CASE WESTERN RESERVE UNIVERSITY

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We hereby approve the thesis of

Randall William Kulnis

candidate for the degree Master of Science

in Dentistry.

Signed: _____
(Chairman)

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CEPHALOMETRIC ASSESSMENT OF SNORING AND NON-SNORING CHILDREN

by

RANDALL WILLIAM KULNIS

Submitted for partial fulfillment of the requirements for the

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Thesis Advisor: Suchitra S. Nelson, Ph.D.

Department of Orthodontics

School of Dentistry

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CEPHALOMETRIC ASSESSMENT OF

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Abstract

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RANDALL W. KULNIS

It is estimated that as many as seventy percent of adults with obstructive sleep apnea syndrome (OSAS) snored during childhood They are at increased risk for hypertension, cardiovascular disease, cerebrovascular disease, and impaired function due to sleepiness. It is unknown if children that snore have radiographic characteristics that might indicate a predisposition to OSAS later in life. Therefore, the purpose of the present study was to test if there are differences in craniofacial variables between snoring and non-snoring children.

Sleep behavior questionnaires were presented to the parents/guardians of children (ages 7-14) treated at CWRU Dental School and local Cleveland orthodontic practices. From 228 completed surveys, thirty-one experimental subjects were identified. Each snoring subject was matched to a non-snoring child of the same gender, race and approximate age. The investigator traced the thirty-one snoring and thirty-one control subject radiographs, unaware of the snoring background of the

subjects. Eleven linear and one angular measurement were recorded and differences between the groups were examined.

An analysis of the results revealed significant differences between snoring and non-snoring children. Snoring children tended to have a greater Body Mass Index. Comparison of cephalometric variables demonstrated that the hyoid bone was positioned more inferiorly to the mandibular plane (H-MP) in snoring children. Also, the superior and most narrow posterior airway spaces (S-PAS and N-PAS) were significantly more constricted in snoring children, compared to non-snoring children. The sagittal plane measurements, including the posterior nasal spine to A point (PNS-A), sella to nasion (SN), and gonion to gnathion (Go-Gn) were shorter in snoring children. There was also a strong tendency for the vertical airway length (VAL) of snoring children to be longer.

Evaluation of questionnaire data revealed that snoring children were at significantly greater risk of: Mouth breathing, sleeping with their head tipped back, falling or staying asleep, and restless sleep.

Therefore, snoring children present craniofacial features that differ from non-snoring children of the same age, gender and race. Consideration of specialty referral of snoring subjects for evaluation of the airway might be warranted.

DEDICATION

To those that have influenced my career in a most positive direction: William J. Kulnis, Sr (Dad), Mary Jane Kulnis (Mom), Dr James R. Paige, Mr Robert E. Kwiat, my wife Debbie and children, Jaison, Ashley and Kadena. You each gave me the encouragement I needed, at the most critical of times, to persevere in the pursuit of my goals, regardless of the obstacles.

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GLOSSARY

Definitions of Anatomic Landmarks

Point/Line	Definition
A	A-point, subspinale: The deepest point in the concavity of the anterior maxilla between the anterior nasal spine and the alveolar crest
Ar	Articulare (Bjork): A point located at the intersection of the lateral radiographic image of the posterior border of the ramus with the base of the occipital bone
B	B-point, supramentale: The deepest point in the concavity of the anterior mandible between the alveolar crest and pogonion
Ba	Basion: The most posterior-inferior point on the clivus
Eb	Base of the epiglottis: The deepest point of the epiglottis
FOP	Functional occlusal plane: A line through PreM and Mo biscecting incisal overbite
Gn	Gnathion: Constructed point determined by bisecting the angle between the tangent of pogonion and the tangent of menton
Go	Gonion: Constructed point determined by bisecting the angle between the line from Me-MIB and a line tangent to the posterior border of the ramus passing through articulare
H	Hyoidale: The most superior-anterior point on the body of the hyoid bone
MCF	Middle cranial fossa: Point represented 40.3 degrees anterior to the line joining Bjork's articulare (Ar) and SE
Me	Menton: The most inferior point on the bony chin

MIB	Mandibular inferior border: Most inferior border of the mandible
Mo	Molar: The mesial-buccal cusp tip of the upper first molar
N	Nasion: Junction of the frontal and nasal bones at the naso-frontal suture
P	Palate: The most inferior point of the soft palate
Phwl	Inferior posterior pharyngeal wall: Junction of the posterior pharyngeal wall and the B-point to gonion plane
PhwN	Most narrow posterior pharyngeal wall: Junction of the posterior pharyngeal wall and a line parallel to the B-Go plane at the most narrow point in the airway inferior to PNS and superior to Go
PhwS	Superior posterior pharyngeal wall: Junction of the posterior pharyngeal wall and a line parallel to the FOP at the level of Psp
PM	Vertical PM: A line from SE through the averaged inferior most points of the left and right pterygomaxillary fissures
PNS	Posterior nasal spine: Process formed by united projecting medial ends of the posterior borders of the palatine bones
PreM	Premolar: The buccal cusp tip of the upper first premolar
Psp	Posterio-superior palate: The most posterior-superior point on the soft palate
Ptm	Pterygomaxillary fissure: The averaged inferior most point of the left and right pterygomaxillary fissure
S	Sella turcica: The center of the hypophyseal fossa lodging the pituitary body
SE	Sphenoethmoidal junction: The junction between the averaged right and left greater wings of the sphenoid bone and the floor of the anterior cranial fossa

STBn	Soft tissue border: The most anterior point of the airway on the soft palate or tongue between PNS and Go at the most narrow point of the pharynx
STBi	Soft tissue border: The most anterior point of the airway on the soft palate or tongue between PNS and Go on the B-Go plane
Tt	Tongue tip: The most anterior point of the tongue which touches the lingual surface of the mandibular incisors

Description of Cephalometric Measurements

Variable	Interpretation
H-MP	Measurement from the most anterior-superior point on the hyoid bone (hyoidale) perpendicular to the plane from menton (Me) to the inferior border of the mandible (MIB).
P-PNS	Length of the soft palate.
VAL	Vertical Airway Length: The linear measurement from the posterior nasal spine (PNS) to the base of the epiglottis (Eb)
BaN	Linear measurement from basion (Ba) to nasion (Na).
SN	Linear measurement from sella (S) to nasion (Na).
A-PNS	Anterior-posterior length of the maxilla measured from A-point (A) to the posterior nasal spine (PNS).
TL	Tongue length measured from the base of the epiglottis to the most anterior point of the tongue which touches the lingual surface of the mandibular incisors.
S-PAS	The width of the pharyngeal airway at the level of the most posterior-superior point on the soft palate (Psp). Measured parallel to the functional occlusal plane (FOP) from (Psp) to the superior posterior pharyngeal wall (PhwS).
I-PAS	The width of the pharyngeal airway at the level of the plane from B-point (B) to gonion (Go). Measured from the inferior posterior pharyngeal wall (PhwI) to the most anterior point in the airway on the soft palate or tongue (STBi).

N-PAS	The width of the pharyngeal airway at it's most narrow point inferior to the posterior nasal spine (PNS) and superior to gonion (Go). Measured parallel to the plane from B-point (B) to gonion (Go) from the posterior pharyngeal wall (PhwN) to the most anterior point in the airway on the soft palate or tongue (STBn).
Go-Gn	Effective length of the body of the mandible measured from gonion (Go) to gnathion (Gn).
MCF	Measurement of the position of the maxilla and mandible relative to the subject's vertical line (PM) from the sphenoethmoidal junction (SE) passing through the pterygomaxillary fissure (Ptm). A line from articulare (Ar) to SE represents the posterior part of the cranial floor and intersects with the vertical PM. A neutral angle at the intersection of the cranial floor and the vertical PM is 40.3°. A vertical PM located anterior to the "neutral" angle represents a forward alignment of the middle cranial fossa and results in a protrusively positioned maxilla. A vertical PM located behind the "neutral" angle represents a posteriorly positioned middle cranial fossa. As a result, the mandible is positioned protrusive. When the subject's vertical PM is coincident with the "neutral" angle, neither a maxillary nor mandibular protrusion effect is present.

INTRODUCTION

Unlike adults, it is very difficult to identify children at risk for developing airway disease. Clearly recognized in adults, and supported by the literature, obstruction of the airway can predispose one to debilitating disease. The relationship between specific risk factors associated with snoring and airway obstruction in children, and their affects on cardiovascular and respiratory control, excessive sleepiness, cognitive, behavioral, and neuropsycological behavior need further investigation. In children, little is known about how development affects airway anatomy, neuromuscular control of the pharynx, and airway regulation. In addition, the influence of gender, obesity and race warrant further study in children with ventilation disturbances.

Criteria for identifying children with clinically significant airway problems need to be established. If one supports a genetic basis for abnormalities in ventilation, it might be expected that anatomic differences exist between subjects predisposed to airway disease and non-affected subjects. Differences in craniofacial structure, soft tissue structures, body mass, and neural control of the airway can be expected. It has been demonstrated in the literature that most apneic adults, snored as children. The potential for obstruction of the airway is greatest in snorers, and pathology can result when airway obstruction exists. Early recognition of those

predisposed to obstruction, might result in intervention that would significantly reduce the development of debilitating disease in the future. Therefore, it is significant to test for anatomic differences between snoring and non-snoring children.

Epidemiologic study of habitual snoring in children between four and five years of age, suggests a prevalence of as high as 12.1%. Snoring children also exhibited restless sleep, hyperactivity and excessive daytime sleepiness (Ali et al. 1993). A similar prevalence (11%) was observed by Owen et al. in 1996. Owen recognized peaks in snoring at ages four and nine The peaks were speculatively associated to lymphoid hypertrophy and allergic rhinitis, respectively. Corbo et al. investigated the prevalence of snoring "often" in children from ages six to thirteen. They found a prevalence of 7.3% among the subjects. A decrease in snoring with increasing age was observed. The decrease was attributed to an increase in pharyngeal airway dimension with growth (Corbo et al. 1989).

Although there have been a number of studies on the etiology and pathogenesis of snoring, the exact mechanisms that lead to snoring are not clearly defined. Studies have investigated the anatomy and physiology of the pharynx, turbulence of airflow within the airway and irritant receptors in the larynx. The affect of blood gases on nerve centers and ultimately pharyngeal muscle tone have been researched; as have the affects of varying lung volumes on airway dimension.

It appears that both genetic and environmental factors influence snoring, and most studies support an anatomic etiology. Most investigators observed the soft palate to flutter against a constricted airway. In situations of airway obstruction, the

blockage is often located at the level of the soft palate; but has also been identified elsewhere within the entire extent of the pharynx.

Attempts to measure snoring subjectively and objectively have been limited in their accuracy, however, the standard for differentiating apneic subjects from non-apneic is to study sleep using polysomnography. But, difficulty is encountered when trying to determine the presence of apnea in children. Compared to apneic adults, obstructive episodes are uncommon, and blood oxygen saturation levels rarely decline in children. Therefore, one should be cautioned against using adult criteria in the diagnosis of obstructive sleep apnea in children.

Several studies have used cephalometrics to examine for differences in snoring and apneic subjects. Most frequently, cephalometric radiographs of adults with apnea were compared to radiographs of non-apneic adults. The tendency was for apneic adults to be older males. Apneic adults also tended to have an increased hyoid to mandibular plane distance, longer soft palates, a diminished sagittal cranial base dimension and narrowed posterior airways. Few studies have investigated differences in cephalometric features in children. However, Shintani et al. (1992) found that children with apnea had lower hyoid bones. In the most severe apneics, the children presented enlarged adenoids and narrow airways.

The present study was conducted to determine if there are differences in craniofacial features between snoring and non-snoring children using measurements made from cephalometric radiographs.

REVIEW OF THE LITERATURE

Prevalence

Snoring was once considered to be harmless and only an annoyance (Kleitman 1963). Years later, the significance of snoring and its role in health and daytime functioning has begun to be recognized. Today, snoring is a factor in a spectrum of medical disorders that have effects that range from no physical debilitation to failure to thrive.

