From the Department of Orthodontics, Institute of Odontology,
Karolinska Institutet, Stockholm, Sweden

A LONGITUDINAL STUDY OF DENTOFACIAL MORPHOLOGY IN YOUNG CHILDREN TREATED FOR THE OBSTRUCTIVE SLEEP APNOEA SYNDROME

Lena Zettergren Wijk

Karolinska Institutet

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ABSTRACT

Obstructive sleep apnoea (OSA) is the most extreme variant of mouth breathing and snoring in a wide spectrum of symptoms of upper airway obstruction during sleep. Typical night-time symptoms in children suffering from OSA are disturbed sleep and snoring. Common daytime symptoms are mouth-breathing, noisy breathing, behavioural problems, and failure to thrive. When OSA leads to physical and/or mental consequences it is diagnosed as a syndrome (OSAS). The prevalence of OSAS in children is 1-3%, with the peak incidence at the ages 2 to 6 years. In the pre-pubertal period, girls and boys are equally affected. Left untreated, OSAS may lead to developmental delay, cor pulmonale or even death.

The most common cause of OSA in children is enlarged tonsils and/or adenoids. Consequently, the treatment for OSA in children with enlarged tonsils and/or adenoids is adeno-/tonsillectomy. There are few longitudinal studies presented in the literature on dentofacial morphology in children suffering from and treated for OSA.

The aims of the present study were:

- to study the dentofacial morphology in children with OSAS and make comparisons with the morphology in children without obstructed airways;
- to evaluate prospectively the clinical manifestations, sleep recordings, and facial and dental development in children with OSAS before and one and three years after tonsillectomy and/or adenoidectomy;
- to longitudinally evaluate, over a five year period, the development of dentofacial morphology, soft tissue profile, and airway space after successful treatment of the OSA (adeno-/tonsillectomy), and to carry out a comparison with the normal dentofacial development in non-obstructed children.

Material and methods

The original sample comprised 20 consecutive prepubertal children, 8 girls and 12 boys, mean age 6 years (age range 4 to 9 years). They had been referred to, or were seeking medical attention at the Department of Otorhinolaryngology, Söder Hospital in Stockholm, because of a typical history of the OSA syndrome. All children underwent tonsillectomy and/or adenoidectomy.

Study I: A 1-year follow-up study of 20 OSAS children who were their own controls.

Study II: A 3-year follow-up study of 14 OSAS children and 14 age and sex-matched control children who showed no signs of obstructed upper airways.

Study III: . A 5-year follow-up study of 17 OSAS children and 17 age and sex-matched healthy controls.

Pre-surgically, the OSA children underwent clinical examination by an otolaryngologist, sleep registration with polysomnography, and orthodontic examination. Follow-up examinations and registrations were made at 1, 3, and 5 years after surgical treatment.

The clinical examination consisted of an evaluation of the lymphoid tissues in the naso- and oropharynx, general health, and body weight. The parents´ reports of day and night-time signs and symptoms of obstructive breathing were also recorded.

Sleep stage, pulse frequency, airflow, oxygen saturation, body movement, and activity in accessory respiration muscles during sleep were recorded during a whole nights´ sleep in a sleep laboratory. At the 1-year follow-up, a less extensive sleep recording was made.

Study models of the dentition were used for registration of the width of the upper dental arch and the occlusion. Measurements of angular and linear variables were made on lateral radiographic head films for evaluation of dentofacial morphology.

Results
Before adeno-/tonsillectomy, the most frequent observation during sleep was increased respiratory labour with increased use of accessory respiration muscles. In three children who were videofilmed parallel to the sleep recording, it was confirmed that they had extended head posture to increase airflow. The heavy respiratory labour was caused by obstructed nasopharyngeal airways, which were significantly reduced in width in the patients as compared to the controls. In several children, apnoeas/hypopnoeas > 1/hour of sleep and low oxygen saturation in arterial blood (<89%) were observed.

Compared to non-obstructed controls, the OSAS children exhibited a narrow upper dental arch, high frequency of lateral crossbite, and reduced overbite. The patients also had a large lower anterior face height, a posteriorly inclined mandible and facial axis, and retroclined incisors.

At the 1-year follow-up examination, none of the children exhibited clinical signs or symptoms of obstructive breathing. Furthermore, a catch-up in body weight was recorded.

One year after surgery, it was also observed that the width of the upper dental arch had increased significantly more ($p<0.01$) in the OSAS children than in the controls, and the lateral crossbite had spontanously resolved in two children. Furthermore, changes towards a more normal dentofacial development were seen in the patients. This beneficial trend could also be confirmed in the 3-year follow-up study.

At the 5-year follow-up, there was no significant difference between the OSAS children and the controls in the dimension of the nasopharyngeal airways. With the exception of the length of the anterior cranial base and the nose, which were still significantly smaller ($p<0.01$) in the OSAS children than in the controls, there were no differences in dentofacial morphology between the groups.

Conclusion
Young children suffering from OSAS have a different dentofacial morphology compared with non-obstructed control children.
Treatment of OSA with adeno-/tonsillectomy was successful in the present patients, and the dentofacial development was normalised after surgery.
It is important that OSA in young children is diagnosed early, and that the patients are evaluated both from a medical and dentofacial point of view.

LIST OF PUBLICATIONS

This thesis is based on the following articles, which are referred to in the text by their Roman numerals (I-III):

I. Ågren K, Nordlander B, Linder-Aronson S, Zettergren-Wijk L, Svanborg E. Children with nocturnal upper airway obstruction: Postoperative orthodontic and respiratory improvement. Acta Oto-Laryngologica 1998; 118: 581-587

II. Zettergren-Wijk L, Linder-Aronson S, Nordlander B, Ågren K, Svanborg E. Longitudinal effect on facial growth after tonsillectomy in children with obstructive sleep apnea. World Journal of Orthodontics 2002; 3: 67-72

III. Zettergren-Wijk L, Forsberg C-M, Linder-Aronson S. Changes in dentofacial morphology after adeno-/tonsillectomy in young children with obstructive sleep apnoea - a 5-year follow-up study. European Journal of Orthodontics 2006; 28: 319-326

CONTENTS

LIST OF DEFINITIONS AND ABBREVIATIONS

Apnoea	Cessation of airflow. Persistence of respiratory effort distinguishes obstructive apnoeas from central apnoeas.
Hypopnoea	Respiratory event with reduced airflow, but not completely abolished. There is no general consensus on the exact definition of hypopnoea.
Arousal	An abrupt change from a “deeper” stage of NREM sleep to a “lighter” stage, or from REM sleep toward wakefulness, with the possibility of awakening as the final outcome. Arousal may be accompanied by increased tonic EMG activity and heart rate as well as body movements.
Fragmentation	(Pertaining to sleep architecture) The interruption of any stage of sleep due to appearence of another stage or wake, leading to disrupted NREM - REM sleep cycles. Sleep fragmentation connotes repetitive interruptions of sleep by arousals and awakenings.
Pickwickian syndrome	Syndrome characterised by obesity, hypersomnolence, periodic breathing with hypoventilation, and cor pulmonale.
Waldeyer´s ring	A collection of pharyngeal lymphoepithelial tissues, constituted of the adenoids, the tonsils, the lingual tonsils, tubal tonsils and tubopharyngeal plicae. The adenoids and tonsils are involved in regulation of mucosal immunity in pharynx, a defence system against microbial invasion. The adenoids and tonsils undergo most of their development after birth.

AI	Apnoea index = average number of apnoeas per hour of sleep
AHI	Apnoea/hypopnoea index = average number of apnoeas plus hypopnoeas per hour of sleep
ALTE	Acute life-threatening event
CNS	Central nervous system
CPAP	Continuous positive airway pressure
ECG	Electrocardiography
EEG	Electroencephalography
EMG	Electromyography
EOG	Electrooculography
GH	Growth hormone

nadir SaO_2	Lowest oxygen saturation in arterial blood
NREM	Non-REM sleep
ODI	Oxygen desaturation index
OSA	Obstructive sleep apnoea
OSAS	Obstructive sleep apnoea syndrome
PSG	Polysomnography
PVDF	Polyvinylidene fluoride
REM	Rapid eye movement
SaO_2	Oxygen saturation in arterial blood
UPPP	Uvulopalatopharyngeoplasty

Pediatric polysomnography (Carroll, 1996)

Measurement	Purpose
EEG	Sleep staging, detection of seizure activity
EOG	Eye movement - sleep staging
EMG	Sleep staging, body movement detection, detection of respiratory related muscle activity
ECG	Heart rate, to detect arrythmias, bradycardia
Chest/abdomen movement	To detect respiratory effort
Nasal/oral thermistor	To detect nasal/oral airflow
Oxygen saturation by pulse oximetry	To detect hypoxemia
End-tidal CO_2	To detect hypoventilation
Body position	To correlate body position with respiratory events
Audio/video taping	To verify breathing patterns and for review of parasomnias (such as sleepwalking) or other events (e.g. seizure activity)

1 BACKGROUND

1.1 INTRODUCTION

In 1872, C.V. Tomes described dentofacial changes in association with chronic upper airway obstruction and introduced the “adenoid face” concept. In 1889 W. Hill specified the characteristics of “the stupid-lazy child” who showed day and night-time symptoms typical for the obstructive sleep apnoea syndrome (OSAS), and recommended the school medical officers to pay special attention to these children. However, even though the clinical manifestations of obstructive sleep apnoea (OSA) were recognized long ago, it was not until 1966 that Gastaut presented the first scientific report of OSA in the medical literature. That report was based on a polygraphic study of a group of adults suffering from the Pickwickian syndrome. The first report on OSA in children was published by Guilleminault *et al.* in 1976. Both these studies focused on the effect of OSA on general health.

