved sense of well-being was reported by 93% of subjects. None of the reported side effects precluded the subjects from wearing the splint.

Subjects were contacted 1 year after the completion of the trial. Eight subjects (57%) reported that they were continuing to use the splint on a nightly basis with good effect. One subject was wearing the splint "sometimes". Two subjects were wearing alternative, less bulky oral appliances; one had chosen to use nCPAP; one had chosen not to use any device; and one was having other medical therapy for an unrelated illness.

DISCUSSION

Our study confirms that a titratable MAS is an effective therapy in the management of OSA over a broad range of clinical severity (AHI 11 to 79/hr) and that efficacy is associated with specific changes in cephalometric variables. There were significant reductions in AHI and arousal index, and in 50% of subjects, the AHI improved to fewer than 10/hr. For the remaining subjects, the AHI was in the suboptimal range (> 10 < 16/hr). These outcomes compare favorably with the results obtained in other studies using oral appliances.[10,15,26,34]

Our results demonstrated that with the MAS in situ, subjects were able to obtain significant anterosuperior elevation of the hyoid bone (baseline HYML 25.3 ± 7.8 mm; with MAS 16.5 ± 9.6 mm; $p = 0.002$), thus achieving a more normal hyoid position. This suggests that significant improvements in AHI during sleep were achieved either by establishing a new anterosuperior HYML relationship or by maintaining the existing hypopharyngeal dimensions as measured while awake. Preventing the mandible from rotating posteroinferiorly during sleep, with subsequent hypopharyngeal occlusion, may be the principal mechanism of action for the MAS.[25]

Our study was also designed to evaluate whether any cephalometric measurements, obtained awake in the supine posture, might be used to assess likely clinical outcomes. The measures and angles taken from the cephalograms were in agreement with the findings in earlier studies of head form and postural angles in Caucasians with OSA.[4] However, the baseline HYML was the only cephalometric variable associated with PSG improvements, namely change in AHI (adjusted $R^2 = 0.37$; $p = 0.012$) and arousal index (adjusted $R^2 = 0.455$; $p = 0.005$). There were no significant relationships between any other cephalometric or anthropometric variables and treatment outcomes.

The results of previous investigations have been varied in this regard. In the study by Ferguson et al[35] no indicators of likely clinical success were identified in their study of 24 OSA subjects who used a mandibular advancement device. In an earlier study, the same authors reported that although neck circumference correlated with AHI, this was not true for any craniofacial or soft tissue measurement. Interestingly, they observed that an increased HYML distance was associated with a larger neck circumference ($p < 0.05$). In the study by Battagel et al,[28] of 58 OSA subjects, no cephalometric fea-

tures which could predict an improvement in AHI were identified. Similarly, in the study by Liu et al,[26] in which an adjustable appliance was assessed in 16 adult OSA patients, although a significant decrease in mean HYML was found with the appliance (before 22.7 ± 5.3 mm; with 19.7 ± 4.8 mm; $p = 0.05$), the reported results did not indicate the strength of any relationship between these changes and treatment response in terms of AHI.

The contrast between the present results and those obtained in previous studies may be due to methodological inconsistencies. The reproducibility of cephalometric measures in the natural head position while standing has previously been validated[33] but hitherto this has not been the case for measurements obtained in supine. The latter is likely to be more clinically relevant in OSA. One supine study reported by Hiyama et al[36] was on normal subjects and although there was no significant difference in the relationships reported it focused on the mandibular posture and did not include craniocervical angles and relationships. In the present study, subjects lay supine with their heads held in the natural head position, cephalostat rods were then placed carefully, a fluid level was used to check head position, and reliable bony landmarks were used for cephalometric measures. Using this standardized approach, reproducibility for cephalometry in the supine position was confirmed: there were no significant differences in the craniovertical and craniocervical variables, or in the inclination of the cervical column in relation to cervicohorizontal variables, between the films before and with MAS. The only significant difference was in the hyoid-mandibular relationship. In contrast, Battagel et al[28] reported that cephalometric rods could not be adequately positioned, and in the study by Liu et al,[26] subjects were asked to mimic their usual position rather than adopt a standard posture. The Frankfort Horizontal Plane, which is not a true horizontal plane, was used by Ryan et al[25] to fix the head position in order to measure upper airway cross-sectional area by videoendoscopy. However, it is not clear whether any of the measurements made by Battagel et al[28] and Ryan et al[25] in their studies were reproducible.

Our study had a number of shortcomings. First, titration of the splint was based on subjective rather than objective responses. This was considered appropriate so that patients might adjust gradually to wearing the splint. Given that significant improvement in AHI was obtained in the majority of patients, this appears to have been a justified approach. Second, the sample size in our study was small, and it was not possible to derive predictive values for changes in AHI based on changes in HYML. Nevertheless, by using a carefully standardized approach, we were able to confirm a strong association between HYML and treatment outcomes. Third, the sample was selected in that it included patients who had failed CPAP therapy, but such subjects would normally be offered an alternative therapy, such as a trial of a mandibular splint, in the clinical setting.

Compliance with treatment for OSA is a major clinical issue. Data regarding long-term compliance with the MAS are limited. In our study, a telephone survey 1 year after commencing treatment revealed that 57% continued to obtain therapeutic benefit from the MAS with improved quality of sleep and daytime function. Pancer et al[18] reported 86% of subjects had continued to use a mandibular appliance when followed up an average of 1 year later; Clark et al[37] reported that 50% of subjects in their study were continuing to use a mandibular device 3 years after commencing treatment.