In 1978, Lugaresi et al. first reported that the prevalence of snoring increased across the life-span. Fifteen years later, Lugaresi et al. more specifically stated that greater than 20% of adults snore, and in men over forty, 60% are snorers (Lugaresi et al. 1993). In 1991, a cross-sectional study investigating the prevalence of snoring and apneas in 5,201 adults over sixty-five years of age was conducted. The results of that study revealed that men and women aged 65-69 reported that "others" had complained of their loud snoring at a prevalence of 41% and 20%, respectively. In contrast to the statement that snoring increased across the lifespan made by Lugaresi et al. in 1978, snoring prevalence in these subjects did not appear to increase with age. In fact, the prevalence of snoring in those over the age of 80 was reported to decrease 24% in men, and 10% in women (Enright et al. 1996).

The Enright et al. 1996 study was dependent upon self-reports. Wiggins et al., in 1990, compared self-reported snoring to that reported by the spouse. The author concluded with the opinion that the spouse was of greater validity than that of the sleeping subject. A Finnish study that relied upon self-reported data was conducted by Martkainen et al. in 1994. The self-reports revealed a peak snoring prevalence in the 50-60 year-old age group. The study compared the change in snoring prevalence taking place between 1985 and 1990. The study questioned men and women between the ages of 36 and 50. Five years later, the same cohort was questioned again. They found the prevalence of snoring increased in both men and women when they reached 55 years of age. A weakness of the study, besides self-reporting, was a very narrow subject age range of fourteen years. Over the five year span of the study, no one under 36 or over the age of 55 was surveyed, and a decrease was not observed in snoring prevalence up to that particular age of maturity (Martikainen et al. 1994). Janson et al. also recognized a rise in prevalence with increasing age, male gender and body-mass index. However, the age of the male and female subjects in the study was again too narrow (ages 20-44) to detect significant age related changes (Janson et al. 1995). Honsberg et al. surveyed 1,476 U.S. subjects 18 years of age or older in 1985; then again in 1990 (nearly six years apart). In that study, self-reported habitual snorers were determined based upon their answer to the question "How often do you snore loudly?" Response options included (1) every night, (2) most nights, (3) some nights, (4) rarely, (5) never, and (6) do not know. Subjects responding with every night and most nights were categorized as "habitual snorers". Of an initial 158

snorers, 58.2% persisted after six years, and 35.5% remitted. In line with the results of Enright et al., Honsberg et al. also observed that snoring remission was significant in the elderly. In the over age 65 population, snoring remission was 53.3%. No mention was made as to the specific ages of the older group. The most elderly group was classified only as "over 65" (Honsberg et al. 1995).

Being aware that loud snorers and sleep apneics as a group frequently have associated physiologic risk factors, the observed decline in prevalence with increasing age may be a reflection of survival bias In a sample of 441 subjects surveyed in Australia between the ages of thirty-five and sixty-nine, those that reported a history of snoring "often" or "always" were categorized as "snorers". Again, a trend of snoring with increasing age was observed. Increasing age "should be considered a significant predictor of snoring" according to the authors. Of even greater significance and considered by the authors to be "the most powerful predictor of snoring" was male gender. (Olsen et al. 1995).

Guilleminault and Dement studied thirty loudly snoring patients (ages twenty-three to sixty-five) referred for evaluation of daytime sleepiness. All but one patient had a history of snoring initiated before the age of twenty-one. Forty-eight percent had a snoring history that went back to "a very young age" (Guilleminault and Dement 1978).

When investigating the presence of snoring in the children and adolescents much less research has been conducted. In the opinion of Westbrook from the Mayo Clinic, snoring, not associated with upper respiratory infection, is not normal in

children. He further stated that chronic snoring in children is "cause for concern", possibly indicating impaired inspiration during sleep. He concludes that snoring in children warrants examination (Westbrook 1983). In 1993, Ali et al. surveyed 782 British parents regarding their child's sleep behavior, 95 (12.1%) reported that their 4-5 year old child snored on most nights. These children were considered "habitual snorers". Compared with children of the same age that never snored, it was significantly more common for "habitual snorers" to experience restless sleep, hyperactivity and daytime sleepiness (Ali et al. 1993). A year later in 1994, Ali again surveyed 507 of the same 782 parents surveyed in 1993. Although snoring resolved in nearly half of the children, the prevalence remained close to the same (11.4%) secondary to the emergence of new snorers. As was the case in Ali et al.'s 1993 study, restless sleep, hyperactivity and daytime sleepiness were still common to the habitual snorers (Ali et al. 1994). In Italy, Corbo et al. questioned the parents of 1615 children aged 6-13 and found that 7.3% reportedly snored "often". They also noted a significant decrease in snoring with increasing age, which was attributed to an increase in the size of the pharynx in older children (Corbo et al. 1989). A more recent study from the United Kingdom surveyed the parents of 245 randomly selected children up to the age of ten. Parents were asked if their children snored and were to respond to the question with one of the following answers: Never, rarely, sometimes or often. The definition of snoring used in the study included children that were reported to snore both "sometimes" and "often". According to the definition of snoring used in the study, children snored at a prevalence of 27%. The prevalence of

children reported to snore "often" only was 11%. Within the study's definition of snoring, no significant differences were found between boys and girls; however, age did appear to be a factor. Regardless of sex, prevalence of snoring peaked around four years of age, and then again at 8-9 years of age. The peaks were related to adenotonsillar hypertrophy and allergic rhinitis, respectively. When comparing snoring and non-snoring children, restless sleep, snoring with a cold and poor hearing reports by parents were of the greatest significance (Owen et al. 1996).

A review of the studies conducted on snoring prevalence revealed great variation and were dependent on the definition of snoring used and the particular characteristics of the population studied.. Data on snoring has frequently been measured using self-reported or informant-reported responses to a variety of questionnaires. The studies have sampled populations of different age ranges and other demographics, different geographic locations, and varying social environments.

Bliwise et al. evaluated the relative validity of self-reported snoring and recognized the degree of difficulty involved in constructing truly objective measurements for comparison. Considering the decline in self-reported snoring in some studies with very old age, the question arose regarding the subject's perception concerning their own sleep related behavior. Of the elderly, how many have spouses available to report observations? Since male predominance of snoring is unquestioned, Bliwise et al. asked if women were less accurate in snoring self-assessment. Other questions, possibly affecting data validity, involved the accuracy of data from subjects living alone, and those prone to deny or exaggerate responses

(Bliwise et al. 1991). Young summed up the difficulty in obtaining measurement validity best when stating "validity is likely to vary by demographic, psychosocial and other factors, such as age, sex, ethnicity, health status, and household composition" (Young 1991)

Pathophysiology of Snoring

The association between a history of heavy snoring and obstructive sleep apnea syndrome, first recognized in the 1960s, suggested a pathologic relationship. Sleep apnea has been generally recognized as a cessation of breathing and can vary in definition and severity. Subsequent studies, relevant to snoring, strongly suggested that the presence of snoring could result in a myriad of adverse medical sequale of which apnea was included. With the increased use of polysomnography in the 1960's for the study of sleep behavior, the pathophysiologic consequences of snoring were recognized as important (Gastaut et al. 1966).

Anatomically, the upper airway (superior to the larynx) is routinely divided into the upper nasopharynx, the middle oropharynx and the lower hypopharynx. Snoring results when anatomic or physiologic changes occur within these compartments during sleep and result in varying degrees of obstruction. Lugaresi et al. has suggested that during inhalation, the lower respiratory tract inferior to the larynx, lengthens. As the laryngo-tracheal-bronchial tract lengthens, it exerts a downward force on the oropharyngeal airway causing it to narrow (Lugaresi et al. 1989). Particular to the snoring population is a greater (more negative) inspiratory pressure (resulting in turbulent airflow), greater pulmonary resistance during snoring

ventilations, longer inspiratory time, and limited inspiratory flow. The above changes although frequent to snorers, are not pathologic in themselves and can occur in non-snorers too. The snorers differ from non-snorers in the severity of the above symptoms upon an airway that is more susceptible to obstruction (Hoffstein 1996).

Essentially lacking rigid support, the airway consists of the soft tissues of the pharynx, the soft palate, uvula and tonsillar pillars. Snoring results when either inspiratory or expiratory breathing, in combination with narrowing of the pharyngeal airway, results in vibration or "fluttering" of these soft tissues anywhere along the upper airway. Ellis et al. (1993) compared two surgical procedures for the correction of snoring that results from obstruction identified specifically at the palatal level of the airway. Ellis et al. believed palatal flutter was the most important cause of snoring. However, they also recognized that snoring could be generated from other areas in the upper airway. McGehee and Sly believed fatty deposition within the airway was a factor in air exchange problems associated with snoring and obstructive sleep apnea (McGehee and Sly 1996). In 1991, Croft and Pringle, using a nasoendoscope, directly visualized the site of pharyngeal obstruction in sleeping subjects. Their study revealed the sites of snoring origin and areas of obstruction and narrowing in the snoring subject's airway. The subjects were divided into three groups: Simple palatal snorers, a single level palatal obstruction group and a group with multisegment obstruction and airway compromise. The palatal snorers had a normal oro/hypopharyngeal area and palatal vibration occurred at the velopharyngeal level, where the airway was often narrowed. No obstructive episodes presented in the

palatal snoring group However, the single level palatal obstruction group demonstrated obstructive episodes associated with apnea at the velopharyngeal level. The oro/hypopharyngeal airway was without obstruction. The multisegment subjects exhibited similar circumferential closure at the velopharyngeal level as seen in the single level palatal obstructed group. In addition, the multisegment group displayed oro/hypopharyngeal involvement ranging from narrowing to complete.

Studies have demonstrated that it takes a highly negative inspiratory suction pressure to collapse the airway of a normal non-snoring individual, a mild to moderately negative pressure to collapse the airway of snorers and mild apneics, and in severe apneics the airway may collapse with even positive pressures. Pae et al. suggested that a more negative pressure was generated in an airway that was lengthened. Subjects with an incompetent airway extended their head posture which resulted in a taut and lengthened airway. A volume of air traversing a long pharynx generated a more negative pressure than the same volume passing through a shorter airway and resulted in a decreased airway diameter. The authors contended that resistance in such an extended and narrowed airway can result from turbulent airflow and not strictly anatomic blockade (Pae et al. 1997). To the contrary, Huggare and Laine-Alava observed neck extension to act favorably on nasorespiratory function (Huggare and Laine-Alava 1997). Miljeteig et al. investigated a small sample of eight adult non-apneic snorers and was not able to establish a significant relationship between snoring severity and nasal resistance to airflow during nasal breathing

exclusively. They pointed out that nasal obstruction could lead to oral snoring and, in such cases, relief of the obstruction might be beneficial (Miljeteig et al. 1993).

Negative pharyngeal air pressure, associated with inspiration, has also been shown to decrease lateral pharyngeal fat pad pressure in pigs (increased with expiration). This demonstrates a close relationship between respiration and pressure within the lateral fatty tissues of the pharynx (Winter et al. 1996). Another component to airway collapse is the compliance of the pharynx. A study by Brown et al. examined the muscle tone (or compliance) of the pharynx in a group of twenty loud snorers. Polysomnography studies were conducted and the subjects were divided into two groups. A group of thirteen were diagnosed with obstructive sleep apnea; the remaining seven did not present with obstructive sleep apnea. The cross-sectional area of the pharynxes of each group was determined by the acoustic reflection technique. The results of the study of apneic and non-apneic snorers revealed that obstructive sleep apnea victims were prone to have smaller pharyngeal cross-section resulting from an observed increase in pharyngeal compliance. The increased compliance was likely a result of the hypotonic state of the muscles. It was also determined that no correlation existed between pharyngeal compliance and obesity (Brown et al. 1985). A study using magnetic resonance imaging techniques, to evaluate the structure and function of the upper airway and surrounding soft tissues was performed by Schwab in 1996. Thicker lateral walls were demonstrated in apneics and an inverse relationship was observed during respiration between the caliber of the airway and the thickness of the lateral pharyngeal walls in both apneic

and normal subjects. The results of the study demonstrated the significance of the lateral pharyngeal walls in determining upper airway caliber. Although Burger et al. (1992) suggested that lateral pharyngeal walls thin with increasing lung volume, Schwab (1996) indicated that lung volume played an insignificant role in lateral pharyngeal wall change. Schwab revealed, with the use of magnetic resonance imaging, that the walls of the lateral pharynx were most thin at mid-expiration. He believed the lateral pharyngeal walls to be "key" to controlling changes in airway dimension. Imaging studies have also revealed the shape of the normal airway to be ovoid (wide laterally, but narrow anterior-posteriorly). The shape of the snoring subject's airway have been shown to decrease in width and increase in depth anterior-posteriorly. The airway of the apneic subject is constricted laterally to the point where its greatest dimension is anterior to posterior (Schwab et al. 1995). The thought that an increase in fat or water (edema) in the lateral pharyngeal walls could account for the increase in wall thickness was disproven with the use of magnetic resonance proton spectroscopy (Schwab et al. 1991) and computerized tomography (Haponic et al. 1983).