During the last few decades, many articles dealing with OSA in children have been presented. These studies have covered various topics such as diagnostic criteria, behavioural problems, learning difficulties, relation to acute life threatening event (ALTE), treatment, obstructive breathing, and dentofacial development and morphology.

1.2 MODE OF BREATHING

Nose-breathing is the natural way of breathing in humans. In the newborn, normal breathing is considered essential for a healthy development (Gaultier, 1991). The space of the nasal airway and pharynx is dependent on the skeletal dimensions and morphology of the nasomaxillary complex, the naso-pharynx, and the oro-pharynx. The smallest area of the pharyngeal airway perpendicular to the air stream through the upper

airways is the critical factor for the resistance to the air flow (Trenouth and Timms, 1999). When the resistance increases as a result of unfavourable morphological and/or functional factors, a change from nose to mouth-breathing may occur. Furthermore, the soft tissue thickness in these areas is also of importance. In children, the lymphoid tissues (tonsils and adenoids) in the so called Waldeyer´s ring are of particular significance in this context.

Depending on the cause of the obstruction, the mouth-breathing condition can last for a short period of time or be virtually permanent. Habitual mouth-breathing which is not caused by obstructed upper airways may develop in combination with incompetent lip closure, large overjet, or open bite.

Mouth-breathers with obstructed upper airways often snore during sleep, and in extreme circumstances may develop OSAS (Carroll, 1996; Mindell *et al.,* 1999). The patency and/or collapsibility of the soft tissues in the airways is influenced by the neuromuscular function. When the individual is awake, activity in accessory respiration muscles and/or pharyngeal dilator muscles increases the space and/or enhances the patency of the pharyngeal soft tissues to establish an open airway passage. Obstructive apnoeas do not, therefore, occur in this situation. At sleep onset, this activity decreases and a rise in upper airway resistance occurs (Fogel *et al.,* 2003). In cases where airflow is already negatively affected by large tonsils and adenoids it is reasonable to assume that a decrement in the activity of the pharyngeal dilator muscles during sleep is a factor of importance in the development of a condition such as OSA.

1.3 CHARACTERISTICS IN DENTOFACIAL MORPHOLOGY ASSOCIATED WITH OBSTRUCTIVE BREATHING

1.3.1 Children

The morphology of the facial skeleton and dentition is the result of hereditary and environmental factors. The heredity is polygenetic, and Moss (1968) stressed the morphogenetic role of the naso-, oro-, and pharyngeal cavities in the growth of the splanchnocranium. As the volume of these functioning spaces increases, the surrounding capsule expands and the embedded macro-skeletal units are passively translated in space. However, environmental influences of various kinds (health status, hormones, nutritional status, the neuromuscular functional pattern and activity, and soft tissue morphology and growth) may modify the genetically determined pattern of growth.

Several studies (e.g. Linder-Aronson, 1970; Hannuksela, 1981; Bresolin *et al.*, 1983; Behlfelt, 1990) have demonstrated that children with obstructed airways exhibit a dentofacial morphology which differs from that of non-obstructed controls. Experimental studies on primates, in which a change from nose to mouth-breathing has been induced, indicate that an altered breathing pattern actually causes a change in dentofacial morphology (Harvold, 1968). On the other hand, the opinion that mouth-breathing does not affect dentofacial morphology, has been put forward by Kingsley (1888), Whitaker (1911), Sillman (1942), Hartsook (1946), Leech (1958), Tulley (1966), Vig (1985), and others.

Linder-Aronson (1970) has shown that children with obstructed upper airways, mouth-breathing and enlarged adenoids have an increased anterior face height, a large angle between the upper and lower jaws, reduced dimension of naso-pharynx in the sagittal plane, a narrow upper dental arch, crossbite or a tendency for crossbite, and retroclined upper and

lower incisors. The same study also demonstrated that nasal obstruction and mouth-breathing was associated with a low tongue position. Furthermore, it has been shown that the facial growth pattern changes and follows a more normal course after adenoidectomy (Linder-Aronson, 1975; Linder-Aronson *et al.*, 1986; Linder-Aronson *et al.*, 1993).

Behlfelt (1990) studied the effect of enlarged tonsils and tonsillectomy on breathing pattern and dentofacial development. The findings were similar to those reported by Linder-Aronson (1970, 1975).

1.3.2 Adults

Lateral cephalometric radiographs have been used in an attempt to analyse and identify morphologic parameters that might be characteristic in adult apnoeics. The following two-dimensional deviations from normal morphology have been reported:

1. The cranial base may be short and the cranial base angle reduced (Andersson and Brattström, 1991; Battagel and L´Estrange, 1996).
2. Bimaxillary retrusion (Lowe *et al.*, 1986; Solow *et al.*,1996).
3. Retrognathia of the mandible alone (Jamieson *et al.*, 1986).
4. Increased mandibular plane angle (Lowe *et al.*, 1986; Andersson and Brattström, 1991).
5. Increased anterior face height (Lowe *et al.*, 1986).
6. Extended head posture (Solow *et al.*,1996).
7. Lowered hyoid bone position in relation to the mandibular plane (Jaimison *et al.*,1986; Partinen *et al.*, 1988).
8. Reduced dimension of the pharynx (Lowe *et al.*, 1986; Andersson and Brattström, 1991; Battagel and L´Estrange, 1996).
9. Long soft palate (Jamieson *et al.*, 1986; Lyberg *et al.*, 1989).
10. Reduced functional area for the tongue (Battagel and L´Estrange, 1996).

An approximate estimation of the degree of airway obstruction may also be obtained on lateral radiographic head films. However a true assessment of the airway dimensions requires a three-dimensional recording technique such as computerised tomographic scanning (CT) or magnetic resonance imaging (MRI) (Lowe *et al.*, 1986; Guilleminault and Stoohs, 1990).

2 OBSTRUCTIVE SLEEP APNOEA SYNDROME

Sleep apnoea can be of central, peripheral (=obstructive) or mixed central and peripheral origin. In central sleep apnoea, a deficient respiratory stimulus from the CNS causes the apnoea and there is no thoracical respiratory movement. In the peripheral sleep apnoea, an obstruction of the upper airways prevents air passage into the lungs, despite intense thoracical movement. The labelling of the OSA condition as a syndrome implies the presence of symptoms in addition to laboratory findings (Table 1).

2.1.1 Children

The definition of OSAS in children is under debate, and different authors may use different criteria to describe this condition. The "gold standard" to diagnose OSAS in children is polysomnograhic sleep recording in a sleep laboratory (Carroll, 1996; Ågren *et al.*, 1998; Mindell *et al.*, 1999; Marcus, 2001). In children, an AI >1 is classified as sleep apnoea (Marcus *et al.*, 1992; Marcus, 2001). Criteria for sleep apnoea in adults are not applicable in children (Rosen *et al.*, 1992).

2.1.2 Adults

In 1993, a State of the Art Conference focusing on the diagnosis and management of OSAS in adults was held in Stockholm. The consensus of opinion at the conference was that a previously proposed definition (Siuntio, Finland 1988) should be adopted by the conference. According to this definition "*OSAS is an intermittent, complete or partial, upper airway obstruction during sleep causing mental and/or physical effects*". The American Academy of Sleep Medicine (1999) states that the diagnostic criteria for OSAS should include an AHI ≥ 5 recorded during nocturnal PSG, and each apnoea should last for at least 10 seconds.

Table 1: Comparison of some characteristics of OSAS in children and adults (Carroll, 1996; Marcus, 2001).