In conclusion, our study has demonstrated that in patients with OSA, the treatment response to a titratable MAS is most likely to occur in subjects with an increased HYML. Although there was a wide individual variation in the extent or degree of anterosuperior hyoid repositioning with the MAS, it is postulated that either the reduction in HYML or the maintenance of an adequate hyoid position while asleep is the mechanism whereby upper airway patency is achieved. Supine cephalometry is therefore an important tool in determining relationships between craniofacial bony and soft-tissue structures and the likely outcomes of treatment for OSA. When adequately standardized, cephalograms provide clinically helpful imaging in

patients for whom mandibular advancement is being considered as a treatment option.

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Dental and occlusal changes during mandibular advancement splint therapy in sleep disordered patients

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SUMMARY The aims of this longitudinal, observational study were two-fold: first, to determine in adults with sleep disorders the extent of dental and occlusal changes following the use of a mandibular advancement splint (MAS) and, second, to determine the time course of these changes.

One hundred adult subjects (87 males, 13 females) diagnosed with obstructive sleep apnoea (OSA) and/or asymptomatic snoring were treated with non-adjustable MAS. At the outset each subject was randomly assigned to a group and reviewed 6, 12, 18, 24 or 30 months after placement of a splint. There were 20 subjects in each group. Craniofacial changes were measured on lateral cephalometric radiographs taken at the initial and review appointments.

When the changes in all subjects were examined, the SNA, ANB angles, ANS–PNS length and face height increased, and the mandibular first molars and the maxillary first premolars significantly over-erupted. Significant retroclination of the maxillary incisors and proclination of the mandibular incisors were accompanied by reductions in maxillary arch length, overbite and overjet. When the changes over time were determined, the mandibular symphysis was significantly lower at all review periods. An increase in face height and reductions in overbite and overjet were evident at 6 months, and over-eruption of the maxillary first premolars and mandibular first molars, and proclination of the lower incisors were found at 24 months. Significant positive correlations were also found between the amount of anterior opening by the appliances and changes in overbite at 24 and 30 months.

The appliance used produced small, unpredictable changes in the occlusion that tended to occur after 24 months' wear. It is postulated that the changes in overbite might be lessened by keeping the bite opening to a minimum.

Introduction

Oral appliances, such as mandibular advancement splints (MAS), are an accepted method for the treatment of mild to moderate obstructive sleep apnoea (OSA) and snoring (American Sleep Disorders Association Standards of Practice Committee, 1995; Schmidt-Nowara, 1999). The aim of these appliances is to enlarge the oropharyngeal airway by repositioning the mandible downwards and forwards.

Recent studies of patients with moderate sleep disorders have reported that long-term wear of MAS was accompanied by small changes in the position of the mandible, reductions in both overbite and overjet, and a forward shift of the mandibular molars (Bondemark, 1999; Pantin *et al.*, 1999; Fritsch *et al.*, 2001; Marklund *et al.*, 2001). Significant dental side-effects in individual cases have also been reported (Panula and Keski-Nisula, 2000; Rose *et al.*, 2001). The molar changes can be questioned because some of the observations were derived from study casts and, consequently, may be confounded by a downward and forward posturing of the mandible. While there may be some disagreement about the site(s) of the dental and occlusal changes due to MAS, there is less information about their onset.

The aims of this cephalometric study were two-fold: first, to determine the extent of dental and occlusal changes following the use of a non-adjustable MAS for the treatment of sleep disorders, and, second, to determine these changes over time.

Subjects and methods

From 114 consecutive adult patients with OSA and/or habitual snoring medically referred to an orthodontic practice for a MAS, 100 subjects (87 men, 13 women) agreed to participate in this study. Of the 114 patients, one subject declined to participate and 13 subjects were excluded because, on questioning, they were found to be wearing the appliance less than 5–6 hours per night. The mean age of the men was 49.0 years (standard deviation 8.3 years) and for the women 51.0 years (standard deviation 10.2 years). Further details of the subjects and methods are given in Robertson (2002).

The appliance used in this study was a non-adjustable rigid splint (Robertson, 1997) which covered the occlusal surfaces of the maxillary and mandibular teeth (Figure 1).

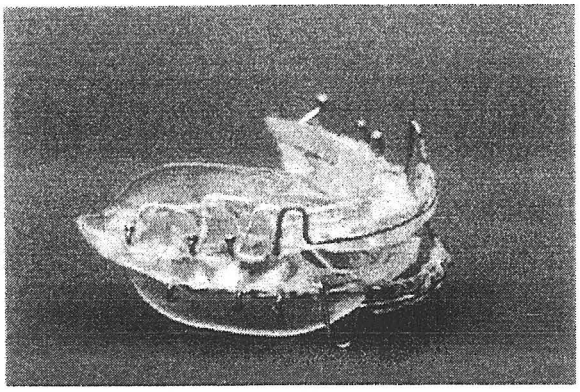

Figure 1 The mandibular advancement splint used in this study.

The splints were constructed to advance the mandible 75 per cent of the maximum protrusion obtained by each subject. In this sample, maximum protrusion ranged from 3 to 14 mm. For appliance construction, mandibular advancement was measured clinically with a George gauge to the nearest millimetre (George, 1992).