Investigators have suggested two hypotheses for airway collapse. The first suggests that pharyngeal collapse can result from neuromuscular control disturbances resulting in reduced upper airway dilator muscle activity. The other describes structural narrowing of the airway itself rendering it collapsible. In a recent study performed on felines at John Hopkins University, the purely structural basis for pharyngeal collapse was examined. The examiners blocked all neuromuscular

innervation and studied the pressure at which the airway collapsed while manipulating airway length and tongue position. Upper airway collapsibility was measured with the neck both flexed and extended, the trachea extended caudally, and with combinations of tracheal extension and both extended and flexed neck postures. It was found that, regardless of how the upper airway was displaced as its length increased, the pressure necessary to collapse and occlude the airway at the soft palate increased. Further examination of the taut airway revealed no changes in the cross-sectional lumen of the pharynx. A thinning of the rim of the soft palate was observed which suggested tension might account for the diminished collapsibility at that site. The affect of anterior tongue positioning on pharyngeal lumen dilation was significant only when elongation of the trachea was simultaneous. With the trachea in a neutral position, anterior displacement of the tongue had little affect on the flaccid airway's collapsibility because the pressure surrounding the collapsible site did not lessen significantly in a field of flaccid tissue. In contrast, as the tongue was displaced anteriorly in the taut airway, the pressure surrounding the airway decreased, diminishing collapsibility. Extrapolating from feline to man, the author attributed the increased collapsibility in apneics to possible imbalances between forces that dilate (bone and muscle) and compress (surrounding soft tissues) the airway; and recognized the potential of reduction in the longitudinal tension of the airway in collapse and obstruction (Schwartz et al. 1996). One can't help but think of the technique applied to open the airway in basic life support. Head-tilt and chin-lift maneuvers are routinely employed in lifesaving maneuvers. The airway is elongated

and the tongue displaced anteriorly. Another study, which removed the neuromuscular component of pharyngeal collapse, was conducted on three groups of human subjects. A group of seventeen normal, eighteen mild apneics and twenty-two moderate apneics, matched for age and body mass, were placed under general anesthesia while endoscopic measurements of the pharynx area in cross-section were performed at varying luminal pressures. The results indicated that the apneic subjects had narrower and more collapsible airways. This finding was also supportive of the anatomic hypothesis for pharyngeal collapse (Isono et al. 1996). Further support for the hypothesis that structural narrowing of the airway results in collapse was seen in the results of a cephalometric study conducted by Bacon et al. in 1990. They found the bony pharyngeal opening, defined as the distance from basion (Ba) to the posterior nasal spine (PNS) was reduced in forty-three adult male apneic subjects when compared to forty normal adult males (Bacon et al. 1990). In 1986, Bradley et al. determined that the pharyngeal airway of non-apneic snorers were less likely than apneic snorers to collapse at diminished lung volumes. They also determined that both snoring groups presented narrower pharyngeal areas than non-snorers. The snorers without apnea appear protected from complete collapse and obstruction of their airways. There was less tendency for airways of non-apneics to narrow to a critical point between obstruction and patency (Bradley et al. 1986).

Irritant receptors, located within the larynx, are responsible for defensive mechanisms such as laryngeal closure and the cough. A study examining the response of these receptors to three different aerosols was performed. The receptor

response to saline, dextrose and distilled water were tested in dogs. The results suggest that the upper airway dilating muscles, specifically the genioglossus and posterior cricoarytenoid muscles, may be stimulated by the laryngeal receptors, in turn, ensuring airway patency (Kuna et al. 1996)

In 1985, Koskenvuo et al. stated that pharyngeal dilator muscle tone decreased in heavy snorers in response to the affect of blood gas composition on medullary nerve centers. Recent work by Series et al. challenged the neuromuscular control hypothesis for pharyngeal collapse which suggests that decreased neural output to the airway dilator muscles results in reduced muscle activity. The dilator muscles of the pharynx function to open the upper airway. The muscle that retracts the uvula and contributes highly to nasopharyngeal closure is the musculus uvulae. Series et al. examined the contractile, histochemical, and biochemical features of this muscle in apneic and non-apneic snorers. The muscle samples were excised when the subjects underwent surgery for the treatment of snoring. An analysis revealed that, in the apneic subjects, the muscle presented all the signs of hyperstimulation and showed significant differences between the apneic and non-apneic groups. The muscle in the apneics was greater in cross-sectional mass, produced greater twitch and tetanic tension, and higher protein content and anaerobic enzyme activity. The authors concluded that the muscle adapted as a consequence of over stimulation much like any well exercised muscle. The belief that the dilator muscles fatigue, or become incompetent as a result of sleep-related breathing disorders, was not

supported by the study. Electrical stimulation of such, already hyperstimulated, muscle could result in adverse fatigue and function (Series et al. 1995).

Evaluation of Snoring

Numerous studies have been conducted in an effort to objectively measure snoring sounds. Despite all attempts, at present, there are no agreed upon standards with which data can be compared.

Much of the research of the late 1980s used suspended microphones from the subject's face at a variety of distances. The sounds were recorded and analyzed for density or average amplitude (Leiberman et al. 1986; Wilson et al. 1985).

In the early 1990s, Dalmasso et al. used two microphones, one placed over the sternum and the other held at the mouth (Dalmasso et al. 1990). Shortly after, investigators placed sound transducers on the sternum notch to measure sound pressures (Issa et al. 1993). The polygraph was more recently used to record the intensity of sound recorded from a microphone placed over the frontal sinus (Hoffstein et al. 1994) and suspended over the subject (White et al. 1994). Over all, investigators have measured snoring sounds with different recording devices, from varying locations and distances, and have interpreted the data with little agreement on an objective definition of snoring.

The evaluation of snoring from a subjective perspective is most frequently applied in the form of questionnaires and surveys. However, as with objective measurement of snoring, subjectively measuring snoring also presents problems. One must rely upon the report of the observer since few snorers are aware of their snoring

(Hoffstein et al. 1994). Self-reported snoring frequency or volume has been demonstrated to be inaccurate and unreliable (Wiggins et al. 1990; Bliwise et al. 1991).

Limited in the degree of accuracy obtainable in the objective and subjective evaluation of snoring, one is confined to research based on less than perfect measures (Young 1991).

A subjective study by Carroll et al. in 1995 questioned the parents of over eighty children (ages 5.4 months to 14.8 years) with snoring and sleep related breathing problems referred for evaluation of possible obstructive sleep apnea. The objective of the study was to determine if, based solely on the child's sleep history, one could reliably distinguish primary snoring from obstructive sleep apnea. After the parents completed a sleep behavior questionnaire, each child was studied with overnight polysomnography. Sleep efficiency and architecture, chest and abdominal motion, cardiac rate and rhythm, arterial oxygen saturation and airflow were recorded. Each polysomnographic study was audio-taped and video-recorded. Based on the sleep study results, the subjects were classified as either primary snorers (snoring without obstructive sleep apnea syndrome), or as children with obstructive sleep apnea. After classification, the nominal data from the questionnaire was statistically analyzed. The results showed sleep efficiency and architecture not to differ significantly between the primary snoring and the apneic groups. The percentage of time spent in rapid eye movement sleep declined with increasing age as expected; and so did the number of rapid eye movement periods per night and the

number of movements per hour (Prinz and Raskind 1978). When basing the diagnosis of obstructive sleep apnea syndrome on the questionnaire alone, without polsomnography study, 29% of subjects with primary snoring only, would have been incorrectly labeled as apneic. Of the subjects identified as apneic with polysomnography, 21% were incorrectly classified as not being apneic based only upon the questionnaire responses. Therefore, only 42% of subjects would have been correctly grouped as primary snorers or obstructive sleep apneics based solely upon the questionnaire. Although the parents witnessed apneic events during sleep, their child struggling to breath, shook their children to make them breath, and watched them sleep in fear, with greater significance in the obstructive sleep apnea group, parent responses were also high in the primary snorer group. Significant differences did not exist between the groups regarding snoring character, obesity, daytime symptoms or failure to thrive; and cyanosis was infrequently reported in both groups. Mouth breathing was significant for the apneic group. In conclusion, there appeared to be a great deal of overlapping symptoms between the primary snorer group and the obstructive sleep apnea group. The results of this study indicated that the diagnosis of obstructive sleep apnea syndrome, based primarily upon clinical history in children, was inadequate, and definitive diagnosis should include objective measures (Carroll et al. 1995).

When comparing objective measures of obstructive sleep apnea in children and adults, many differences are observed. One cannot extrapolate adult polysomnographic criteria to children and adolescents (Marcus et al. 1992). Carroll

et al. have shown that sleep is less disrupted by obstructive sleep apnea, and excessive daytime sleepiness is much less frequent in children than adults. The apneic child should not be strictly envisioned as obese, having morning headaches and excessive daytime sleepiness. Children that breath through their mouths, snore loudly most nights, have witnessed apnea and difficulty breathing might not be apneic after all. Objective study, measuring breathing during sleep, can reveal that many once thought of obstructive sleep apnea victims, suffer only from primary snoring (Carroll et al. 1995).

Marcus et al. (1992), in a study of fifty children from age one to eighteen, screened for minimal likelihood of obstructive disease and considered "normal", were studied with polysomnography. In agreement with Carroll et al., the sleep study results of this age group differed from those in adults. Obstructive apneas, where chest motion was present, but airflow was absent, were uncommon in children, and never over ten seconds duration. Obstructive apnea is measured by counting the number of apneic episodes lasting ten seconds or more, that occur per hour of sleep (apnea index). In general, although it varies with the authority conducting the study, the normal for adults is less than five to ten. The children in this study had an apnea index of only 0.1 ± 0.5. The author thought that more than one obstructive apneic episode per hour of any duration should be considered abnormal in children. Central apneas, where chest wall motion and airflow were both absent, occurred in fifteen of the fifty children studied. Only one child demonstrated a decline in oxygen saturation below 90%. The authors believed central apneas should be considered abnormal if

the oxygen saturation drops below 90%, regardless of the length of the episode. No mixed apneas, where both obstructive and central components occur together, were witnessed in children or adolescents. Pauses in breathing greater than three seconds in duration, and more than twenty seconds apart, occurred in 8% of the children examined. In those children, arterial oxygen saturation was maintained between 92-97%. The study supported previous findings suggesting that sleeping arterial oxygen saturation levels for adolescents should not fall below 92%. The authors cautioned against applying adult criteria for diagnosing obstructive sleep apnea syndrome to children and adolescents for fear of underdiagnosis (Marcus et al. 1992).

In a study conducted to examine differences in oxygen saturation levels between snoring and non-snoring children up to ten years of age, no significant differences were found. The study used pulse oximetry recording only, no other measurements were employed. Based upon the results of the study, the use of overnight pulse oximetry alone, to exclude a diagnosis of obstructive sleep apnea, would not be an effective screening method (Owen et al. 1995). Pradhan et al. attributed the inability of pulse oximetry alone to screen reliably for obstructive sleep apnea syndrome because significant arterial oxygen desaturation might not occur during apneic and/or hypopneic events. In developing a screening algorithm to detect obstructive sleep apnea in snoring populations, the authors determined that when the lowest oxygen saturation recording from overnight pulse oximetry examination was combined with significant clinical predictors of apnea (male sex, body mass index,

age and increased complaints of snoring), the number of polysomnogram studies could be decreased by 13%, and remain extremely sensitive (Pradhan et al. 1996).

Snoring and Disease

In adults, sleep disordered breathing has been associated with numerous pathologic outcomes. Dependent upon severity, physiologic changes can result in failure to thrive, cor pulmonale, excessive daytime sleepiness, learning disorders, developmental delay and hyperactivity (Loughlin 1992).