	Children	***Adults***
Clinical characteristics:		
Peak age	Preschool children	Elderly
Sex	Equal	Predominantly men Females post menopause
Aetiology	Adenotonsillar hypertrophy	Obesity
Weight	Failure to thrive, normal, obese	Obese
Mouthbreathing (daytime)	Common	Less common
Excessive daytime somnolence	Less common	Very common
Neurobehavioural	Hyperactivity, developmental delay	Cognitive impairment, impaired vigilance
Polysomnographic characteristics:		
Obstruction	Cyclic obstruction or prolonged obstructive hypoventilation	Cyclic obstruction
Sleep architecture	Normal	Changed
Sleep stage with OSA	REM	Non-REM (or REM)
Cortical arousal	In <50% of apnoeas	At termination of each apnoea
Treatment:		
Surgical	Adeno-/tonsillectomy Tracheostomy (rarely)	UPPP (selected cases) Tracheostomy (rarely)
Medical	CPAP (occasionally) Corticosteroids	CPAP Dental splints

2.2 SYMPTOMS

The symptoms of OSA in children differ in many respects from those in adults. The typical clinical manifestations are disturbed sleep and noisy breathing while snoring is not a prominent feature in some children (Guilleminault and Stoohs, 1990). The upper airway obstruction may present itself mainly as paradoxical breathing and inward rib cage motion with or without the occurrence of apnoea. In addition to excessive daytime sleepiness, failure to thrive is a common symptom in such children. Many

children experience only mild symptoms with spontaneous improvement, whereas in others the condition is progressive.

Secondary effects of nasal obstruction have been previously studied. Disturbed acid-base balance in blood (Lüscher, 1930; Kreewinsch, 1932) and influence on pulmonary function (Ogura *et al.*, 1964; Togawa and Ogura, 1966) have been reported. In 1965 Noonan, and soon thereafter Luke *et al.* (1966), described an association between cronic upper airway obstruction and cardio-respiratory complications from moderate cardial enlargement to cor pulmonale and polmonary edema that improved after adeno-/tonsillectomy. Morbidities that primarily affect the cardiovascular and neurobehavioural systems in children suffering from OSA have also been reported by Carroll (1996) and Gozal and O´Brien (2004).

2.3 DIAGNOSIS

The diagnosis OSAS in children is based on history (thorough anamnestic examination - questionnaire), physical examination, and laboratory tests. Common complaints from parents are noisy or disturbed breathing during sleep, in some cases aggravated by upper airway infections or allergies. Other complaints may be excessive daytime sleepiness, behavioural problems, enuresis, failure to thrive, or difficulty to swallow. The physical examination is often made by an otolaryngologist in order to evaluate the patency of the upper airways and the medical status. Furthermore, measurements of height and weight, and evaluations of respiratory, cardiovascular and neurological function are also carried out.

The accepted standard for laboratory examination is nocturnal polysomnographic sleep recording, which must show sleep, respiration, heart rate (ECG), body position, and leg movements. The recording should also include oximetry, EEG, EOG, and chin-EMG to show the quality of

sleep and the registration of the occurrence and frequency of arousals (Swedish Medical Research Council, 1994; Carroll, 1996).

2.4 AETIOLOGY

In children with OSAS, hypertrophic tonsils and/or adenoids are the most common aetiologic factors. Other less common factors which may be related with OSA are severe craniofacial anomalies, micro- or retrognathia, macroglossia, neurological lesions, and Down´s syndrome (Strohl *et al.*, 1978; Guilleminault and Stoohs, 1990; Carroll, 1996). Furthermore, epigenetic influence on developing craniofacial structures may result in narrow airways or closure during sleep (Jaimieson *et al.*, 1986; Partinen *et al.*, 1988), and studies have shown that the OSAS also has a strong familial component (Guilleminault *et al.*, 1995; Mathur and Douglas, 1995; Ovchinsky *et al.*, 2002).

Obesity is an additional factor which may increase the risk for developing OSA, particularly in adult males (Schwartz *et al.*, 2008).

2.5 EPIDEMIOLOGY

In a British study, 0.7 per cent of 4.5 to 5.5 years old children were reported to have significant sleep and breathing disorders (Ali *et al.*, 1993). A study from Iceland indicates a prevalence of at least 2.9 per cent in children aged 6 months to 6 years (Gislason and Benediktsdóttir, 1995), while in a Swedish cohort study on breathing obstructions in 4-year-old children, a frequency of 0.9 per cent was recorded (Löfstrand-Tideström *et al.*, 1999). In prepubertal children, the frequency of OSAS is the same in girls and boys (Carroll, 1996).

OSAS is most frequent in children at the ages of 2 to 6 years (Guilleminault *et al.*, 1981; Mindell *et al.*, 1999), which coincides with the

age when the lymphoid tissues occupy the largest volume in relation to the skeletal nasopharyngeal cavity (Linder-Aronson and Leighton, 1983). Other reasons for enlargement of the adenoids and tonsils are infections, allergic reactions, and tumors.

During puberty, boys with reduced airway space in particular are at risk of developing abnormal nocturnal breathing patterns. This can be caused by extensive soft tissue growth (Guilleminault and Stoohs, 1990) or enlargement of the tongue muscle from testosterone stimulation (Guilleminault *et al.*, 1989). Mindell *et al.* (1999) mentioned that the pattern of symptoms seen in early to mid-adolescent boys appears to more closely resemble the adult OSAS symptoms and their frequent association with obesity.

The prevalence of OSAS at the ages 30 to 60 years is about 4 per cent in men and 2 per cent in women (Young *et al.*, 1993).

2.6 TREATMENT

Children with suspected OSAS should be carefully evaluated and treated as early as possible since an impaired breathing pattern has an unfavourable influence on dentofacial development. Once the diagnosis OSAS has been established and the predisposing factors are identified, a rational treatment plan should be made. The treatment of choice depends on the causative factors and the location of the obstruction. Alternative treatments in children are surgery, orthodontic/orthopaedic treatment, improvement of the airway passage with certain devices, and medical therapy.

Surgery:

The goal of complete adeno-/tonsillectomy, which is carried out in patients with adeno-/tonsillar hypertrophy, is to increase airway space, and not primarily to treat recurrent inflammations or infections of the lymphoid tissues (Guilleminault *et al.,* 2004). That this operation has a dramatically relieving effect of the symptoms in OSA patients with adeno-/tonsillar hypertrophy, has been demonstrated by Ågren *et al.* (1998). For OSA patients who do not respond to usual treatment options it may be necessary to perform craniofacial surgery or tracheostomy to relieve or bypass the airway obstruction (American Academy of Pediatrics, 2002).

Orthodontic/Orthopaedic treatment:

A narrow maxilla with a high palatal vault, and/or a retropositioned mandible may contribute to a reduced naso and/or oro-pharyngeal airway space. In growing children functional orthopedic appliances which advance the mandible (Villa *et al.,* 2002) or expand the maxilla (=RME) (Guilleminault *et al.,* 2005; Villa *et al.,* 2007) may increase the upper airway space and improve respiratory function. An increased airway space might also be obtained in cases where orthodontic uprighting of lingually inclined teeth results in a change in the resting posture of the tongue (Guilleminault *et al.,* 2005). However, no long-time evaluations of the possible effect of these treatments have been found in the literature (Carvalho *et al.,* 2007).

Devices to improve the airway passage:

The treatment method called CPAP (continuous positive airway pressure) is based on a device consisting of a blower, a pressure control unit, a tube, and a mask which is applied to the patient’s nose. An air stream increases the volume of the airway thereby relieving the breathing obstruction. The CPAP method has been reported to be effective, but requires extensive

training, and in children it should be applied with care. In a case report Li *et al.* (2000) described the detrimental effect of using CPAP for a long time in a growing child. The pressure from the mask was suspected to have severely depressed the mid-face causing a maxillary hypoplasia.

Medical treatment:

Medical treatment could be indicated if the upper airway obstruction is caused by allergic enlargement of adenoids and/or mucosal membranes. Obese children, suffering from OSA, should be recommended weight control.

Treatments which make use of CPAP or medication are not generally succesful in curing OSA. The use of CPAP emerges as a secondary line of treatment in children who fail to respond to surgery, or when surgical treatment is contradicted.

It is important that the treatment of OSA is evaluated continuously during growth, especially in children with narrow upper airways, in whom even a minor soft tissue enlargement may cause relapse of the obstruction.