Assessment of dental and occlusal changes

At the outset all subjects had two lateral cephalometric radiographs taken: one with the teeth in the intercuspal position and with the head in the so-called natural head position, and a second film with the appliance *in situ*. This latter film was used to confirm the amount of mandibular advancement and opening obtained with each appliance. At this visit each subject was randomly assigned to a group to be reviewed either 6, 12, 18, 24 or 30 months later. There were 20 subjects in each group, with no more than three women in any one group. The subjects received their appliances 1 week later. At the review appointment a third lateral cephalometric radiograph with the teeth in the intercuspal position and the head in the natural head position was obtained. The subjects were seated for all radiographs which were taken in the afternoon by the same operator.

The reference points and planes given in Figure 2 were transferred to mylar film, digitized twice with a reflex metrograph (Scott, 1981), and converted to linear and angular measurements (Table 1). The positions of the first molars, maxillary first premolars, and maxillary and mandibular central incisors, relative to anatomically stable reference lines in the maxilla and mandible, were measured on the initial and review films. The position of the mandible was measured relative to an anatomically stable reference line in the cranial base (Johnston, 1996). The differences between each subject's review and initial film, taken with the teeth in the intercuspal position, were used to determine the skeletal, dental and occlusal changes.

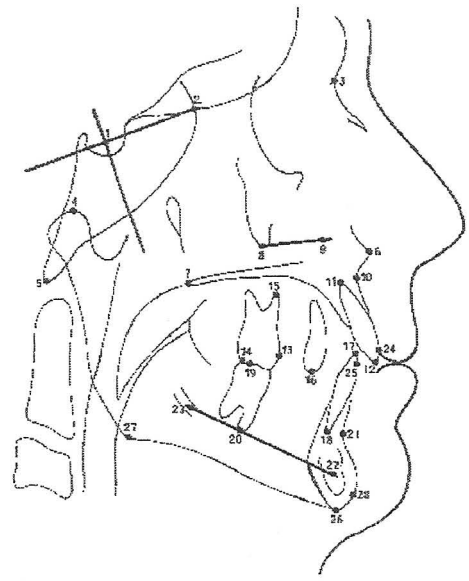

Figure 2 Cephalometric reference points. (1) S, sella; (2) SE, spheno-ethmoidal junction; (3) N, nasion; (4) Cd, condylion; (5) Ba, basion; (6) ANS, anterior nasal spine; (7) PNS, posterior nasal spine; (8) IZ_1, inferior zygoma (1). The lower most point on the average of the right and left outlines of the zygoma; (9) IZ_2, inferior zygoma (2). The constructed point 2 cm rostral to IZ_1 along the line parallel to the floor of the nose through IZ_1; (10) A, point A; (11) UIA, upper central incisor apex; (12) UIE, upper central incisor edge; (13) U6MCpt, upper first molar mesial contact point; (14) U6DCT, upper first molar distal cusp tip; (15) U6MRA, upper first molar mesial root apex; (16) UPMCT, upper first premolar cusp tip; (17) LIE, lower incisor edge; (18) LIA, lower incisor apex; (19) L6DCT, lower first molar distal cusp tip; (20) L6MRA, lower first molar mesial root apex; (21) B, point B; (22) Fid_1, fiducial point 1. A point on a natural structure within the mandibular symphysis; (23) Fid_2, fiducial point 2. A point on a natural structure of the posterior body of the mandible; (24) overbite point. The point of intersection between the perpendicular through UIE and a line drawn parallel to the functional occlusal plane through the point LIE; (25) overjet point. The point of intersection of the line drawn parallel to the functional occlusal plane through UIE and the labial surface of the most prominent mandibular incisor; (26) Me, menton; (27) Go, gonion; (28) Pg, pogonion. Cephalometric measurements and planes: S–SE, cranial base reference line, joining 1 and 2; IZ_1–IZ_2, maxillary reference plane, line joining 8 and 9; Fid_1–Fid_2, mandibular reference plane, line joining 22 and 23; FOP, functional occlusal plane. The average occlusal plane of the buccal teeth including the first molars and premolars. This plane is used to determine overbite and overjet points; Cd vert, the vertical distance between the condylion and an extension of the line S–SE; Cd horiz, the horizontal distance between the condylion to a line perpendicular to the line S–SE through S.

To determine the errors in the radiographic method, 10 randomly selected films were retraced and remeasured by the same operator (CR) and compared with the *t*-test for paired data and the errors calculated using Dahlberg's formula (1940). There were no statistically significant differences between the duplicate measurements. The errors, which fell within the ranges of 0.17–0.50 mm and 0.20–0.95 degrees, are comparable with those reported in a similar study (Coltman *et al.*, 2000). Associations between the amount of mandibular advancement and

Table 1 Changes in sleep disordered subjects (n = 100) during mandibular advancement splint therapy.