In 1985, Koskenvuo et al. identified significant associations between snoring and hypertension in men and women, and habitual snoring and angina pectoris in men. The association between snoring and myocardial infarction or hospital admission for ischemic heart disease was non-significant.

In a retrospective investigation that examined the prevalence of cardiac arrhythmias in 458 patients who had undergone polsomnography, it was determined that subjects with sleep apnea and nocturnal hypoxemia had a significantly higher prevalence of arrhythmia than snorers and non-apneic subjects. In addition, the subjects with the most severe apnea also demonstrated the most serious arrhythmias. The study results support the evidence that higher mortality from cardiovascular causes is seen in apneic subjects (Hoffstein and Mateika 1994).

A year later, Olson et al. studied 441 randomly selected subjects for associations between sleep apnea and cardiovascular disease including hypertension. It was concluded that when confounders, such as age, sex, body mass index, current alcohol consumption and smoking were considered, no significant association

between sleep disordered breathing and cardiovascular disease, including hypertension, existed (Olson et al. 1995). Young et al. studied 580 adults the following year, also adjusted for the confounders of age, sex and body mass, but concluded that simple snorers (snoring without significant apnea and hypopnea) were four times more likely to suffer from hypertension and cardiovascular disease than those without sleep disorders (Young et al. 1996).

Hoffstein et al. verified associations in adults between sleep arousal and snoring (Hoffstein et al. 1995). Ulfberg et al. observed an association between headache upon awakening and heavy snorering and apnea (Ulfberg et al. 1996). Although positive correlation has been observed between adult snoring and body mass, age, male gender, and in some reports, smoking (Janson et al. 1995), no significant link has been made between habitual snoring and brain infarction (Neau et al. 1995). Guilleminault and Dement have delineated the clinical symptoms of adult snorers that include numerous psychological components and abnormal motor activity; sleepwalking, sleep "attacks", amnesia and loss of judgment, hallucination, depression, confusion, irritability, impotence and bed wetting (Guilleminault and Dement 1978).

Guilleminault and others have observed that snoring in childhood and adolescence can ultimately result in adulthood apnea and associated pathologies (Guilleminault and Dement 1978; Westbrook 1983). Marcus et al. suggested that more labored breathing in children with obstructive sleep apnea syndrome resulted in

increased calorie burning. Therefore, as a result, normal growth was observed to have been stunted in apneic children (Marcus et al. 1994).

In 1981, a prospective study was performed on ninety-two children between the ages of one and thirteen who underwent tonsillectomy, adenoidectomy, or both. Three children presented with a history of snoring, apnea and excessive daytime sleepiness and two of these had otitis media. Electrocardiographic evidence revealed right heart strain in all three of these children. It was suggested that the snoring, apnea and pulmonary hypertension were the result of upper airway obstruction. In all three cases, symptoms were relieved and the hemodynamics returned to normal after the tonsils and adenoids were removed. A weakness of the study was the lack of polysomnographic measures to support their labeling of children as apneic. Only one child had such sleep study carried out and in that particular case the results were indicative of an obstructive type event. The results suggested that over 3% of children who presented with inflamed tonsils and adenoids had electrocardiographic evidence of early pulmonary hypertension, and such conditions were reversible with the elimination of obstruction. The serious nature of such innocuous signs and symptoms in childhood and adolescence must not be overlooked (Wilkinson et al. 1981).

In a case series report of twenty-five full-term infants having had "near-miss sudden infant death syndrome (SIDS)" followed for their first year of life, Guilleminault and Stoohs (1992) suggested a frequent familial history of obstructive

sleep apnea. Also coincident to these children were small posterior airway spaces and an increased upper airway resistance.

Regarding the prevalence of allergic sensitization in snoring children, McColley et al. studied thirty-nine children between the ages of one and seven, referred for sleep studies. The study demonstrated an incidence of allergic sensitization three times that reported in children in general. The results of the study also determined that the presence of allergy was associated with increased risk of obstructive sleep apnea syndrome (McColley et al. 1997).

Loughlin (1992) has listed the clinical manifestations frequently considered as possible consequences of obstructive sleep apnea syndrome in children as the following.

I. Asleep.

   A Hypoventilation with hypoxemia and hypercapnia.

   B. Endocrine dysfunction.

   C. Enuresis.

   D. Anoxic seizures.

   E. Cardiac arrhythmias.

II. Awake.

   A. Excessive daytime sleepiness. (EDS)

   B. Mood and personality changes.

   C. Developmental delay.

   D. Poor or deteriorating school or work performance.

E. Failure to thrive.

F. Morning headaches.

G. Digital clubbing

H. Cardiovascular dysfunction.

1. Systemic hypertension

2. Pulmonary hypertension

3. Polycythemia

Cephalometric Assessment

Investigators of snoring have hypothesized that facial growth and appearance can be a reflection of the processes of development. Facial form, therefore, would be a result of an individual's growth circumstances and characteristics (Enlow et al. 1971). The affect of chronic airway obstruction on the dentition and dental arch form has been documented (Gross et al. 1994; Linder-Aronson 1974; Subtelney 1954). Guilleminault et al. has suggested that enlarged tonsils, adenoids and lymphoid tissues in childhood could induce unfavorable craniofacial bony changes with maturity (Guilleminault et al. 1989). Potsic and Wetmore suggested that the development of the long face was associated with chronic nasal obstruction (Potsic and Wetmore 1990).

The most useful and easily obtainable radiograph in the study of subjects prone to snoring and apnea is the lateral head film (cephalometric radiograph). Both hard and soft tissues of the entire airway superior to, and including the larynx, can be observed in the sagittal dimension. Much research has been performed using

cephalometric radiographs to measure a variety of hard and soft tissue landmarks identified on the films. In studies, primarily of adult subjects, correlation between soft and hard tissue landmarks and breathing disorders have been sought. Few studies have examined cephalometric variation within a child and adolescent cohort.

Most studies are a variation of a recurring theme. Generally, polysomnographic studies are performed to discern apneic subjects from non-apneic snorers and non-snorers. Body mass index (weight(kg)/height(m)^2) is frequently used as a measure of obesity (Khosla and Lowe 1967), and many different hard and soft tissue linear and angular comparisons are made. The results of several studies are presented.

Partinen et al. stated that cephalometrics can be useful in determining the appropriate treatment for obstructive sleep apnea patients. Risk factors for obstructive sleep apnea in adults included a distance from the most anterior-superior point on the hyoid bone to the mandibular plane of greater than or equal to 24 mm, a posterior airway space less than or equal to 5 mm and obesity. Posterior airway space is measured along the B point-gonion plane. As body mass index increased, especially in the most obese subjects, the cephalometric measures became less significant as predictors of apnea (Partinen et al. 1988).

In 1990, Bacon et al. used cephalometric analysis to determine morphologic differences between adult sleep apnea syndrome subjects and normal controls. The observations indicated the following variation in the apneic group: A shorter cranial base, posterior facial compression with a reduced bony pharynx, a vertical

lengthening of the lower face with retruded chin and tongue and a lengthened soft palate.

Maltais et al. compared cephalometric measures between adult snorers, non-snorers and apneics. The results indicated greater hyoid to mandibular plane distance and increased soft palate length in the apneic group In the snoring group, hyoid to mandibular plane distance was similar to controls. However, distance increased with increasing age and the soft palate was longer in snorers compared to non-snorers. Body mass index was not correlated with any of the cephalometric measures (Maltais et al. 1991).

Zucconi et al. studied the importance of cephalometric variables in adult snorers with and without apnea. Snorers were divided into two groups based upon the average number of apnea or hypopnea events presented per sleep hour. One group (group A) presented less than ten events per hour, the more severely affected group (group B) presented more than ten. A subset of the severely affected group averaged over thirty events per hour. Their results were in agreement with Maltais et al., in that both hyoid to mandibular plane distance and soft palate length were increased. In the more severe group, body mass was greater and they had a longer snoring history. Otolaryngology examination revealed greater oropharyngeal obstruction and macroglossia in the severe group. Since little association could be identified between mildly apneic snorers and cephalometric variables, the authors suggested the differences seen in the severe group might have been a consequence of the snoring rather than a cause (Zucconi et al. 1992).

Zucconi et al. again studied snorers with and without obstructive sleep apnea syndrome. An increase in hyoid to mandibular plane distance and soft palate length was again confirmed. A decrease in the sagittal cranial base, maxilla and mandible were observed by reduction in Ba-N, Ba-PNS, PNS-A, S-N, Go-Me and Go-Gn compared to controls. The posterior airway space was reduced in apneics and the posterior facial height was not decreased. The lower facial height did not increase in this study, however, Bacon et al. demonstrated the opposite in 1990 (Zucconi et al. 1993).

Will et al. compared cephalometric measurements of Caucasian, black and Hispanic groups diagnosed with obstructive sleep apnea syndrome. The following landmarks were compared between the different ethnic groups: SNA, SNB, ANB, hyoid to mandibular plane, soft palate length and posterior airway space. The data from apneic subjects in each group revealed minor variation from the ethnic norm in Caucasians, significant bimaxillary prognathism from the norm in blacks and bimaxillary retroposition in the Hispanic subjects (Will et al. 1995).

Guilleminault et al. investigated the familial tendency for obstructive sleep apnea and concluded that certain craniofacial features, which appeared to have a genetic basis, were risk factors for the development of obstructive sleep apnea (Guilleminault et al. 1995). Others have supported these findings (Strohl et al. 1978; Redline et al. 1992).

Recently, Nelson and Hans examined the contribution of craniofacial risk factors in increasing apnea in obese and non-obese snorers. In the non-obese group,

greater tongue length was determined the most reliable predictor of apnea. A shorter anterior-posterior alignment of the middle cranial fossa (more posterior position of the maxilla) correlated to more frequent apnea, and increasing age was also related to increased apneic activity. Within the obese group, increased hyoid to mandibular plane length and tongue length correlated higher with obstructive sleep apnea syndrome (Nelson and Hans 1997).

In 1983, Adamidis and Spyropoulos evaluated over one hundred children between the ages of seven and twelve with class I malocclusion and consistent mouth breathing secondary to enlarged adenoid tissue, confirmed radiographically. Controls were of similar ages and presented ideal occlusion, facial harmony and no nasal obstruction. The objective of the study was to determine the effects of hypertrophic adenoid tissue on the position of the tongue, the mandible and the hyoid bone. The results indicated that mouth breathers had a significantly more down and forward tongue position. Also of significance in mouth breathers with hypertrophic adenoids was a downward inclination of the mandible as evidenced by increases in ANS-Me, PNS to the mandibular plane and angular measurements. The angle between the long axis of the hyoid bone and the palatal and basion nasion planes was significantly increased, as was the distance from the palatal plane to the hyoid. The authors suggested the resultant tongue position could account for the frequent high palatal vault, narrow maxilla and open bite tendency observed in "long face syndrome" subjects.

A year earlier Brouillette et al. studied twenty-two infants and children with sleep apnea. The authors attributed twelve cases of right ventricular hypertrophy (cor pulmonale), six subjects with weight below the tenth percentile, and neurologic and behavioral disturbances to delay in referral of the subjects for care. Radiographic examination demonstrated narrowed upper airways resulting from tonsil and adenoid hypertrophy, and anatomic abnormalities including mandibular posterior displacement, temporomandibular joint ankylosis, syndrome children and cleft repair subjects. Surgical treatment of the subjects ranged from surgical excision of tonsils and/or adenoids to tracheostomy alone. Surgery was aimed at relieving airway obstruction and all patients experienced improved activity and development. Obstructive sleep apnea was relieved effectively in two thirds of the subjects (Brouillette et al. 1982). The cephalometric radiographs in the Brouillette et al. study were obtained with the subject in a supine position. In a study investigating the affect of positional change from upright to supine position on cephalometric variables, Pracharktam et al. found that significant differences existed between snoring and obstructive sleep apnea subjects. However, the distance from the posterior pharyngeal wall to the soft palate decreased, and the hyoid bone elevated in the supine position. The authors recommended upright cephalometric radiography (Pracharktam et al. 1992).