3 THE PRESENT STUDY

The original sample comprised 20 consecutive prepubertal children, 8 girls and 12 boys, with a mean age of 6 years (age range 4 to 9 years), who had either been referred to or who had independently sought medical attention at the Department of Otorhinolaryngology, Söder Hospital in Stockholm. All children had a typical history of OSAS and were treated with tonsillectomy and/or adenoidectomy. Adeno-/tonsillectomy was performed in 16 children, tonsillectomy alone in three children, and adenoidectomy alone in one child. An outline of the investigation materials of the Studies I, II, and III is presented in Table 2.

3.1 STUDY I

3.1.1 Aim

To evaluate prospectively the clinical manifestations, sleep recordings, and facial and dental development in children with OSAS before and at one year after tonsillectomy and/or adenoidectomy.

3.1.2 Material and methods

The children were their own controls. For reference purposes, the orthodontic variables were compared with mean values of well-known studies of dentofacial development and growth for age-matched groups (Moorrees, 1959; Bhatia and Leighton, 1993).

Signs and symptom

The parents were interviewed with regard to the children´s nocturnal breathing, daytime sleepiness, hyperactivity, enuresis, and upper airway infections. Health records with weight development from birth to the time when the patients entered the study were available. Weight was also recorded at the time of surgery. The size of tonsils and adenoids was

Table 2: Description of the investigation materials used in Studies I, II, and III.

Study		**Baseline** Mean age: OSAS 5.6 years Control 5.8 years			**1-year follow-up** Mean age: OSAS 6.8 years Control 6.8 years			**3-year follow-up** Mean age: OSAS 9.0 years			**5-year follow-up** Mean age: OSAS 10.9 years Control 10.7 years		
		Sleep recording	Study model	Lateral roentgen cephalogram	Sleep recording	Study model	Lateral roentgen cephalogram	Sleep recording	Study model	Lateral roentgen cephalogram	Sleep recording	Study model	Lateral roentgen cephalogram
I	OSAS (n=20)	X	X	X	X	X	X						
II	OSAS (n=14)	X	X	X	X	X	X		X	X			
	Control (n=14)		X	X		X	X						
III	OSAS (n=17)			X			X			X			X
	Control (n=17)			X			X						X

Study I: The 20 OSAS children were Swedish and caucasians. At the 1-year follow-up three patients droped out from the orthodontic registrations - one because of orthodontic treatment, one because of missing registrations, one that did not turn up.

Study II: It was possible to find only 14 control children without breathing obstruction to match 14 of the original 20 OSAS children. All controls were Swedish and caucasians.

Study III: The patients comprised the 17 OSAS children who were evaluated at the 1-year follow-up in Study I. The controls comprised 11 of the 14 children from Study II and 6 children from a British longitudinal study of dentofacial morphology. The British control children were caucasians.

scored by an otolaryngologist (tonsils on a 5-graded scale, and adenoids on a 3-graded scale).

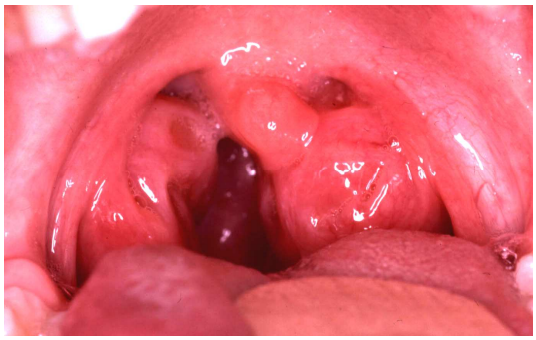

Figure 1a: Enlarged tonsils

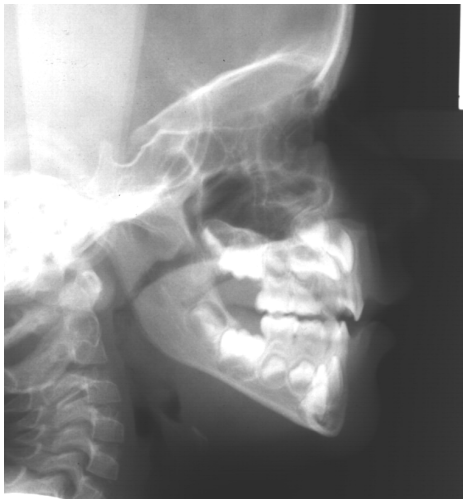

Figure 1b: Enlarged adenoids

Respiratory sleep recording

All 20 children underwent a whole-night polysomnography (PSG). Simultaneously ear oximetry, respiration and body movement (PVDF-matress) were recorded. At one year post-surgery, only the PVDF-oximetry sleep recording was carried out.

Orthodontic evaluation

All 20 children were examined orthodontically before surgery and 17 of them one year after surgery. Three patients dropped out, one because of orthodontic treatment, one because of missing registrations, and one patient who failed to appear at the 1-year follow-up examination. The children´s dental occlusions were clinically examined by an orthodontist. The registrations also included study models, lateral roentgen cephalograms and photographs. The dentofacial variables which were evaluated are presented in Table 5, and Figures 2 and 3.

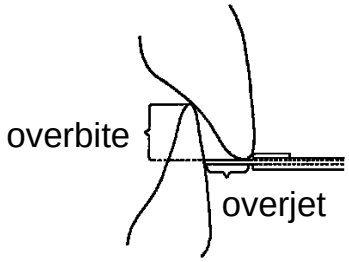

Figure 2: Measurements of overbite and overjet.

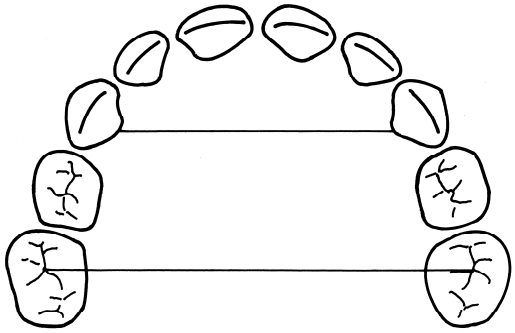

Figure 3: Width of the upper arch, measured between the centres of the cinguli at the gingival margine of the deciduous canines, and between the central fossae of the second deciduous molars.

Statistics

Non-parametric tests were used. The Spearman correlation test was used to analyse the degree of association between AHI and tonsillar-/adenoidal size. The Wilcoxon signed ranks test was used to compare the sleep recordings and orthodontic variables before and one year posttreatment.

3.1.3 Results

Signs and symptoms, and clinical examination

See Tables 3 and 4.

Table 3: Clinical daytime and nocturnal signs and symptoms in children (n=20) with a history of obstructive respiration during sleep.

Signs and symptoms	n
Snoring and apnoeas at night	20
Paradoxical inward rib cage motion	8
Excessive daytime sleepiness	14
Weight stagnation or weight loss	8
Enuresis	1

Table 4: PSG and PVDF-oximetry recording pre-operatively in children (n=20) with a history of obstructive respiration during sleep.

Respiratory sleep recordings:	n
AI>1	17
AHI>5	10
Signs of increased respiratory labour	19
Increased use of accessory respiration muscles in REM and slow-wave sleep	17
Poor sleep quality due to numerous arousals in connection with apnoea/hypopnoea and reduced time in REM and/or slow-wave sleep	9
Increased respiratory frequency (>20/min)	1
ODI ≥ 2	6

An investigation of a possible association between the severity of symptoms in the pre-operative respiratory sleep recording, the tonsillar-/adenoidal size, and the duration of the condition was carried out. No correlation between these variables was found.

One year after surgery, resolution of snoring and daytime symptoms was reported in all but one child, and the respiratory sleep recordings were improved in all but four children. Fifteen children had entirely normal recordings.

In comparison with the Swedish standard weight development curve (Engström *et al.*, 1973) the average weight of the OSAS children deviated from the normal value by -0.6 sd (range -2.2 to +2.2) before surgery. One year after surgery the deviation had been reduced to -0.2 sd (range -1.5 to +1.8) and this change was statistically significant.

Dentofacial recordings

Pre-operatively, there was no significant correlation between the ML/NL angle and the tonsillar-adenoidal size, the duration of symptoms, age when symptoms became manifest and degree of obstruction.

Pre-operativerly, the width of the upper dental arch was reduced and the ML/NL and ML/NSL angles were increased compared to standard values from longitudinal studies of dentofacial morphology (Moorrees, 1959; Bathia and Leighton, 1993). During the year after surgical treatment, the width of the upper arch increased and the ML/NL and ML/NSL angles decreased significantly ($p<0.001$) (Table 5). The combined effect of an anterior rotation of the mandible and a posterior rotation of the maxilla explains why ML/NL exhibited a more pronounced reduction than ML/NSL.

Table 5: Dentofacial variables. Pre-operative arithmetic means ($\bar{x}$) and ranges. Average changes ($\bar{d}$), ranges, and p-values during one year after surgery (n=17).