	Initial		Review			
Skeletal variables	Mean	SD	Mean	SD	Mean difference	P
SNA	81.38	3.74	81.7	3.53	–0.32	**0.023**
SNB	78.41	3.8	78.42	3.59	–0.01	0.927
ANB	3.22	2.1	3.5	2.09	–0.29	**0.013**
N–Me	130.89	7.81	131.66	7.72	–0.76	**0.0001**
N/ANS–PNS	56.94	3.52	57.05	3.56	–0.11	0.356
Me/ANS–PNS	72.96	5.93	73.62	5.9	–0.66	**0.0001**
S–Go	87.2	7.42	87.7	7.41	–0.5	**0.0001**
S/ANS–PNS	47.99	3.76	47.94	3.84	0.05	0.56
Go/ANS–PNS	38.75	6.24	39.31	6.29	–0.56	**0.001**
ANS–PNS	54.4	4.09	55.36	3.81	–0.95	**0.002**
Ba–PNS	45.1	4.53	45.04	4.18	0.06	0.763
Cd–Pg	125.35	7.19	125.18	7.01	0.17	0.362
Cd–Go	67.04	6.32	66.74	6.15	0.31	0.116
Go–Pg	80.68	5.14	80.6	5.19	0.08	0.606
Mandibular position						
Cd vert	17.61	4.05	18.42	3.86	–0.81	**0.0001**
Cd horiz	14.77	3.58	14.84	3.53	–0.07	0.666
Fid_1 vert	118.46	7.69	119.45	7.69	–0.99	**0.0001**
Fid_1 horiz	39.68	12.63	39.49	12.63	0.19	0.471
Dentoalveolar variables						
U6DCT vert	28.81	3.52	29	3.28	–0.2	0.259
U6DCT horiz	7.29	4.39	7.17	4.44	0.12	0.577
$U6/IZ_1$–IZ_2	67.68	8.4	67.03	8.77	0.65	0.264
L6DCT vert	21.54	2.27	21.91	2.33	–0.37	**0.021**
L6DCT horiz	34.94	4.86	34.86	4.71	0.07	0.668
$L6/Fid_1$–Fid_2	103.28	21.28	103.6	20.74	–0.32	0.852
Pm vert	32.63	3.56	33.02	3.45	–0.39	**0.002**
Pm horiz	11.43	4.37	11.58	4.76	–0.15	0.505
UIE vert	37.41	3.59	37.33	3.73	0.08	0.604
UIE horiz	29.31	5.3	28.95	5.26	0.36	0.153
UI/IZ_1–IZ_2	104.75	8.79	103.14	9.1	1.58	**0.001**
LIE vert	34.05	3.15	34.15	3.37	–0.1	0.398
LIE horiz	7.34	4.88	6.87	4.71	0.47	**0.002**
LI/Fid_1–Fid_2	83.81	10.87	86.53	8.21	–2.71	**0.001**
Overbite	4.09	2.62	3.07	2.1	1.02	**0.0001**
Overjet	4.25	2.23	3.19	1.76	1.06	**0.0001**
MaxAL	37.54	4.68	37.07	4.29	0.47	**0.006**
MandAL	30.48	4.17	30.71	4.19	–0.23	0.23

Significant differences at the 5 per cent level are shown in bold.
SD, standard deviation.

opening measured cephalometrically and the antero-posterior skeletal relationship (ANB angle) at the outset were investigated with Pearson product-moment correlation coefficients. Differences between the groups at the outset were investigated with a one-way ANOVA and Duncan's new multiple range test. Unpaired t-tests were used to test for gender differences at the outset and t-tests for paired data were used to determine if there were any statistically significant differences between the initial (T1) and review (T2, T3, T4, T5, T6) measurements. Associations between the changes in the mandibular molars, overbite and overjet, the maxillary and mandibular incisors, and the amount of mandibular opening and advancement were investigated with Pearson product-moment correlation coefficients.

Mandibular advancement

Initial mandibular advancement was established clinically with the George gauge (George, 1992). The amount of advancement and anterior (opposite the incisors) and posterior (opposite the first molars) vertical opening by the appliances was determined by comparing the measurements taken from the two initial cephalometric radiographs (with and without the appliance *in situ*). Mandibular advancement was measured as the horizontal movement of the lower incisors reference point (LIE), along a line parallel to the functional occlusal plane (FOP). Anterior opening was measured as the vertical distance between the upper and lower incisal edges (UIE–LIE) along a line perpendicular to the FOP. The

FOP was the line passing through points 14 and 16 (Johnston, 1996). The degree of posterior vertical opening was determined as the perpendicular distance between the lower first molar distal cusp tip (L6DCT) to the FOP.

Results

At the outset the mean mandibular advancement by the appliances was 6.83 mm (standard deviation 1.78 mm), the mean incisal opening was 5.64 mm (standard deviation 1.86 mm), and the mean opening opposite the first molars was 2.61 mm (standard deviation 1.36 mm). There were no significant cephalometric differences between the groups or between the male and female subjects.

Changes in the combined group

When all the subjects were combined into a single group to determine the extent of the dental and occlusal changes, 12 small but statistically significant increases and five significant reductions were found between the initial and review films (Table 1). A small increase in SNA was accompanied by a similar increase in ANB. The total anterior face height (N–Me), lower face height (Me/ANS–PNS), and posterior face height (S–Go, Go/ANS–PNS) also increased significantly. Approximately 86 per cent of the increase in the total anterior face height occurred in the lower face. Maxillary length (ANS–PNS) increased significantly and the mandible was displaced significantly downward (Cd vert, Fid_1 vert), but not forward. Both the mandibular first molar and the maxillary first premolar (L6DCT vert, Pm vert) over-erupted slightly. Significant retroclination of the maxillary incisors (UI/IZ_1–IZ_2) and proclination of the mandibular incisors (LI/Fid_1–Fid_2) were accompanied by reductions in maxillary arch length, overbite, and overjet. There was a statistically significant positive association between the amount of advancement obtained with each appliance and the ANB angle (n = 100, r = 0.302, P = 0.002).