In 1992, Shintani et al. compared the relationships between cephalometric values in children and obstructive sleep apnea syndrome severity. One hundred and eleven children with obstructive sleep apnea syndrome and a mean age of 4.5 ± 1.7

years were evaluated. It was determined that children with the most severe apnea had larger adenoids and a narrower airway radiographically than the children with mild apnea. Relating cephalometric values and treatment, it was determined that in children with obstructive sleep apnea, the best responders to adenotosillectomy had larger adenoids and tonsils, and lower hyoid bone position. The authors concluded that hypertrophy of the adenotonsillar tissue can easily affect the child's airway. Hypertrophy then, is the most common cause of obstructive sleep apnea in children and can successfully be treated with adenotonsillectomy.

Treatment

It is generally agreed that snoring is a symptom of a compromised airway and can lead to serious health problems. It has been shown that mechanical blockage or growth disorders that result in upper airway resistance lead to an increased risk of obstructive sleep apnea. Recognition in childhood is, therefore, of the utmost importance. Guidance during growth, by the removal of anatomic obstacles, can restore pathologic function to normal. Treatment in children means not just improving facial growth, but avoiding a future of potentially impaired health (Viva et al. 1992).

In adults, both surgical and non-surgical approaches to treatment have been suggested. Non-surgical corrective efforts have included position changes in bed, weight loss, nasal decongestants, avoidance of alcohol, and sedative or hypnotic drugs that relax the pharyngeal musculature. More recently, nasal dilator strips,

tongue retaining devices and removable mandibular advancement devices have been of interest. The use of continuous positive airway pressure has also been effective.

Surgical treatment in adults has focused on alteration of the underlying anatomical variant. Deviated septum repair and resection of redundant pharyngeal tissue and the soft palate can help. Orthognathic surgery might be warranted in severe cases, but commonly used procedures to reduce snoring include uvulopalatopharyngoplasty (UPPP) and laser assisted uvulopalatoplasty (LAUP).

In children, the most common causes of airway obstruction are enlarged tonsils and adenoids. Postoperative improvement is anticipated soon after surgery when the tonsils are the obstructive source. (Potsic and Wetmore 1990). Most children with adenotonsillar obstruction, craniofacial abnormalities and neuromuscular disorders react favorably to adenotonsillectomy (Potsic 1989). Brooks (1993) stated that any factor that decreased pharyngeal size or increased pharyngeal compliance, increased a child's risk for obstructive sleep apnea.

In 1997, Nieminen et al. evaluated seventy-eight Finnish children referred for ENT evaluation because of parental complaints regarding their child's snoring and obstructive symptoms. The children were between the ages of 2.4-10.5, the average age was 5.6. The purpose of the study was to determine which factors in the patient's history and clinical examination were predictive of obstructive sleep apnea syndrome in snoring children. The parents first observed their child's sleep for two weeks before completing a questionnaire designed to help identify obstructive sleep symptoms. During the clinical examination, each child's palatine tonsils were graded

for size. Subjects were excluded from the study if found to have developmental anomalies Children with a history of adenoidectomy were not excluded from the study sample. Next, each child underwent overnight polysomnography. Lastly, the questionnaire answers and the clinical findings were compared to the polysomnographic studies to determine possible risk factors predictive of obstructive sleep apnea syndrome. Analysis of the results indicated that in subjects whose parents witnessed their apnea every night, the risk of obstructive sleep apnea was greatest. Children with a history of constant snoring, restless sleep, previous adenoidectomy and enlarged tonsils were also at a significantly greater risk for obstructive sleep apnea. The authors concluded, based upon the results of their study, that adenoidectomy alone was not sufficient treatment for obstructive sleep apnea syndrome in children; and adenoid tissue does not play a critical role in development of obstructive sleep apnea syndrome. Had the authors conducted polysomnography on the study population before and after adenoidectomy, they might have had a stronger argument supportive of such a claim. Relative to adenoidectomy, the only thing presented in the study was that a greater percentage of apneic patients had histories of adenoidectomy, than did non-apneic subjects. Nothing was done in the methodology to demonstrate either an adverse, or a beneficial affect of adenoidectomy relative to apnea expression (Niemimen et al. 1997).

In 1982, Guilleminault witnessed a reversal of symptoms in one hundred percent of children (N = 25), between the ages of 2 and 14, with sleep disorders and enlarged pharyngeal lymphoid tissues treated with tonsillectomy and/or

adenoidectomy. All subjects presented with heavy nocturnal snoring and many had other clinical symptoms, often behavior related. None of the subjects had sleep apnea or oxygen desaturation evidenced in sleep studies, however, each child presented significant respiratory resistive load during sleep. Ten children presented cardiac arrhythmias during sleep. The conclusions presented emphasize that a narrowed airway during sleep in children can be deleterious to the cardiovascular system, as well as adversely affecting daytime functioning; and that tonsillectomy and/or adenoidectomy can result in the complete disappearance or substantial amelioration of symptoms (Guilleminault et al. 1982).

In 1995, Suen et al. evaluated sixty-nine children between the ages of one and fourteen who were referred to the otolaryngologist for suspected obstructive sleep apnea. All subjects snored, had enlarged tonsils, witnessed apneas and/or daytime somnolence. Polysomnography studies were conducted on all the children. Despite the symptoms, over half of the children had normal results. For thirty of the subjects with significantly disturbed sleep patterns, adenotonsillectomy was performed. At least six weeks after surgery, twenty-six of the thirty children returned for post-operative polysomnography. Significant increases in total sleep time, time spent in stage-1 sleep, and decreases in the number of arousals after adenotonsillectomy were observed. The number of respiratory disturbances decreased and a significant improvement in oxygen saturation occurred. Four patients continued to have a high number of respiratory disturbances and continued to snore. Only three of the twenty-two remaining subjects with infrequent respiratory disturbances (post-surgically),

persisted in snoring. The authors believed that assessment of the severity of obstructive sleep apnea based on clinical examination and history alone, was inadequate. They strongly recommended polysomnographic study for every child with symptoms suggestive of obstructive sleep apnea, since baseline tests were strongly predictive of post-operative outcome. Follow-up polysomnography was encouraged for subjects with more disturbed pre-surgical sleep patterns and when snoring persists despite surgery (Suen et al. 1995).

In 1996, Ishizuka and Kakuta compared the post-adenotonsillectomy improvement in over one hundred severe and non-severely apneic children (age 3-9, avg. 5.6). The degree of apnea severity was determined by the frequency of apneic episodes over a seven hour monitored sleep period. Severe cases presented more than thirty apneic episodes during the seven hour period; the non-severe had less than thirty. Post-operative testing revealed significant reduction in apneic episodes in over 71% of severe apnea cases. The excised adenoid and tonsil tissues were weighed post-operatively in every case. There was a significantly greater mean adenoid mass in the severe group than in the non-severe group. No significant differences were found between the groups regarding tonsil mass. Parent's responses to questionnaires six months post-operatively showed an improvement rate of 100% for snoring, respiratory arrest and sleep troubles. The investigators strongly recommended polysomnography to evaluate upper airway obstruction and suggested significant reduction in the severity of apnea in patients treated with adenotonsillectomy (Ishizuka and Kakuta 1996). A similar resolution of symptoms was observed by

Stradling et al. in 1990. Adenotonsillectomy was held accountable for elimination of severe abnormalities in children's waking and sleep behaviors, including snoring (Stradling et al. 1990). Miyazaki et al. stated, "It is vital not to overlook respiratory disorders (in children), but to diagnose and treat them appropriately" (Miyazaki et al 1989).

In cases of extreme micrognathia the tongue has been sutured to the lip in an attempt to relieve obstruction. Referred to as glossopexy, it is most commonly used in neonates with severe deformity (Potsic and Wetmore 1990)

The UPPP procedure has been successful, especially in combination with adenotonsillectomy in children with neuromuscular disorders (Potsic 1989). The procedure has also been very effective in children with large uvula and redundant pharyngeal mucosa (Potsic and Wetmore 1990).

Management of obstruction in severely affected children may be addressed with temporary airways. A nasopharyngeal airway or tongue retaining device can improve the quality of life over many years if the child is compliant. A nasopharyngeal airway or tongue retaining device can be worn at night.

A tracheostomy, aside from its indication in acute emergency, is often a last resort and, once placed, is demanding of care and maintenance. Where all efforts have failed to result in improved airway compliance in children, the benefit to tracheostomy can be critical to survival. Often placed temporarily, while the child grows, some are needed for years. In older children the quality of life can be improved by blocking the stoma during the day and opening it at night. Nasal

continuous positive airway pressure presents an alternative to tracheostomy in adolescents. However, compliance and tolerance over the long-term generally decline (Potsic and Wetmore 1990).

Less commonly used procedures for the relief of obstructive sleep apnea in children include hyoid suspension and craniofacial surgery. Children suffering from a narrow pharynx resulting from skeletal deformity can benefit from craniofacial surgery. Surgical advancement of the maxilla, mandible, or both, may open the obstructed airway. Advancement of the genioglossus muscle and base of the tongue from the posterior pharynx can open an occluded airway. The advancement is accomplished by ligating the hyoid bone to the mandible in an anterior-superior direction. This surgical procedure is rare in the young, but might be of benefit to older children and adolescents where all else has failed (Potsic and Wetmore 1990).

Respiratory stimulates appear promising in the treatment of children with mixed apneas. Methyphenidate has been shown to decrease excessive daytime sleepiness (Potsic and Wetmore 1990).

In conclusion, limited epidemiological data suggests that 11% of children snore habitually. Those children commonly experience restless sleep, hyperactivity and daytime sleepiness.

A physiologic basis for snoring has been explored. Snoring can result from vibration of the soft tissues anywhere along the upper airway. Snoring can be associated with airway obstruction. However, the shapes of the non-snorer, the snorer and the snorer-apneic (obstructed) airways demonstrate differences. An

anatomic justification for airway collapse and obstruction appears warranted. Non-apneic snorers, although presenting altered airway anatomy, avoid total collapse and obstruction.

Regarding objective measurement of snoring sounds, there has been little agreement on standards with which to compare data. Subjective evaluation of snoring, often in questionnaire form, have present problems and are of limited accuracy. It would appear, unlike adults, little difference exists between snoring children and apneic children regarding snoring character, obesity, daytime symptoms or failure to thrive.

In snoring and apneic adults, much investigation has been directed at determining relationships between sleep disordered breathing and numerous pathologic conditions. However, systematically collected data is lacking in children and adolescents. Little is known about the specific risks associated with snoring and airway obstruction in this younger cohort. Innocuous signs and symptoms of a serious nature may be routinely overlooked. To the contrary, procedures used to diagnose and treat adults are being applied to children with a lack of awareness for the efficacy or appropriateness of the treatments rendered.

Criteria for identifying children at risk for the development of significant obstructive airway disease need to be established. It has been observed that a large percentage of adults with obstructive apnea have histories of snoring as children. It has also been agreed that snoring is a symptom of a compromised airway that can

lead to health problems of a most serious nature. Treatment in children might avoid future impairment of health.

STATEMENT OF THE THESIS

The purpose of the cross-sectional study was to test the hypothesis that there are differences in craniofacial features between snoring and non-snoring children.

MATERIALS AND METHODS

Study Population

The study population enrolled healthy Caucasian and African-American, male and female children, between the ages of seven and fourteen. The snoring and non-snoring subjects were sought from a population seeking orthodontic examination but, had not yet been treated by means of either orthodontics, or orthopedics. Tonsils and adenoids were present in all subjects except for four experimental group children who had adenoids and /or tonsils removed prior to seeking orthodontic care. The snoring subjects snored often, and with a volume ranging from moderate to very loud. The control group was composed of children that never snored. Control subjects were matched to the experimental group according to age, gender and ethnicity.

In order to identify a pool from which snoring and non-snoring children could be located, subjects were recruited from patients seeking orthodontic treatment at Case Western Reserve University, School of Dentistry, Department of Orthodontics, and from twelve different orthodontic private practices in Cleveland, Ohio and the surrounding area (Appendix E). See Table 1. The number of snoring and non-snoring subjects obtained from each practice, is provided in Table 2.