Variable	Pre-operative		Change during one year		
	$\bar{x}$	Range	$\bar{d}$	Range	*p*-value
Width between the second primary molars in the upper jaw (mm)	36.2	30.9-41.1	0.6	0.0-2.6	<0.001
ML/NL (degrees)	34.2	25.7-43.7	-3.1	-(1.0-6.1)	<0.001
ML/NSL (degrees)	38.1	29.9-44.9	-2.1	-(0.6-4.7)	<0.001

Initially 11 children exhibited a lateral crossbite. Orthodontic recordings of two of these patients were lacking at the follow-up evaluation (see page 21). In two of the remaining nine patients the malocclusion had resolved spontaneously one year after surgery.

3.2 STUDY II

3.2.1 Aim

1. To characterise the dentition and facial morphology of children with OSAS.
2. To study the time-linked effects on dentofacial morphology and respiration following elimination of the upper airway obstruction.

3.2.2 Material and methods

For 14 of the original 20 OSAS children (9 boys and 5 girls) in Study I, age and sex-matched controls with no signs of upper airway obstruction were available. The OSAS children were treated with adeno-/tonsillectomy to relieve the pharyngeal obstruction. In the OSAS group, sleep recordings were obtained before and at one year after surgery. Study models and cephalometric recordings were made before, and at one and three years after surgery. With the exception of the 3-year registration, the corresponding records were available in the controls.

Sleep recordings

For details see Study I.

Dentofacial recordings

On the study models overbite, overjet, width of the upper and lower dental arches between the deciduous canines and molars were measured (Figure 2 and 3). The occurrence of lateral crossbite was registered. On lateral cephalometric radiographs the angular variables and growth direction (facial axis) shown in Table 7 and Figure 4 were studied. In addition, lower anterior face height (LAFH) was registered as a percentage of the total face height.

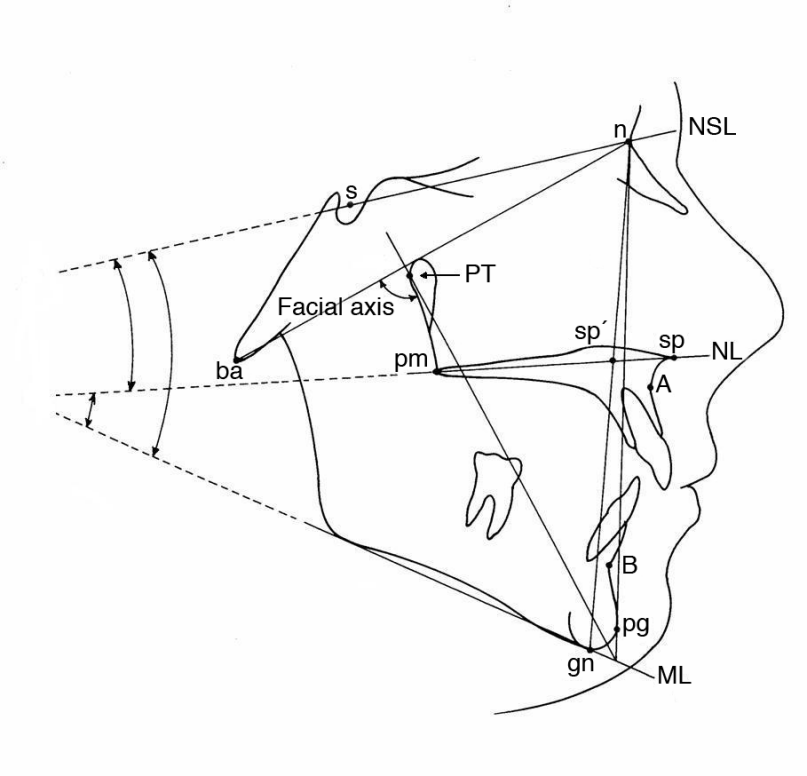

Figure 4: Reference points and lines on cephalograms. Definition of skeletal points and lines are those given by Björk (1960). The reference point PT and the angle "Facial axis" are those given by Ricketts (1981).

Statistics

Arithmetic means for each group at the different time points and mean differences between the paired samples before surgery were calculated. The differences between the groups before surgery, and within the groups during the first year after surgery were tested for statistical significance using the Wilcoxon signed ranks test.

3.2.3 Results

Sleep recordings

See Table 6.

Table 6: Polysomnographic variables before and one year after surgery. Mean values and ranges in OSAS children (n=14).

Variable	Pre-operative		One year after surgery	
	Mean	Range	Mean	Range
AHI	7.5	0-37	not measured	
Obstructive respiration in per cent of total sleep time	37.5	5-76	9.5	0-37
ODI	4.3	0-31	0.1	0-1

The changes in respiration pattern and ODI were statistically significant ($p<0.01$ and $p<0.05$, respectively).

Dentofacial recordings

During the first year after adeno-/tonsillectomy, dentofacial morphology developed favourably in the OSAS children. All variables in Table 7 that differed between OSAS children and the controls, changed in the same direction in both groups, although significantly more in the patient group, with the exception of the variable “lower anterior face height”.
Pre-operatively, seven OSAS children had a lateral crossbite, which had resolved spontaneously in two children one year after surgery. Among the controls at the initial registration, one child had a lateral crossbite that still remained one year later.

In the OSAS children, a further slight normalisation occurred during the following two years, but the changes during the first year post-operatively were more pronounced. There were no 3-year follow-up registrations for the control children.

Table 7: Dentofacial variables. Mean differences ($\bar{d}$), ranges and p-values in OSAS children compared to control children before surgery, and mean changes ($\bar{x}$) within the OSAS and control group, ranges and p-values during the first year after surgery.

	Pre-operative			One year after surgery			
Variable	$\bar{d}$ [1]	range	*p*-value	Group	$\bar{x}$ [2]	range	*p*-value
ML/NSL				OSAS	-2.5	-3.6 - -0.6	<0.001
	6.0	-3.6 - +15.4	<0.01				
(degrees)				Control	-0.3	-2.1 - +0.8	NS
NL/NSL				OSAS	0.8	-0.7 - +2.5	<0.01
	-2.2	-6.7 - +5.3	<0.05				
(degrees)				Control	0.6	-0.5 - +2.1	<0.05
ML/NL				OSAS	-3.5	-6.1 - -1.0	<0.001
	8.1	-3.2 - +15.4	<0.001				
(degrees)				Control	-0.8	-2.7 - 0.8	NS
Facial axis				OSAS	0.9	-1.5 - +2.6	<0.05
	-5.1	-11.6 - +5.0	<0.01				
(degrees)				Control	0.0	-1.6 - +1.3	NS
LAFH				OSAS	-0.5	-1.9 - +0.2	NS
	3.0	-1.7 - +7.4	<0.05				
(per cent)				Control	-0.4	-2.3 - +0.5	NS
Overbite [3]							
	-1.3	-4.1 - +1.9	<0.01				
(mm)							
Width 55-65				OSAS	0.7	0.0 - +1.4	<0.001
	-3.5	-10.1 - +0.6	<0.01				
(mm)				Control	0.4	0.0 - +1.1	<0.001

[1] A negative sign indicates a smaller value in the sleep apnoea children.
[2] A negative sign indicates a decreased value from pre-operative to one year after surgery.
[3] Due to loss of primary incisors in several children it was not possible to measure the variable overbite one year after surgery.

3.3 STUDY III

3.3.1 Aim

1. To compare young children suffering from OSAS with non-obstructed children, with respect to craniofacial morphology, soft tissue profile, and airway space.
2. To longitudinally evaluate the development of these structures after successful treatment of OSA (adeno-/tonsillectomy), and to undertake a comparison with the normal development in non-obstructed children.

3.3.2 Material and methods

The material comprised cephalometric records from 17 Swedish children (10 boys and 7 girls). All children had been diagnosed as suffering from OSAS as a result of enlarged tonsils and/or adenoids, and were to undergo tonsillectomy and/or adenoidectomy as treatment. The OSAS diagnosis was based on a typical history of sleep apnoea and was verified by polysomnographic registration (see Study I).

The control group comprised lateral cephalograms of 17 age and gender-matched children. The dental ages were also matched with one exception. Eleven of these children were Swedish and had been examined by an otolaryngologist who had reported no signs of obstructed upper airways. The remaining six children in the control group were selected from a longitudinal cephalometric growth study (Bhatia and Leighton, 1993). This material did not contain any subjects with facial deformity or severe malocclusion.

In the patient group, cephalographic records were available at baseline (=before surgery), and then at 1, 3, and 5 years post-operatively. With the exception of the 3-year registration, corresponding records were available for the control group. The cephalometric analyses carried out were based

on linear and angular measurements which had been obtained from standardised cephalograms (Figures 5 and 6). Skeletal, dental and soft tissue variables were studied and the development over the 5-year follow-up period was analysed.