Changes over time

The significant changes over time are given in Table 2. A conspicuous finding is that only one variable (Fid_1 vert) was common to all periods. At the first review period (T2, 6 months), small but statistically significant increases in face height (N–Me, S–Go, Go/ANS–PNS) were accompanied by a significant downward position of the mandible (Cd vert, Fid_1 vert) and significant reductions in overbite and overjet. Only one significant increase (Fid_1 vert) occurred at 12 months. Significant increases in both the total and lower anterior face heights (N–Me, Me/ANS–PNS) and the vertical position of the mandible relative to the cranial base (Cd vert, Fid_1 vert) were found after 18 months' wear of the splint. The overbite and overjet were also reduced in this group. By the second year of wear, SNA and face height (N–Me, Me/ANS–PNS) had increased. The mandible continued to be lower (Cd vert, Fid_1 vert) down the articular eminence than at the outset. Both the mandibular first molars (L6DCT vert) and the maxillary first premolars (Pm vert) had over-erupted and the mandibular incisors had been proclined (LI/Fid_1–Fid_2). At the final review period the mandibular incisors were proclined, on average, 4 degrees and the overbite and overjet reduced. The lower face height, although still increased, was less at 30 months than at 18 and 24 months. There were no significant changes in the positions of the first molars or in arch lengths. Statistically significant positive correlations were also found between the amount of anterior opening obtained with each appliance and the changes in overbite at 24 and 30 months (Table 3).

Discussion

There is good evidence that in untreated individuals small dental and occlusal changes occur throughout life (Behrents, 1985). For this reason, reference planes were used based on anatomically stable landmarks (Johnston, 1996) to detect any changes in the positions of the teeth and mandible. Mandibular advancement appliances, used in the treatment of OSA, widen the oropharyngeal airway by repositioning the mandible downwards and forwards during sleep. Therefore, it was not surprising that they produced similar changes to functional appliances (Aelbers and Dermaut, 1996; Collett, 2000). In agreement with Bondemark (1999), a small but statistically significant increase was found in face height, the mandible was lower relative to the cranial base, the maxillary first premolar and mandibular first molar had over-erupted, the overbite and overjet were reduced, and the mandibular incisors were proclined. No significant changes were found in the positions of the maxillary first molars with the appliance used.

The findings from this study show that the so-called skeletal and mandibular positional changes can be mainly attributed to appliance-induced dental changes, although initial facial growth may have contributed (Behrents, 1985). For example, the increases in SNA and ANB, which some might argue are evidence of 'skeletal' change, are more likely to be due to remodelling of point A following retroclination of the upper incisors. This conclusion is supported by a failure to find significant changes in the positions of the anatomically stable reference points in the maxilla relative to the cranial base. The increase in palatal length is somewhat more problematic: it may be due to the difficulty in locating either ANS, PNS or both points, remodelling of ANS secondary to retroclination of the upper incisors, or a

Table 2 Significant changes during mandibular advancement splint therapy, by length of treatment.

Variables	Initial		Review		Mean difference	*P*
	Mean	SD	Mean	SD		
6 months (n = 20)						
N–Me	131.02	5.06	132.05	4.93	–1.03	**0.0001**
S–Go	86.88	7.46	87.89	7.47	–1.01	**0.001**
Go/ANS–PNS	38.59	6.12	39.84	5.86	–1.25	**0.001**
Cd vert	16.83	3.46	17.837	3.6	–1.01	**0.022**
Fid_1 vert	119.11	6.4	119.96	7.14	–0.85	**0.012**
Overbite	3.35	1.98	2.74	1.95	0.61	**0.037**
Overjet	3.67	1.45	2.8	1.61	0.87	**0.001**
12 months (n = 20)						
Fid_1 vert	118.58	9.57	119.78	9.22	–1.2	**0.011**
18 months (n = 20)						
N–Me	131.63	5.36	132.23	5.43	–0.7	**0.033**
Me/ANS–PNS	74.1	4.04	74.66	4.06	–0.56	**0.048**
Cd vert	17.48	3.47	18.41	3.64	–0.93	**0.043**
Fid_1 vert	119.81	5.73	120.46	5.93	–0.66	**0.031**
Overbite	3.83	2.45	2.93	1.74	0.9	**0.008**
Overjet	4.36	1.97	3.3	1.62	1.06	**0.0001**
24 months (n = 20)						
SNA	81.25	4.48	81.95	4.09	–0.7	**0.024**
N–Me	130.47	8.36	131.75	8.64	–1.28	**0.0001**
Me/ANS–PNS	72	5.57	73.05	5.65	–1.05	**0.007**
Cd vert	18.05	3.58	19.07	3.24	–1.02	**0.007**
Fid_1 vert	117	8	118.4	7.93	–1.4	**0.0001**
L6DCT vert	20.69	2.51	21.87	1.83	–1.17	**0.025**
Pm vert	32.29	3.47	33.02	3.8	–0.73	**0.025**
LIE horiz	7.48	5.9	6.83	5.94	0.65	**0.02**
LI/Fid_1–Fid_2	84.56	10.66	86.71	9.87	–2.15	**0.002**
Overbite	4.5	2.31	3.14	2.21	1.36	**0.0001**
Overjet	4.79	3.49	3.53	2.52	1.26	**0.001**
30 months (n = 20)						
Me/ANS–PNS	71.84	6.24	72.68	6.37	–0.84	**0.015**
Fid_1 vert	117.82	8.61	118.66	8.4	–0.84	**0.023**
UIE vert	37.21	3.75	36.52	3.98	0.69	**0.029**
LIE horiz	9.2	5.58	8.14	4.94	1.06	**0.012**
LI/Fid_1–Fid_2	82.04	9.7	86.34	9.36	–4.3	**0.0001**
Overbite	4.43	2.6	2.61	2.27	1.82	**0.005**
Overjet	3.84	1.71	2.63	1.27	1.21	**0.005**

Significant differences at the 5 per cent level are shown in bold.
SD, standard deviation.