TABLE 1 --Survey Distribution Data

Data Source	Surveys Distributed	Surveys Completed	Dates of Data Collection
CWRU	66	66	Jul 97-Apr 98
Dr Alvetro	2	2	Mar 98
Dr. Amberman	10	8	Jul97-Nov97
Dr. DeLoach	29	20	Jul97-Jan98
Dr. Dworkin	50	41	Jul97-Sep97
Dr Kossoff	17	14	Jul97-Dec97
Dr. Simpson	24	12	Jul97-Oct97
Dr. Souris	14	14	Sep97-Apr98
Dr Wenger	38	23	Jul97-Sep97
Dr White	34	28	Jul97-Oct97

TABLE 2 --Sources of Study Population

Data Source	Snoring Children	Non-snoring Children
CWRU	10	8
Dr Alvetro	2	0
Dr Amberman	1	1
Dr DeLoach	3	3
Dr Dworkin	4	10
Dr Kossoff	1	0
Dr. Simpson	2	2
Dr. Souris	1	0
Dr Wenger	4	1
Dr White	3	6

Recruitment Strategy

Within the Department of Orthodontics at Case Western Reserve University, parents of subjects aged seven to fourteen, presenting for pre-treatment initial records, were asked to participate in the study. Those agreeing to participate were asked to complete a consent form (Appendix A), a parent and child demographics form (Appendix B), and a sleep behavior questionnaire (Appendix C). The records of each patient were reviewed to ensure the subject was healthy, had no craniofacial abnormality and no prior orthodontic/orthopedic treatment. The pre-treatment radiograph was examined for diagnostic quality and to ensure all critical landmarks were included within the boundaries of the film (e.g., hyoid bone). All cephalometric radiographs taken within the Department of Orthodontics on the Bolton cephalometer have metallic registration landmarks spaced 50 mm apart within the nasion rest. In addition, residents routinely record the distance between the patient midline and the cephalometric film at the time of exposure (ML distance). Both recordings were useful in determining image magnification. The date the radiographs were taken were recorded, and the ages of the children at the time the radiographs were taken were calculated to the nearest month. Height and weight were recorded on each subject in inches and pounds, respectively, and each parent/guardian was asked at what age their child's snoring began.

Outside the School of Dentistry, Cleveland area practitioners provided a data base of seven to fourteen year old patients presenting to their respective practices between July 1997 to April 1998 (Appendix E).

The following methods and procedures were used at the first three private offices visited. The investigator reviewed records of "new patient starts" including records of patients presenting for examination up to four months prior to the investigator's visit. The records were evaluated to ensure the individual subjects were healthy, met the age range criteria, and had no craniofacial abnormalities. The pre-treatment cephalometric radiographs were examined for diagnostic quality, and to ensure all critical landmarks were contained within the boundaries of the film. The date the radiograph was taken was recorded, and the age of the child at that time was calculated. The nasion rest on the cephalometer used to expose the cephalometric radiographs was measured to assist in determining the magnification of the radiographic images. Next, the cephalometric radiographs were traced by the investigator immediately following review of the records.

After record review of the medical history and radiographs, the subject's sex and race were documented. Additionally, the addresses and phone numbers of the parents or guardians were recorded.

Within days of the data collection, a letter explaining the investigators background, the intent of the study, and confirmation of the orthodontist's agreement to participate in the study (Appendix D), was mailed to each subject's parent or guardian. Each letter was accompanied by a sleep behavior questionnaire (Appendix C), which the parent or guardian was asked to complete.

A week to ten days after the letters and questionnaires were mailed to the parents or guardians of the children, the examiner or his assistant telephoned the

parent or guardian to obtain their answers to the sleep behavior questions. Once contacted, the answers were recorded by the investigator, or his assistant, on duplicate questionnaires. The parents or guardians were also asked the child's approximate height and weight, and at what age their child began to snore.

At the remaining eight local Cleveland area offices visited, the cephalometric radiographs were not traced simultaneously with the record review. Instead, snoring and non-snoring children were identified by questionnaire first, then their cephalometric radiographs were collected from their respective offices, mixed randomly together, traced and measured.

At one office, questionnaires were mailed to the parents/guardians, and a self-addressed stamped envelope was provided. Seven to ten days after mailing of the questionnaires, phone calls were made to the homes that had not returned their surveys. At another local office, the orthodontist was made aware of the criteria of the study population and asked parents of his patients to complete the questionnaires.

One practitioner, outside the Cleveland area, volunteered to provide questionnaires for new records patients in her office She was thoroughly presented with the study population criteria and reviewed the patient's history to ensure age criteria were met, no craniofacial abnormalities were present, and no prior orthodontic/orthopedic treatment existed. She provided completed questionnaires and cephalometric radiographs on snoring children for inclusion in the study. Only questionnaires of snoring children were provided.

A total of thirty-one snoring subjects were identified. Non-snoring subjects were located from the data pool collected Non-snoring subjects were matched to snoring subjects based upon age, gender and race. The best match of the same sex and race, closest in age to the snoring subject was selected.

Cephalometric Data

The investigator traced each cephalometric radiograph and recorded a combination of eleven linear and one angular measurement (Figures 1-3, Glossary). Thirteen of the measurements were based upon the literature and previous work performed at Case Western Reserve University. The measurement of the most narrow posterior airway space was developed by the investigator.

A three tenths millimeter lead pencil was used to trace the cephalometric radiographs on three thousands of an inch thick matte acetate paper (Masel, Bristol, PA). All landmarks were located by the same investigator and all measurements were corrected for magnification before calculation. The mean enlargement factor of the cephalometric radiographs was 7.5% for both experimental and control groups.

Twelve (19%) of the cephalometric radiographs were retraced by the investigator to determine tracing error. A period of not less than two weeks passed between initial tracing and retracing for landmark error identification. The two radiographs provided from outside the Cleveland area were mixed with the twelve radiographs that were traced to determine landmark identification error. All eleven linear and the one angular variable of the second tracings were measured by the same investigator and compared to the same variables of the first tracings. Intra-Class

Correlation Coefficients were used to examine the reproducibility of landmark identification.

The linear measurements recorded include: Hyoid to mandibular plane (H-MP), soft palate length (P-PNS), vertical airway length (VAL), basion to nasion (BaN), sella to nasion (SN), posterior nasal spine to A-point (PNS-A), tongue length (TL), superior, posterior airway space (S-PAS), inferior, posterior airway space (I-PAS), narrow, posterior airway space (N-PAS), gonion to gnathion (Go-Gn) The only angular variable measured was the cranial base alignment (MCF). See the Glossary for a description of each measurement and definition of anatomic landmarks.

Questionnaire Data

The questionnaire presented to the parents or guardians was one page long and consisted of eleven Likert-type scale questions (never, rarely, sometimes, often, unsure), and seven yes/no questions. One question asked parents or guardians of snoring children, at what age they began to snore. The survey format was adopted from the work of Ali et al. (1993). The responses to the questions on the sleep survey were analyzed. Relationships between the answers and the snoring and non-snoring populations were examined.

Data Analysis

Cephalometric Analysis

In addition to traditional skeletal cephalometric landmarks, some soft tissue structures of the upper respiratory compartment were included in the present study.

Definitions of landmarks and measurements are presented in the Glossary. A portion of Enlow's counterpart analysis was used to analyze cranial base rotation.

Body Mass Index

Each subject's height and weight was converted to kilograms and meters in order to calculate Body Mass Index [weight/height^2,(kg/m^2)].

Statistical Analysis

The objective of the study was to compare cephalometric measurements of children that snore "often", to children that "never" snore. The specific aim was to identify craniofacial variables that differ between snoring and non-snoring children.

The independent variables measured on a continuous scale included the following: Twelve anatomic variables derived from cephalometric radiographs, age and Body Mass Index (kg/m^2). Gender was determined as a dichotomous variable.

The analysis for the present study proceeded as follows:

1. Descriptive Statistics
   Means and standard deviations were calculated for all continuous independent variables.

2. Comparison of Equality of the Means
   Paired t-tests were used for comparison of equality of the means between the two groups of snoring and non-snoring children. A p-value of less than 0.004 using Bonferroni's correction [α of 0.05 divided by the total number of variables (0.05/12=0.004)] was used to determine statistical significance. All computations were performed using Microsoft® Excel computer program for Windows and the Statistical Package for the Social Sciences (SPSS-PC+ for Windows).

3. Questionnaire Analysis

   The relative significance of the categorical data from the sleep questionnaire to the experimental and control groups, was examined statistically. The Chi-Square test was used to test for equality of proportions between questionnaire responses and subject category.

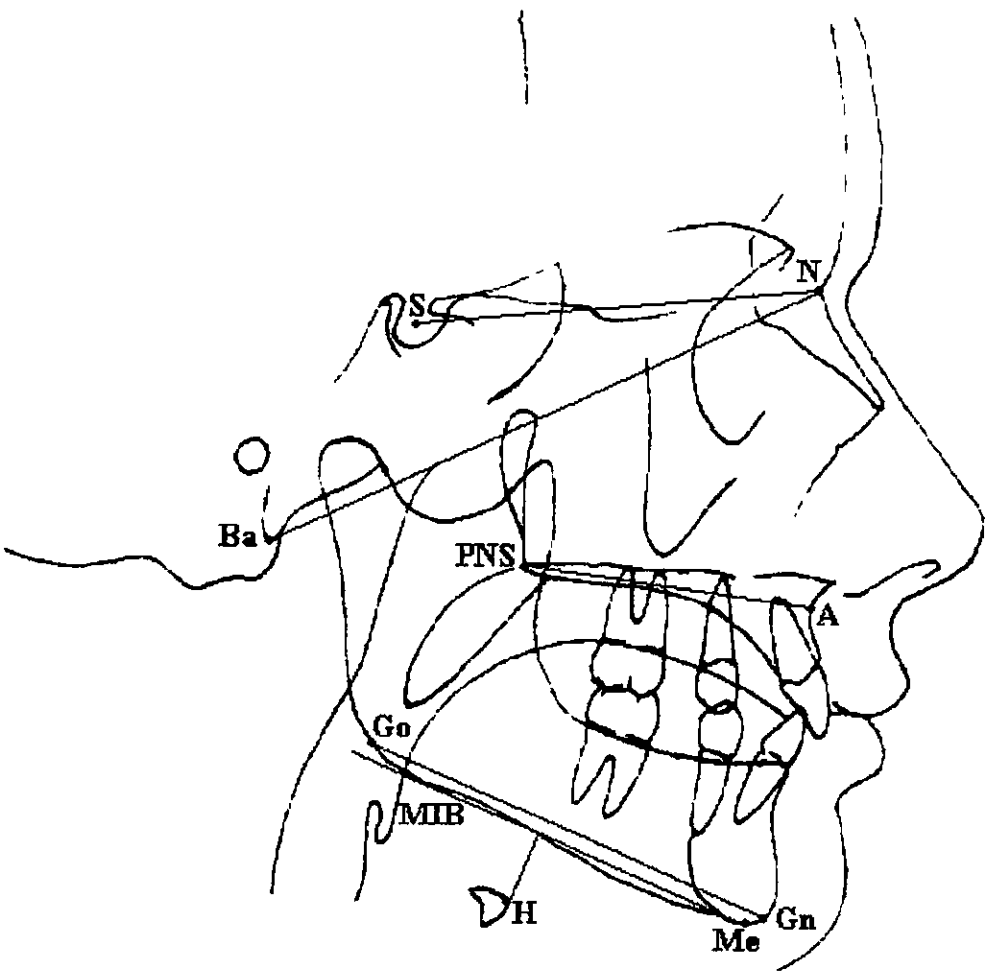


Figure 1. The diagram illustrates five conventional hard tissue linear measurements and the mandibular plane. (Refer to Glossary for an explanation of the abbreviation shown)