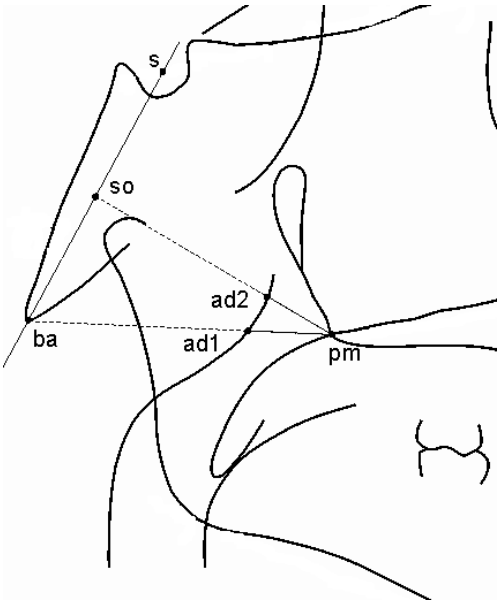

Figure 5: Nasopharyngeal airway space measured at the adenoids. Reference points ad1 and ad2 according to Linder-Aronson and Henriksson (1973).

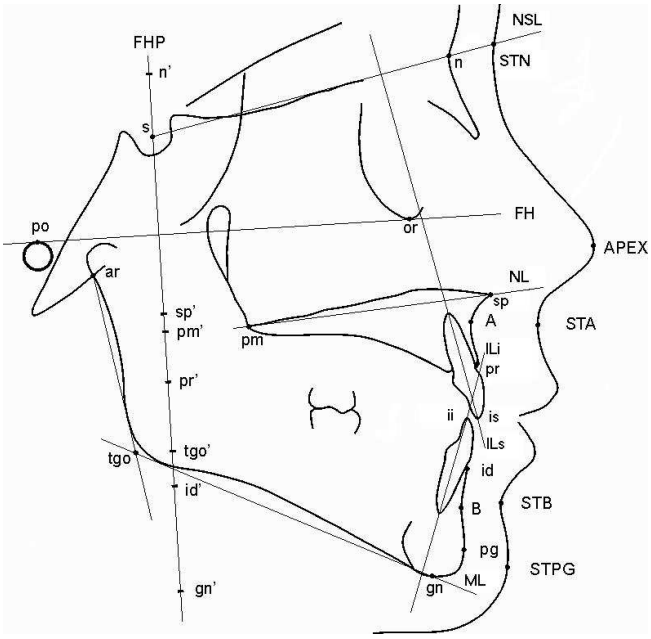

Figure 6: Reference points and lines on lateral roentgenographic head films. The definitions of reference points and the Frankfort Horizontal (FH) are those given by Björk (1960). Definitions of soft tissue points are given in Study III.

Statistics

The differences between the matched pairs at the baseline and the 5-year follow-up registrations were tested for statistical significance with the paired t-test. The differences in development over the five year period were tested for statistical significance (Global F-test) using the procedure for repeated measures (ANOVA). The significance level $p<0.05$ was chosen.

3.3.3 Results

The results are presented in Tables 8 and 9, and in Figure 7.

Table 8: Cephalometric angular (degrees), linear (mm), and ratio (per cent) variables at baseline in patients and controls, mean ages 5.6 and 5.8 years respectively.

Variable	n	Patients		Controls		Patients versus controls		
		Mean	SD	Mean	SD	Mean diff	SD	*P* value
Angular								
ML/NSL	17	38.0	4.41	33.5	4.42	4.5	6.50	0.012*
NL/NSL	17	4.2	2.12	6.7	2.19	-2.5	2.48	0.001***
ILs/NL	16	94.5	7.64	99.3	8.38	-4.8	8.66	0.044*
ILi/ML	11	83.6	7.70	89.6	4.74	-6.0	6.17	0.009**
Linear (skeletal)								
A-FHP	16	59.4	3.21	59.8	2.41	-0.4	4.01	0.718
B-FHP	16	53.5	3.59	54.5	3.75	-1.0	4.86	0.429
pg-FHP	17	51.4	4.73	53.7	4.89	-2.3	5.67	0.121
n-FHP	17	58.1	2.49	59.6	2.26	-1.5	2.17	0.009**
n´-sp´	17	38.6	2.95	39.7	3.25	-1.1	2.90	0.114
sp´-gn´	17	53.4	3.57	50.9	3.12	2.5	4.68	0.047*
n´-gn´	17	92.0	5.49	90.7	4.98	1.3	5.56	0.350
sp´-pr´	15	13.9	2.22	13.8	2.55	0.1	2.25	0.770
id´-gn´	9	21.8	1.72	21.6	2.56	0.2	2.74	0.769
s-pm´	17	32.6	2.06	32.0	2.04	0.6	2.47	0.317
pm´-tgo´	17	24.3	2.93	26.6	2.86	-2.3	4.15	0.041*
s-tgo´	17	56.9	4.01	58.5	3.34	-1.6	5.06	0.192
Linear (soft tissue)								
ad1-pm	17	11.6	4.88	14.2	4.65	-2.6	4.37	0.026*
ad2-pm	17	8.4	2.16	11.0	2.84	-2.6	2.44	<0.001***
STN-FHP	17	66.1	2.75	66.7	2.19	-0.6	2.75	0.368
APEX-FHP	17	78.9	3.72	81.0	2.60	-2.1	3.41	0.025*
STA-FHP	15	69.2	3.46	70.3	2.66	-1.1	3.77	0.283
STB-FHP	17	62.2	4.50	62.3	4.24	-0.1	5.08	0.910
STPG-FHP	17	61.7	5.11	63.7	4.95	-2.0	5.78	0.176
Ratio								
(sp´-gn´/n´-gn´) x 100	17	58.0	1.95	56.2	2.25	1.8	2.87	0.018*

* *P*<0.05; ** *P*<0.01; *** *P*<0.001.

At baseline (Table 8), the inclination of the mandible (ML/NSL) and maxilla (NL/NSL) differed significantly between the patients and controls. In the patients, the mandible was more posteriorly inclined (p<0.05) whereas the maxilla was more anteriorly inclined (p<0.001) compared to

the controls. The relatively greater inclination of the mandibular plane and relatively smaller inclination of the nasal plane in the patients was also reflected in the variables representing lower anterior (sp´-gn´) and posterior (pm´-tgo´) face heights. As compared with the corresponding dimensions in the controls, anterior face height was greater and posterior face height was smaller in the patients ($p<0.05$). The anterior facial ratio was significantly greater ($p<0.05$) in the patients. Furthermore, the length of the anterior cranial base (n-FHP) was significantly shorter ($p<0.01$) in the patients.

The upper and lower incisors were more retroclined in the patients than in the controls ($p<0.05$ and $p<0.01$, respectively).

As regards the facial soft tissues, the only difference was the slightly more advanced tip of the nose (APEX-FHP) in the control subjects ($p<0.05$). The width of nasopharyngeal airways was evaluated with the variables ad1-pm and ad2-pm. Both measurements were significantly reduced in the patients ($p<0.05$ and $p<0.001$, respectively).

At the 5-year follow-up (Table 9), only two significant differences were recorded between the groups. The length of the anterior cranial base (n-FHP) was shorter ($p<0.01$) and the position of the tip of the nose (APEX-FHP) was less advanced ($p<0.05$) in the patient group.

Table 9: Cephalometric angular (degrees), linear (mm), and ratio (per cent) variables at the 5-year follow-up in patients and controls, mean ages 10.9 and 10.7 years respectively.