Table 3 Associations between anterior opening by the appliance and change in overbite.

Duration (months)	n	r	*P*
6	20	0.103	0.667
12	20	–0.036	0.879
18	20	0.019	0.936
24	20	0.611	**0.004**
30	20	0.573	**0.008**

Significant differences at the 5 per cent level are shown in bold.

chance finding. The latter should not be dismissed because the probability of one or more tests being significant by chance alone at the 5 per cent level out of 36 tests is high (P = 0.84). Downward positioning of the mandible by the appliance increases face height which allows the mandibular molars and maxillary premolars to slightly over-erupt. The significant maxillary incisor retroclination, which was only found in the larger combined sample, can be attributed to the appliance acting directly on the incisors. However, increased pressure from the lips due to the altered mandibular posture may also play a part. Although the changes in the positions of the maxillary incisors were small and highly variable, a precision metal casting enclosing the teeth may prevent their movement and the consequent reductions in overbite, overjet and the length of the maxillary arch that were found.

When the changes over time were examined there was considerable variation both within and between

groups. Only downward displacement of the mandibular symphysis was found at all review periods. While an increase in face height and reductions in overbite and overjet were evident at 6 months, over-eruption of the maxillary first premolars and mandibular first molars, and proclination of the lower incisors were not detected until 24 months. Significant positive correlations were also found between the amount of anterior opening due to each appliance and the changes in overbite at 24 and 30 months. The appliances used in this study were, on average, 2.6 mm thick (standard deviation 1.3 mm) posteriorly and 5.6 mm thick (standard deviation 1.8 mm) anteriorly. This finding could indicate that the thickness of these appliances should be kept to the minimum consistent with the depth of the overbite and the use of screws to advance the mandible. At present there is no convincing evidence that this is the case, because if a causal relationship does exists between anterior opening by an appliance and a change in overbite it would be reasonable to expect it to be more obvious with time. No significant association was found between the amount of bite opening and the change in overjet. The only consistent finding in the smaller groups was a downward positioning of the mandibular symphysis.

As a non-adjustable appliance was used in this study, it can be argued that these results are not comparable with those where fully adjustable appliances were used. Pantin *et al.* (1999), however, considered that dental side-effects were largely generic to mandibular advancement and tended not to be device specific. In a review article, Schmidt-Nowara (1999) concluded that the newer titratable appliances created the potential of a greater degree of mandibular advancement, but also a greater likelihood of complications. Overall, very few of the occlusal changes found in this study were of concern to the patients, with treatment discontinued in only one patient due to adverse dental side-effects. Unfortunately, cephalometric predictors for patients who may be at risk from adverse occlusal changes with mandibular advancement treatment have yet to be established. Further investigation in this area is needed, especially for those patients who are at present undergoing treatment with fully titratable appliances as, to date, no data of the long-term effects of these appliances on the occlusion have been forthcoming.

Conclusion

The present study provides data on the extent, and time course, of dental and occlusal changes occurring during mandibular splint therapy in sleep disordered patients. Although the response by the subjects in this study was extremely variable, changes in face height, the position of the mandible, overjet, and overbite occurred as early as 6 months. Over-eruption of the maxillary first premolars and mandibular first molars and proclination of the mandibular incisors were not evident for at least 2 years. These preliminary results suggest that overbite changes might be lessened by keeping the bite opening by an appliance to a minimum.

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Treatment of Long-standing Nocturnal Enuresis with Oral Appliances.

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Abstract

Enuresis, the involuntary release of urine during sleep, is one of the most common disorders in childhood. More common in boys than girls, this condition is characterised by night-time wetting in the presence of normal urinalysis and physical examination. At present treatment can be divided into behavioural modification and pharmacological therapy, despite which many enuretic children remain untreated, or are treated ineffectively. Treatment of long-standing chronic enuresis by orthodontic appliances could be used more frequently to give relief to those not responding to conventional treatment. Mandibular advancement therapy is especially beneficial in the treatment of enuretic patients presenting with anteroposterior skeletal discrepancies.

Key Words: nocturnal enuresis, mandibular advancement therapy, maxillary expansion.