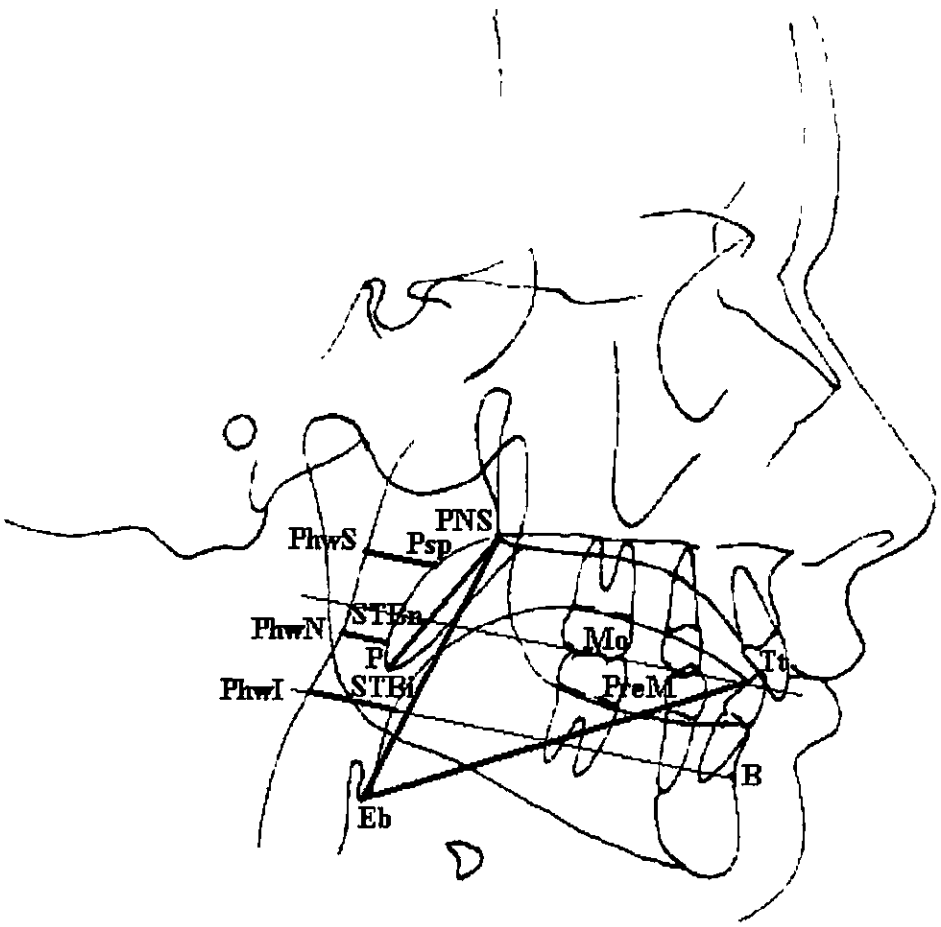


Figure 2. The diagram illustrates six soft tissue linear measurements and the occlusal and B point-gonion reference planes. (Refer to Glossary for an explanation of abbreviations)

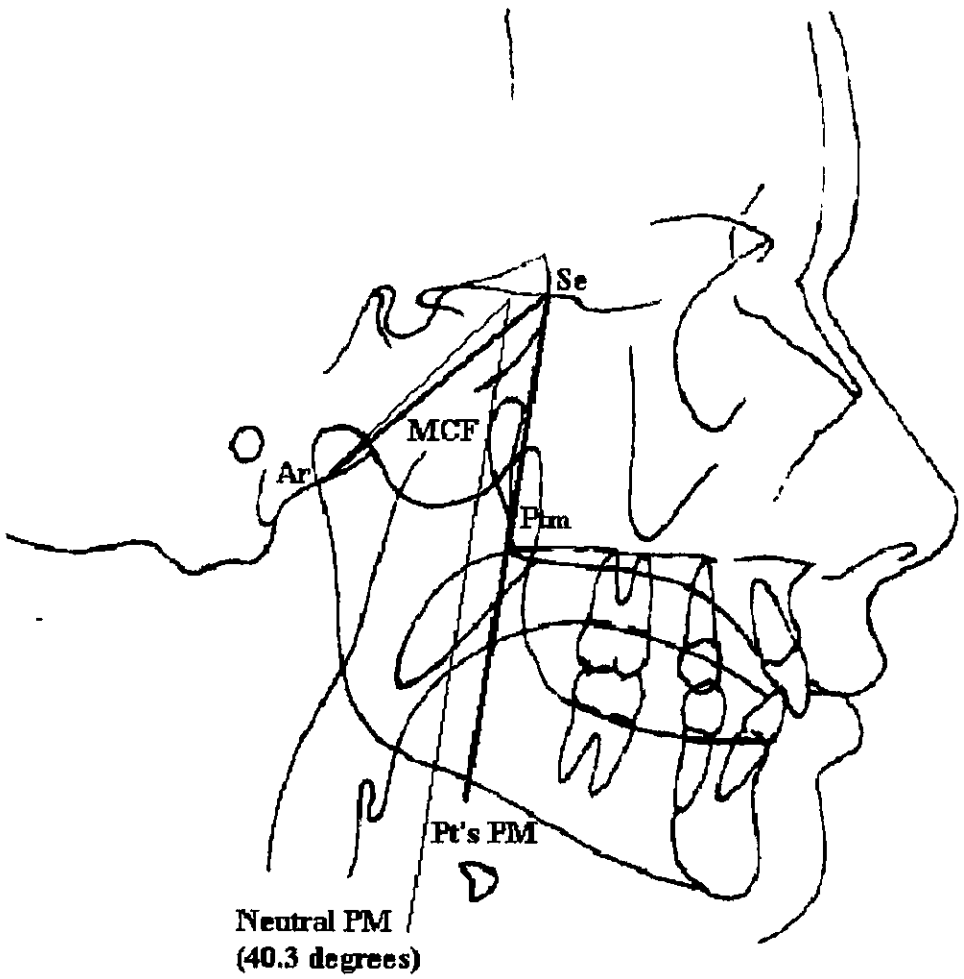


Figure 3. MCF component of the Counterpart Analysis. This diagram demonstrates the effect of a rotational alignment of a subject's own cranial base (heavy line Ar-SE-Ptm) compared to the "neutral" position (light line Ar-SE). A clockwise rotation is show suggesting protrusion of the maxilla. (Refer to Glossary for an explanation of abbreviations)

RESULTS

A total of 228 parents/guardians were surveyed regarding their child's sleep behavior. Thirty-six children snored "often". However, from a pool of seventy-four non-snoring subjects, it was only possible to accurately match thirty-one pairs on age, sex and race.

The sleep questionnaire presented parents with multiple responses to the question of snoring. The parents were given the option to select from the following five choices when asked if their child snored: Never, rarely, sometimes, often or unsure. To ensure the matched sample consisted of non-snorers, only subjects with a reported history of having "never" snored were selected. See Table 3 for a distribution of the total responses to the question of snoring presence.

TABLE 3.- -Responses to the Question, "Does your child snore at night?"

	Never	Rarely	Sometimes	Often	Unsure	Total
Responses	74	70	46	36	2	228
%	32	31	20	16	1	100

Every effort was made to restrict age variation to within six months. For example, if an experimental (snoring) subject was twelve years and zero months old, a control (non-snoring) subject of the same sex and race was chosen that fell within the age range of eleven years six months to twelve years six months. The average age

difference in this study was ± 3.6 months and the range was from 0-8 months. Of the thirty-one subject pairs three matched perfectly, however, one control was eight months younger than the experimental subject and two other subject pairs differed in age by seven months.

Snoring and non-snoring (control) subjects were matched based upon subject age, sex and race. Subjects were grouped as either Caucasian or African-American. However, one Indian female served as a control for a snoring Caucasian female.

The study population was sixty-one percent female, and was predominantly Caucasian (80%). The average age of the study population was ten in both the snoring and control groups, with a standard deviation of two years. The Body Mass Index (BMI) was significantly greater in the snoring subjects (20.3 ± 4.4) compared to the non-snoring group (17.7 ± 3.4). The average age of onset of snoring was three years of age and ranged from birth to nine (mode 0, median 2.5).

Cephalometric Assessment

Equality of means for cephalometric variables between snoring and non-snoring subjects was tested using paired t-tests. Statistical significance was determined using Bonferroni's correction at $p < 0.004$. Comparison of means for the twelve cephalometric variables between the snoring and non-snoring subjects are presented in Table 4.

TABLE 4.- -Comparison of Means of Cephalometric Variables Between Snoring and Non-snoring Subjects

Variables	Snoring Subjects	Non-snoring Subjects	'p value
H-MP	16 95±4 31	13 22±5 07	0 001*
P-PNS	30 75±4.03	29 57±3 77	0.252
VAL	58.64±6.96	54 97±7 26	0 011
Ba-N	97 97±6 31	99 58±5 71	0.200
SN	65 19±3 46	66 75±4.40	0 069
PNS-A	43 41±3 19	45 03±3 05	0 024
TL	70 67±6 06	69 74±6 04	0 579
S-PAS	8 05±4 13	11 87±3 78	0.002*
I-PAS	12 22±2.94	11 68±2 96	0 555
N-PAS	5 06±2.76	8 00±2 40	0 000*
Go-Gn	66 47±5 24	68 63±5 79	0 093
MCF	-0.207±3 19	0 584±2 48	0.257

* significant at p <0.004

Craniofacial variables that differed significantly between snoring and non-snoring subjects were determined. The variables presenting the most significant difference between the groups included the distance from the hyoid bone to the mandibular plane, the most narrow anterior-posterior measure of the posterior airway space and the anterior-posterior airway space measured from the posterior wall of the pharynx to the most posterior-superior point on the soft palate. Also significant were the measures of the vertical airway length (VAL) from the base of the epiglottis to the posterior nasal spine, and posterior nasal spine to A-point (PNS-A).

The magnification of the radiographs from CWRU School of Dentistry and the twelve private practices ranged from two to fourteen percent.

Questionnaire Assessment

Chi-Square testing of the responses to the sleep questionnaire revealed significant differences in some key questions between snoring and non-snoring subjects (Tables 5 and 6). Over 93% of the snoring responses were "sometimes" or

"often" when asked if the child tended to breath through their mouths rather than their nose. In the control group, 70% "never" or "rarely" breathed through their mouths. Sixty-four percent of snoring subjects slept with their heads tipped back "sometimes" or "often", when 77% of non-snorers never did, and another 20% did so rarely. When asked if their child had problems related to sleeping, nearly 50% of snoring group children had problems, whereas less than 10% of controls responded the same. It was also observed that 50% of snorers have trouble getting to or staying asleep, and are restless sleepers "sometimes" or "often". In contrast, control subjects responded with "never" and "rarely" nearly 80% of the time. A significant number of snoring children had a history of having been told their adenoids were enlarged, but the same was not true for tonsils. Responses favored snoring children having a strong tendency toward having difficulty paying attention in school, falling asleep during the day, and getting coughs and colds. Based upon responses, it was also more expected to have had tonsils and/or adenoids removed, and to have morning headaches in the snoring children. When inquiring at what age the snoring children began to snore, 74% of subjects began at age four or younger.

TABLE 5 - -Questionnaire responses (Never, Rarely, Sometimes, Often) %Experimental/ %Control

Question	N	R	S	O	x^2
Restless sleeper?	17/32	37/45	13/16	33/7	0.058
prob. Getting to or staying asleep?	36/55	19/26	19/16	26/3	0.068
fall asleep during the day?	48/74	26/16	13/10	13/0	0.088
Hyperactive?	74/74	10/13	10/7	7/7	0.951
Frequent coughs and colds?	13/29	29/45	45/23	13/3	0.067
sleep with head tipped back?	18/77	18/20	43/3	21/0	0.000*
mouth breathing?	4/40	4/30	18/20	75/10	0.000*
morning headaches?	45/71	29/23	26/7	0/0	0.059
difficulty with attention in class?	32/59	32/28	26/7	10/7	0.122

*Significant at $p < 0.05$

Key for TABLE 5

KEY. N=Never, R=Rarely, S=Sometimes, O=Often	x^2 = Chi-Square Distribution

TABLE 6..- -Questionnaire responses. (Yes, No)

QUESTION	SNORING		NON-SNORING		x^2
	%YES	%NO	%YES	%NO	
Tonsils or adenoids removed?	10	90	0	100	0.075
Told tonsils were enlarged?	32	68	13	87	0.075
Told adenoids were enlarged?	38	62	7	93	0.004*
Problems with hearing?	13	87	10	90	0.688
Taking medications?	26	74	20	80	0.589
Problems related to sleeping?	48	52	10	90	0.000*

*Significant at $p < 0.05$ x^2 = Chi-Square Distribution.