Variable	n	Patients		Controls		Patients versus controls		
		Mean	SD	Mean	SD	Mean diff	SD	*P* value
Angular								
ML/NSL	17	34.4	4.91	31.9	4.97	2.5	7.62	0.184
NL/NSL	17	5.8	2.72	7.0	2.95	-1.2	3.49	0.182
ILs/NL	16	109.2	4.35	108.1	6.19	1.1	6.72	0.492
ILi/ML	11	91.9	6.54	91.9	7.79	0.0	8.35	0.999
Linear (skeletal)								
A-FHP	16	62.9	3.54	64.1	3.82	-1.2	5.19	0.348
B-FHP	16	59.0	4.47	59.7	5.32	-0.7	6.44	0.698
pg-FHP	17	58.9	5.05	60.2	6.32	-1.3	7.05	0.444
n-FHP	17	61.7	2.45	64.0	2.78	-2.3	3.17	0.008**
n´-sp´	17	45.2	3.20	46.1	2.87	-0.9	3.41	0.307
sp´-gn´	17	58.7	4.36	56.3	3.47	2.4	6.44	0.139
n´-gn´	17	103.9	5.37	102.4	4.77	1.5	6.53	0.343
sp´-pr´	15	13.2	2.17	12.8	1.85	0.4	3.14	0.612
id´-gn´	9	24.2	2.39	23.6	1.89	0.6	2.66	0.525
s-pm´	17	37.8	2.15	37.6	2.15	0.2	2.80	0.805
pm´-tgo´	17	29.2	3.36	29.4	3.37	-0.2	5.27	0.906
s-tgo´	17	67.0	4.06	67.6	4.36	-0.6	5.03	0.639
Linear (soft tissue)								
ad1-pm	17	17.5	4.43	19.7	5.46	-2.2	7.25	0.215
ad2-pm	17	14.3	3.35	15.4	3.99	-1.1	5.31	0.403
STN-FHP	17	70.2	2.80	71.3	2.90	-1.1	3.17	0.145
APEX-FHP	17	87.2	4.04	89.7	3.97	-2.5	4.63	0.045*
STA-FHP	15	76.0	3.59	76.5	4.31	-0.5	5.97	0.786
STB-FHP	17	68.7	5.24	68.5	5.56	0.2	6.20	0.896
STPG-FHP	17	69.7	6.51	70.3	6.73	-0.6	7.62	0.776
Ratio								
(sp´-gn´/n´-gn´) x 100	17	56.5	2.54	55.0	2.04	1.5	3.69	0.113

* $P<0.05$; ** $P<0.01$.

The statistical analyses (ANOVA test) of the growth pattern over the 5-year period showed that development of the variables ML/NSL, ILs/NL, and ILi/ML differed significantly (Figure 7). The inclination of the mandible was reduced in both groups but to a greater extent in the patients (-3.6 degrees compared with -1.6 degrees in the controls). At baseline, the upper and lower incisors were retroclined in the patients compared to the

controls. At the 5-year follow-up, the inclination of both upper and lower incisors was very similar in the two groups.

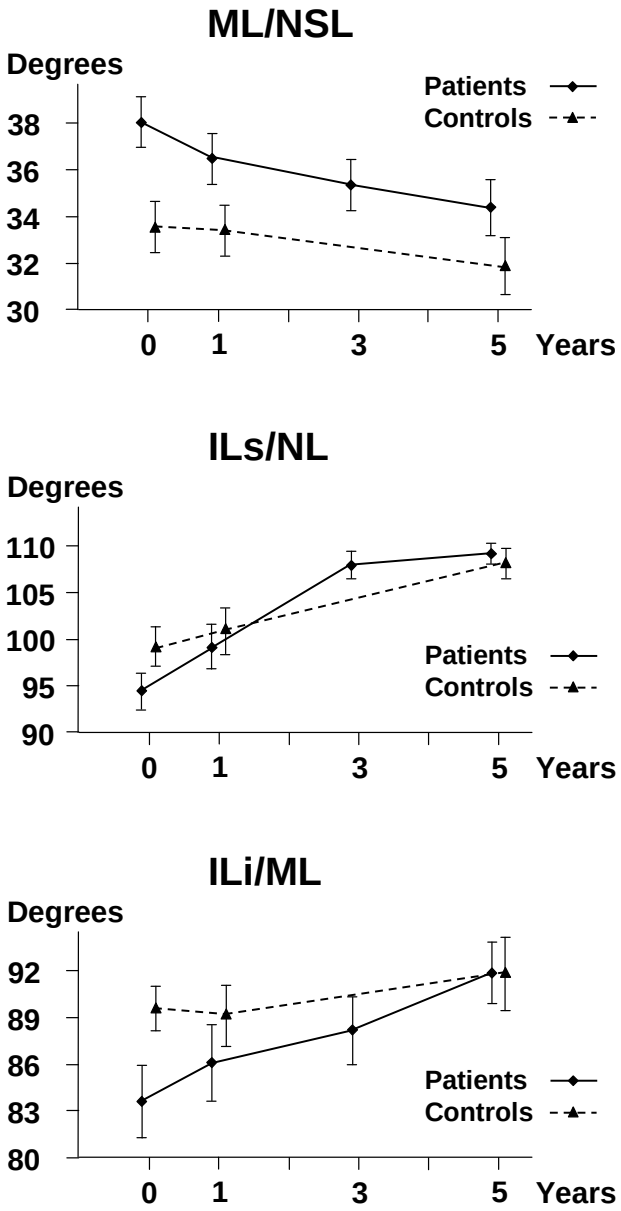

Figure 7: Diagrams illustrating the means, standard errors, and the changes of the variables ML/NSL, ILs/NL, and ILi/ML during the 5-year follow-up in the OSA and control groups. In order to avoid overlapping of the error lines, the breakpoints of the curves have been slighly displaced in relation to each other.

The pattern of change for all other variables was similar in the two groups.

4 DISCUSSION

In the present prospective investigation, OSA caused by adenoid and tonsillar hypertrophy in young children has been studied. The following questions were of interest:

- Does adeno-/tonsillectomy normalise the breathing function and resolve the day and night-time symptoms of OSA?
- Does dentofacial morphology differ between OSAS children and children whose airways are not obstructed?
- How do the dentofacial structures develop after surgical treatment of OSA?

Obstructive sleep apnoea

In the wide spectrum of symptoms of upper airway obstruction, OSA is the most extreme variant of mouth breathing and snoring. Potential causes for OSA in children are enlarged tonsils and adenoids, which reduce upper airway space and may make normal breathing difficult.

The initial criterion for the possible inclusion of the patients who were evaluated in this study, was that the parents had reported typical symptoms of obstructive sleep apnoea syndrome in their children. Using a combination of laboratory diagnostic indicators, the diagnosis OSAS was then verified in the 20 consecutive young children (12 boys and 8 girls, mean age 5.6 years) who eventually comprised the patient group.

It would have been desirable to obtain an age and sex-matched control group of healthy children without nocturnal respiratory disturbance, simultaneous to the collection of the OSAS children. From an ethical point of view, however, this was not possible, and therefore the control groups had to be selected from previous longitudinal studies of dentofacial

morphology. Due to this fact, the control group records were about 20 to 30 years older than those of the OSAS children. During that time span, it is possible that minor secular changes in dentofacial morphology have occurred, for example due to variations in frequency of allergic condition (Bresolin *et al.*, 1983) and caries (von der Fehr, 1994). Although such changes should not be disregarded, it is not likely that they would have any significant influence on the results of the present study.

The surgical treatment performed in the OSAS children was tonsillectomy and adenoidectomy in 16 patients, tonsillectomy alone in three patients and adenoidectomy alone in one child. The three children who dropped out of the study were all treated with adeno-/tonsillectomy. In study II, the investigation material comprised only 14 patients due to difficulties in finding a sufficient number of matching controls.

The adeno-/tonsillectomy gave an immediate relief of symptoms and still one year after surgery, none of the patients exhibited any subjective signs and symptoms of OSAS. However, five children showed signs of obstructive respiration in their PVDF-oximetry records one year after surgical treatment. On lateral roentgen cephalograms, it was observed that two of these children had a somewhat enlarged adenoid tissue which spontaneously diminished before the 5-year follow-up. In two others, an enlargement of the adenoid tissue had continued however, and before the 5-year follow-up they were subjected to an additional adenoidectomy. In one child, no tissue enlargement was observed on the lateral cephalogram. At the 5-year follow-up there was no significant difference between patients and controls in respect of width in the naso-pharynx measured as the distances ad1-pm and ad2-pm.

Airway obstruction which results in mouth breathing, may in turn lead to an extended head posture, and a lowered mandible, hyoid bone, and tongue position. Some of the present OSAS children were videotaped simultaneously with the sleep recordings. Activation of accessory respiration muscles and extended head posture were particularly obvious during REM and slow-wave sleep. This is in agreement with Guilleminault *et al.* (1981) who stressed that the severity of the respiratory obstruction in children may rather be reflected in respiratory labour than in the number of apnoeas/hypopnoeas or the degree of oxygen desaturation in the blood.