Background

Nocturnal enuresis is one of the most prevalent and persistent sleep problems in children. Despite intensive clinical research, many enuretic children remain untreated or are treated ineffectively.[1] Nocturnal enuresis is characterised by the frequent occurrence of normal complete uncontrolled micturition during sleep in children older than 5 years of age.[2] Research suggests that enuresis is most prevalent in the United States with as many as 8% of boys and 4% of girls still enuretic at age 12.[1]

The most widely used criteria for enuresis specifies two or more incontinent events in a month over 5 years of age with the absence of a physical disorder associated with incontinence such as urinary tract infection or diabetes. Enuresis is divided into primary, where the child has never had a long period of nocturnal continence, and secondary, where night-time wetting reoccurs after one year of continence and is more likely to have a pathologic etiology. Primary enuresis is far more frequent than secondly with an approximate ratio of 10:1.[2]

Polyuria is a common feature in enuretic children. Normal subjects show a marked circadian variation in urine output and osmolality with a reduced volume of concentrated urine during night-time.[2] Urine production during night-time being about half that of day-time. It has therefore been suggested that a substantial number of enuretic children may lack physiological circadian variation in urine excretion rate and urine osmolarity due to no increase in the antidiuretic hormone vasopressin (ADH) at night.[3]Enuresis is reported to be a common symptom of obstructive sleep apnea in children [4,5] and is suggested to be secondary to increased intra-abdominal pressure from paradoxical breathing associated with increased respiratory effort.[2]

Current treatment of nocturnal enuresis is varied and is often dependent on the age of the child. Children under the age of 6 years may be simply managed by reassurance

and preventing irregular sleep – wake patterns and sleep deprivation. Other behavioural treatments include urine retention control training (RCT), alarm monitoring, stream interruption and waking the child at night for urination. Described in the literature as the single most effective treatment for enuresis, the urine alarm works by negative reinforcement awakening the child at night at the onset of urination using a moisture sensitive switching system attached to the child's pyjamas or underwear. In any case, whatever the age of the child, a micturition frequency chart associated with volume evaluations will allow determination of the severity of bedwetting. Evidence shows that enuretic children do exhibit a 15% annual spontaneous remission rate, which is consistent with the opinion that these children are simply lagging behind in the acquisition of continence.[6] Medical treatment for long-standing enuresis includes the use of tricyclic antidepressants and antidiuretics. Thanks to its safety, the antidiuretic desmopressin has now replaced the antidepressant imipramine in the pharmacological approach to enuresis. Desmopressin acts by reducing nocturnal urine production and is thought to also improve the patients ability to awaken and improve bladder instability. Although desmopressin offers better short-term results than the alarm system, the latter is found to be significantly more effective in the long-term.[2] The use of desmopressin is usually limited for the treatment of enuresis in older children and adolescents once other behavioural methods have been unsuccessful owing to the potential of adverse side effects. Although an overall 70-80% success rate of adequate treatments is reported in the literature, others do however report an apparent resistance to all available treatments. A subgroup of enuretic children has subsequently been identified with normal circadian levels of ADH, without polyuria that are unresponsive to either desmopressin or behavioural methods. The following case report describes the

successful treatment of such a patient with long-standing nocturnal enuresis with the use of mandibular advancement therapy.

Case Report.

A 12.3 year-old boy was referred from the Paediatric Outpatient Clinic, Dunedin Public Hospital for snoring and long-standing nocturnal enuresis (1-2 events per night). Previous unsuccessful treatment included adenotonsillar surgery corticosteroidal nasal sprays, behavioural programs (RCT), alarm monitoring and the use of tricyclic antidepressants and antidiuretics. Overnight polysomnography was normal with the exception of intermittent snoring and one episode of enuresis in deep sleep. As this patient presented with a Skeletal II facial pattern (mandible post-normal to maxilla) (Figure I) an orthodontic functional appliance (Clark Twin Block) was inserted for mandibular advancement therapy. (Figure 2). Following one months' treatment, snoring had ceased and nocturnal enuresis was reduced from a nightly occurrence to 1-2 events per week. After three months appliance wear and further mandibular advancement (2mm), the patient was now aware of the onset of enuresis. After eight months of treatment, the appliance was discontinued after one month of continence, this however, resulted in a reoccurrence of enuresis (1-2 events per week). Continence was again achieved by reinsertion of the appliance. After one year of treatment use of the appliance was restricted to sleep-overs and school camps as prolonged periods of dryness were now being achieved without the appliance insitu. From an orthodontic perspective, use of a mandibular advancement appliance in this patient also doubled as a 'functional' orthodontic appliance for the partial correction of a skeletal II facial pattern.

Case Discussion

Nocturnal enuresis has been reported as a common symptom among children with sleep disordered breathing. [3,4] Recent studies of enuretic children who underwent adenotonsillar surgery have shown a marked reduction in nocturnal enuresis[7,8] which suggests that upper airway obstruction is probably a more common etiological factor in this condition than previously recognised.

Topal et al [9] investigated whether the presence of nocturnal enuresis was related to the severity of obstructive sleep apnea (OSA) in 160 enuretic children suspected of sleep disordered breathing. Following polysomnography, nocturnal enuresis was found to be more prevalent in children with a respiratory disturbance index (RDI) greater than one, but more severe OSA did not further increase the risk of enuresis.