It is likely that this increased respiratory labour and a presumably higher energy consumption during sleep is responsible for some of the present OSAS children´s failure to thrive. However, these children rapidly gained weight after the adeno-/tonsillectomy had normalised their breathing pattern. This development is in agreement with the results of the study by Marcus *et al.* (1992). The failure to thrive could also be related to unfavourable variations of growth hormone (GH) levels as described in studies of OSAS in children (Bar *et al.*, 1999; Nieminen *et al.*, 2002; Peltomäki, 2007). Secretion of GH is associated with the sleep-wake cycle, and is assumed to increase during slow-wave and REM sleep. GH levels have not been recorded in the present study. However, in nine children numerous arousals in connection with apnoea/hypopnoea and reduced time in REM and/or slow-wave sleep were observed. Bar *et al.* (1999) and Nieminen *et al.* (2002) have shown that improved respiration after adeno-/tonsillectomy has resulted in increased level in the serum of GH mediators and catch-up growth.

The risk of OSA symptoms reappearing should not be disregarded and especially not in cases where the upper airways are narrow. In subjects

with a reduced nasal-pharyngeal dimension even a small enlargement or decreased patency in the pharyngeal tissues may trigger a relapse in obstructive breathing. Guilleminault *et al.* (1989), and Guilleminault and Stoohs (1990) have reported the existence of such a risk in boys during puberty. Shintani *et al.* (1998) mentioned that the possibility of subclinical neuromuscular dysfunction may reduce the degree of breathing improvement after adeno-/tonsillectomy. Rosen *et al.* (2001) suggested that obese children, and those with a family history of breathing disorders during sleep were at higher risk of developing residual OSA after adeno-/tonsillectomy. These are important issues which indicate that possible residual obstructive breathing problems after adeno-/tonsillectomy should always be carefully monitored.

Dentofacial morphology

The present study has shown that OSAS children in comparison with controls exhibited significant differences in the dentition, such as reduced overbite, narrow upper dental arch, crossbite and/or crossbite tendency, and retroclined incisors. Similar differences in slightly older children have previously been reported in the literature (Linder-Aronson, 1970; Behlfelt, 1990) to be associated with mouth breathing, extended head posture, and narrow upper airways. It was interesting to note, however, that the deviations from normal values were large enough to reach statistical significance in the present patients even at the early ages that they represented.

The developmental pattern of the upper dental arch and the occlusion changed in a favourable way after surgery. Between 5 and 6 years of age, and between 6 and 7 years of age, the normal average increase in width between the upper second deciduous molar is 0.4 mm and 0.3 mm respectively (Moorrees, 1959). This is in accordance with the average

increase in the control group. In the patient group, the width of the initially narrow upper arch increased by 0.7 mm ($p<0.001$) during the first year post-surgery. During the subsequent two years, the increase was 0.4 mm per year. It was not surprising, therefore, that the lateral crossbite had resolved spontaneously in two patients. The inclination of the incisors showed an even more positive development. At the end of the 5-year follow-up period, the average inclination of both upper and lower incisors was normal. The OSA diagnosis had been established and treatment carried out when most of the patients had only deciduous teeth. At the end of the 5-year follow-up period, the deciduous incisiors had exfoliated and the permanent incisors erupted. Due to the successful treatment of the patients, the permanent incisors could erupt in a normally functioning environment, and this fact may explain the result that the average inclination of both upper and lower permanent incisors was completely normal after treatment.

The OSAS children had a significantly different facial morphology before treatment compared to age and sex-matched non-obstructed controls. The lower anterior face height (sp´-gn´) was greater, the mandible (ML/NSL) was posteriorly inclined, the maxilla (NL/NSL) was anteriorly inclined, and the inclination of the facial axis in the OSAS children before surgical treatment indicated that they exhibited a more vertical growth direction in general than the controls. After treatment for OSA, the patients´ dentofacial growth was normalised. This development is in agreement with other studies on dentofacial growth following treatment for obstructive breathing in children who were 4 to 5 years older than the children in the present study (Linder-Aronson, 1975; Behlfelt, 1990).

It has been reported that both children with obstructed upper airways and adult apnoeics exhibit some degree of maxillary and mandibular

retrognathism (Linder-Aronson, 1970; Jamieson *et al.*, 1986; Lowe *et al.*, 1986; Guilleminault *et al.*, 1989; Behlfelt, 1990; Solow *et al.*, 1996; Shintani *et al.*, 1998; Finkelstein *et al.*, 2000; Kawashima *et al.*, 2002). A tendency for retrognathism was also recorded in the present study although the differences between patients and controls did not reach the level of significance. This is in line with the findings of Shintani *et al.* (1997) who compared OSAS children in different age groups with control children without airway obstruction. They found no difference between the youngest OSAS children and their controls regarding the SNA and SNB angles, while in the age groups 5 to 6 and 7 to 9 years OSAS children exhibited reduced SNA and SNB angles compared to controls.

As regards the length of the anterior cranial base (n-FHP), it was interesting to note that it was significantly shorter in OSAS children than in controls. Since the spheno-ethmoidal synchondrosis in the youngest children was probably not yet ossified before treatment, it could be expected that there would also be an increased growth in this area after treatment for OSA. However, at the 5-year follow-up registrations the difference between patients and controls in respect of cranial base length was still present. In the adenoid children studied by Linder-Aronson (1970), a similar but not significant trend was found. A short cranial base in OSAS children has also been reported by Shintani *et al.* (1997) and Kawashima *et al.* (2002). In a study on adult apnoeics, Andersson and Brattström (1991) showed that the cranial base was shorter in the apnoeics than in controls without snoring problems, but SNA and SNB angles did not differ between these groups. Further studies are needed in order to establish whether a short anterior cranial base is a significant characteristic in the craniofacial morphology of apnoeic patients.

The morphology of the soft tissue profile was evaluated on the basis of measurements of the distances from FHP to five reference points, namely the soft tissue nasion (STN), the tip of the nose (APEX), and the soft tissue A, B, and Pg points. It was found that the tip of the nose was significantly less prominent in the patients than in the controls ($p<0.05$). The other distances exhibited a similar tendency but the deviation of the patients´ values from the control values was small and did not reach the level of significance. However, the tendency for patient retrognathism noted in respect of skeletal morphology was also reflected in the soft tissue profile. After treatment, the tip of the nose was still significantly less prominent in the patients than in the controls. No differences between the groups were found as regards the position of the soft tissue A, B, and Pg points in relation to FHP.

The observed change in growth direction in the OSAS children can be explained by a differential amount of growth in the anterior and posterior part of the facial skeleton. The ratio (sp´-gn´/n´-gn´) x 100 indicated that the lower anterior face height in relation to total anterior face height was greater ($p<0.05$) in the patients than in the controls, the difference being 1.8 units. Five years post-treatment, the mean ratio in the patients approached that of the controls, but a difference of 1.5 units still existed. Posteriorly, on the other hand, the lower face height (pm´-tgo´) before treatment was 2.3 mm smaller, on average, in the patients than in the controls. Five years after treatment, the difference between patients and controls was only -0.2 mm. This means that in comparison with the controls the posterior face height in the patients had increased significantly more. Taken together, these growth events resulted in an average anterior rotation of the mandible in the patient group which was most pronounced during the first year post-treatment and significantly greater than the average mandibular rotation recorded in the control group. A similar

development has been observed in children after tonsillectomy (Behlfelt, 1990).

The increase in posterior face height might be explained either by changed position of the condyle, growth in the condylar cartilage that increases mandibular ramus height, or bone apposition at the gonial angle, or by a combinations of these alternatives. A possible growth stimulation in the condylar cartilage could very well be the result of a favourable change in the GH balance following the normalisation of the patients´ sleep. The variations in the mandibular growth pattern seen in the OSAS patients, therefore, could be explained by the earlier discussed failure to thrive, and the catch-up growth after adeno-/tonsillectomy, and the possible association of these conditions with secretion of GH. In studies on patients suffering from Turner´s syndrome (Rongen-Westerlaken *et al.,* 1993), GH therapy has been shown to have a favourable effect on mandibular growth. In young patients with GH deficiency due to total body irradiation (TBI), administration of GH has been observed to stimulate mandibular growth and result in an upward and backward displacement of the mandibular condyle (Forsberg *et al.,* 2002). Furthermore, the mandibular length increased significantly more in GH treated patients than in controls.

Additional studies of factors which may possibly determine the growth and shape of the mandible, could further elucidate these issues.

5 CONCLUSIONS

- Children exhibiting symptoms of OSAS should be given an examination based on a combination of laboratory diagnostic tests, EMG activity in relevant muscles, arousals, and oxygen saturation.
- OSA in young children caused by hypertrophic adenoids and/or tonsils was succesfully treated with adeno-/tonsillectomy.
- Young children suffering from OSA have a different dentofacial morphology compared with non-obstructed control children.
- Dentofacial morphology was normalised after the treatment of OSA.
- It is important that children with OSA are diagnosed early and evaluated both from a medical and dentofacial point of view. This requires close co-operation between paediatricians, otolaryngologists, orthodontists and paedodontists.

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