The use of orthodontic appliances to treat nocturnal enuresis is not new. Timms [10] successfully treated 10 patients with long-standing nocturnal enuresis by orthodontically expanding the maxilla 6-10 mm. Kurol et al [11] used a similar device but with less maxillary expansion (3-5 mm) resulting in 7 out of 10 enuretic children wetting the bed less often within one month and 4 children becoming completely dry. Rhinomanometry before and after maxillary expansion showed less nasal resistance after treatment but as only minimal transverse expansion was found in the region of the interior concha nasalis (0.4 – 1.2 mm) no statistical association could be found between nocturnal enuresis and maxillary expansion. In the study of Kurol et al [11] only one patient presented with a unilateral posterior cross-bite (transverse occlusal discrepancy), the remaining nine patients in this study had 'normal' transverse occlusions. From an orthodontic perspective, it is not surprising therefore that the use of maxillary expansion in patients with normal transverse occlusions has not been widely accepted as a treatment choice for nocturnal enuresis. Conversely, the use of

mandibular advancement in enuretic children may also be helpful in the orthopedic correction of certain facial types (skeletal II facial pattern) as well as aid in the treatment of nocturnal enuresis. As many enuretic children present for orthodontic treatment at adolescence (given the prevalence of enuresis in 12 year olds[1]), mandibular advancement therapy is most likely used in some of these patients under the guise of "functional orthodontic treatment" for the correction of anteroposterior skeletal discrepancies. In many cases, the clinician is probably unaware of the patient's enuresis and hence subsequent treatment of this condition by mandibular advancement goes unreported. In this case report, treatment of persistent, long-standing nocturnal enuresis in a 12 year old boy was successfully treated by mandibular advancement.

Recommendations for Clinical Management and Research

Most investigators consider nocturnal enuresis to be a multicausal disorder involving genetic, developmental, organic and psychological factors. The pathophysiology of this disorder, however, remains uncertain as it is likely that there are several forms of nocturnal enuresis according to their clinical features. From a practical point of view most patients respond well to either behavioural methods (alarm system) or antidiuretics (desmopressin). There appears however, to be a subgroup of patients that are non-responsive to either behavioural or pharmacological treatments. Use of polysomnography in such patients may be of benefit to determine any causal relationship between nocturnal enuresis and upper airway obstruction. Use of orthodontic appliances could be considered as a viable treatment alternative to those patients who have not responded either to conventional treatment or to surgical management of upper airway obstruction and/or who present with occlusal

discrepancies that would benefit from orthodontic intervention. In patients that present with transverse occlusal discrepancies, use of maxillary expansion is recommended. Conversely, in patients who present with antero-posterior skeletal discrepancies, the use of mandibular advancement therapy is considered the treatment of choice.

In conclusion, use of orthodontic appliances could be used more frequently to give relief to those children not responding to conventional treatments for long-standing nocturnal enuresis. Further research is necessary however, to determine the causal relationship between long-standing nocturnal enuresis and sleep-disordered breathing and the effect of orthodontic treatment as a viable treatment alternative.

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Figure 1: Lateral cephalogram of enuretic patient.

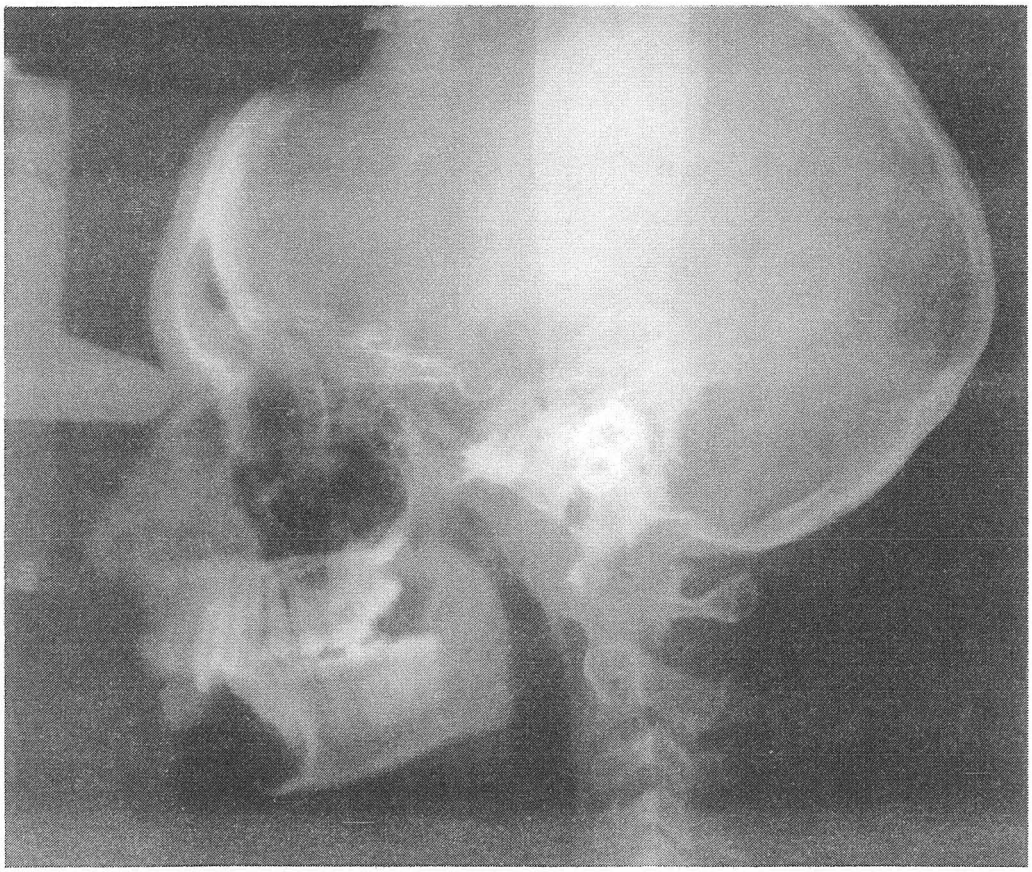

Figure 2: Mandibular advancement splint insitu.