Measurement Reproducibility

Table 7 demonstrates the means and standard deviations of the twelve cephalometric measurements traced at time 1 (T1) and time 2 (T2). Intra-Class Correlation Coefficients for measurement reproducibility were calculated. There was a highly significant correlation between the measurements made at time 1 and time 2. The least reliable variable was the measurement from P to PNS.

TABLE 7 - - Measurement Reliability

Variable	Intra-Class Correlation Coefficient*
H-MP	0.99
P-PNS	0 75
VAL	0 94
Ba-N	0 82
SN	0 98
PNS-A	0 81
TL	0 99
S-PAS	0 90
I-PAS	0.92
N-PAS	0 98
Go-Gn	0 99
MCF	0 96

*reliable at 0 75

DISCUSSION

The objective of the present study was to identify cephalometric differences between snoring and non-snoring children 7-14 years of age. Since the "empirical" age range of most children presenting for orthodontic evaluation is between the ages of seven and fourteen, that was the range selected for subject inclusion in the present study. Only thirty-six (15%) out of two hundred twenty-eight subjects surveyed answered "often". This percentage is slightly higher than the results of previous investigations of snoring prevalence in children (Ali et al. 1993; Ali et al. 1994; Owen et al. 1996). In a study of Italian children aged 6-13, the prevalence of snoring "often" was found to be 7.3% (Corbo et al. 1989). The above average percentage of snoring children might be attributed to selection bias introduced in gathering the experimental subjects. One of the twelve contributors of study subjects submitted completed questionnaires and cephalometric radiographs on snoring subjects only. No attempt was made to randomly survey patients as they entered the practice. The total contribution to the study from that practice consisted of two experimental subjects.

The onset age of snoring in the present study (mean = 34.6 mos ± 30 mos) was somewhat higher than the results of Brouillette et al. in 1982. They examined over seventy infants and children with obstructive sleep apnea and found that the

mean age at onset of breathing difficulties, including snoring, was 14 months ± 12 (Brouillette et al. 1982). Most of the subjects in the Brouillette et al. study were under the age of five. The younger sample population probably accounts for the variation in age between the two studies.

Body Mass Index (BMI) was observed to be significantly greater in the snoring, than in the non-snoring children (20.3 ± 4.4 vs 17.7 ± 3.4). There is agreement with the literature that BMI in adults is significantly increased in snoring and apneic populations (Partinen et al. 1988; Zucconi et al. 1992; Zucconi et al. 1993; Nelson and Hans 1997). However, others have found BMI to be of no significance in their investigations of adults (Maltais et al. 1991; Olson et al. 1995). In an attempt to distinguish snoring children from apneic children, Carroll et al. (1995) found no significant differences between the groups with regard to obesity. Contrary to the finding in the present study, some suggest that snoring, and apneic children, may be underweight or underdeveloped (Brouillette et al. 1982; Guilleminault et al. 1982).

In the present study, to ensure the control population consisted of non-snorers, only subjects having a reported history of having "never" snored were selected as controls. It is believed that substantial overlap between snoring and non-snoring subjects is unlikely. Control subjects were matched to the experimental subjects based upon age, sex and race. A "best match" principle was used to pair controls to experimental subjects. For each snoring subject, a control was selected that most closely matched the age of the snorer. The average age difference between experimental and control subjects was 3.6 months. The advantages of matching

subjects on age is that it eliminates the influence of changes brought about by aging, for example: Changes in lung volume, fat deposition in the upper airway, increases in muscle mass and pubertal growth changes, including reduction in lymphoid tissue mass.

Twelve cephalometric variables reported to be important based upon the literature and previous studies at CWRU were measured. Nine of the variables were measures of hard tissue landmarks, three were measures of the soft tissue pharynx. The width of the pharyngeal airway at it's most narrow dimension (N-PAS) was constructed by the investigator. No previous measure of this dimension could be found in the literature. Based upon the pathophysiology of snoring, and the relative significance of pharyngeal constriction in snoring and obstruction (Croft and Pringle 1991; Isono et al. 1996), the investigator determined a measure of the width of anterior-posterior pharyngeal constriction at it's most narrow dimension to be important.

In comparing the means of the twelve cephalometric measurements, three variables demonstrated statistical significance. Those variables included the distance from the hyoid to the mandibular plane (H-MP) and two of the three measures of pharyngeal width (S-PAS and N-PAS). Numerous investigators support the finding of an increased hyoid to mandibular plane distance (Partinen et al. 1988; Maltais et al. 1991; Zucconi et al. 1992; Zucconi et al 1993; Nelson and Hans 1997; Shintani et al. 1992). Regarding the significant reduction in posterior airway space observed in the present study, Brouillette et al. and others, attributed the narrowing of the airway to

tonsil and adenoid hypertrophy, and anatomic abnormalities (Brouillette et al. 1982; Shintani et al. 1992). In the present study, significant differences were observed in the width of the posterior airway space between experimental and control subjects, but only at two of the three pharyngeal airway measurement points. Significant differences were observed at the most superior (S-PAS) and the most narrow (N-PAS) airway space measurements. No significant difference was observed in the posterior airway space (I-PAS) measured along the B-point to gonion plane. Zucconi et al. (1992) is in agreement with such findings. To the contrary, Partinen et al. (1988) found a narrowed posterior airway space measured along the B-point to gonion plane to be predictive of increased respiratory disturbance when associated with an increased hyoid to mandibular plane distance.

The present study results also support, with statistical significance, an increase in the length of the airway from the base of the epiglottis to the posterior nasal spine (VAL), and a shortened maxilla (PNS-A) in snoring subjects. Pae et al. (1997) claim that an extended airway results in narrowing and turbulent airflow. Others have observed neck extension to act favorably on nasorespiratory function (Huggare and Laine-Alava 1997; Schwartz et al. 1996). The fact that snorers had a shortened maxilla (PNS-A) may suggest a narrowing in the sagittal dimension. However, one would also anticipate other sagittal measures such as S-N, Go-Gn and Ba-N to be significantly reduced also. In the present study, there was a trend for all three sagittal measures to be shorter in the experimental group. Bacon et al. demonstrated a significant reduction in the sagittal dimension of the anterior cranial base in apneic

adults (Bacon et al. 1989). Zucconi et al. (1993) demonstrated a reduction in cranial base and mandible in snoring adults, and a shorter maxilla in apneic adults. They suggested habitual snorers might have an anatomic predisposition to airway obstruction. Whether, or not, snoring subjects in the present study represent a population of apneic children has not been determined. However, in a recent study of snoring children, it was discovered that the detection of apnea every night by the parents was the most significant historical risk factor for obstructive sleep apnea. In addition, a history of restless sleep every night, previous adenoidectomy, constant snoring, and enlarged tonsils were also important signs and symptoms of risk for obstructive sleep apnea in children (Nieminen et al. 1997).

Two hypotheses for the development of adulthood obstructive apnea exist at present. One, a developmental hypothesis, suggests that an extended history of constant snoring and apnea results in physiologic changes evidenced in adults as anatomic variation from the normal. Numerous studies have shown differences in cephalometric measures between normal non-apneic, snoring and non-snoring adults, and subjects with obstructive sleep apnea (Zucconi et al. 1992; Zucconi et al. 1993). In such cases the abnormality of the cephalometric findings may be the consequence of having had a long-standing obstructive airway problem.

Another hypothesis suggests that the cephalometric differences between apneics and normals existed prior to the exacerbation of apnea, and prior to aging, weight increase, alcohol consumption and other precipitating factors.

The present study tests the second hypothesis to a degree. Since it is not known which, if any, of the snoring child subjects present symptoms of obstructive sleep apnea, it is assumed that those presenting the most severe snoring histories are at significantly greater risk of an apneic future than subjects which have no history of snoring. Snoring is universal in children with symptomatic obstructive sleep apnea (Gastaut 1966; Lugaresi et al 1978; Westbrook 1983; Brouillette et al. 1982; Nieminem et al. 1997). Therefore, by testing for differences in cephalometric variables in snoring and non-snoring children, one may be able to speculate that the anatomic variation exists from childhood. Since most (90%) of the growth of the craniofacial skeleton is obtained by the age of twelve, 60% by age four (Krogman 1973); and the average age of the present study population was ten, one can speculate that facial growth and development might have already been modified, especially in the presence of airway obstruction early in life. Based on the above, one could conclude that the best cohort to study, would be one of an age where minimal craniofacial development has occurred. The best group would be one in which the influence of age on the craniofacial skeleton has been little. Such a population would likely be well under the age of four. Shintani et al. (1992) studied young apneic children (avg. age 4.5) and determined that the children with the most severe apnea had larger adenoids and a narrower airway. The best responders to adenotonsillectomy treatment had larger adenoids and tonsils, and low set hyoid bones. In cases where lower hyoid bone position was observed, skeletal morphology was a likely contributor to airway obstruction. Brouillette et al. (1982) also

demonstrated nasopharyngeal and oropharyngeal constriction in infants and children with respiratory disorders

The most significant differences between snoring and non-snoring children, in the present study, regarding questionnaire responses were: mouth breathing, sleeping with the head tipped back, problems related to sleeping, enlarged adenoids, restless sleep and frequent morning headaches. There was also a tendency for snoring children to have trouble getting to, or staying asleep, having frequent coughs and colds, falling asleep during the day, having enlarged tonsils, and having had tonsils and/or adenoids removed.

As reported in previous studies (Ali et al. 1993; 1994), the present study also found a tendency for restless sleep, and a trend for daytime sleepiness in snoring children. But, unlike Ali's results, no relationship between hyperactivity and snoring was observed. The significance of a relationship between hyperactivity and snoring in Ali's study may be attributed to his sample size. Ali's sample sizes were much larger (782 in 1993, and 507 in 1994) in relation to the present study sample (31).

The thirty-one snoring subjects in the present study tended to have had their tonsils and/or adenoids removed, have been told either or both were enlarged and restless sleep. Nieminen et al. (1997), while testing snoring children, found that those experiencing restless sleep, adenoidectomy, and enlarged tonsils, present greater risk for having obstructive sleep apnea.

A striking similarity in percentages of symptoms was observed between a small sample (25) of children referred to a Sleep Disorders Clinic (Guilleminault et

al. 1982), and the outcomes of the responses to the questionnaire in the present study. In both studies, the percentage of subjects with excessive daytime sleepiness was over 26%, learning problems over 36%, frequent morning headaches and special sleeping positions greater than 24% and 40%, respectively.

In the present study, there was a tendency for snoring children to have had a history of tonsillectomy and/or adenoidectomy. Ten percent of snoring children had tonsils and/or adenoids removed, whereas none of the non-snoring subjects had surgical intervention. Nieminen et al. (1997) believed adenoid tissue did not play a critical role in the development of obstructive sleep apnea. The results of their research actually demonstrated a greater risk of being apneic when adenoids were removed. Others believe the adenoid tissue plays an important role in the development of clinical symptoms (Suen et al. 1997). The present study also revealed a significant trend toward having been told the child's adenoids were enlarged; they had also been told their tonsils were enlarged but not as frequently. Nieminen et al. (1997) found that children with enlarged tonsils also had greater risk of being apneic.

A large percentage of snoring children were said to "breath through their mouths", "sometimes" or "often" (93%). Only thirty percent of control subjects did the same. Carroll et al. (1995) also found a high percentage of mouth breathing in both snoring and apneic children. A history of daytime sleepiness was not demonstrated in that study. However, morning headache was observed in twenty-three percent of snoring subjects, apneic subjects had none.

Although the present study revealed no significant differences in hearing problems between snoring and non-snoring children, Owen et al. (1996) found a significant association between snoring children and poor hearing, restless sleep and having a cold.

Limitations of the Study

Sample size was limited by the number of experimental subjects that met the criteria of the study. Size was also restricted by the timeframe over which the data was collected. As with most studies, a larger sample may have demonstrated greater variability in results and increased reliability. Since the sample was gathered predominantly within the local Cleveland area, it can conceivably be assumed that the sample in the local geographic area might not be representative of the target population of interest. If so, this would be an example of sample distortion bias. Also, the potential for social class to confound the results existed and might have altered results. Selecting a sample in accordance with stringent selection criteria, limited to healthy children devoid of craniofacial abnormalities and no prior orthodontic/orthopedic treatment helped in controlling for bias. Matching on age, sex and race, and blinding of the investigator during measurements additionally limited confounding biases. Although the presence of morning headaches was significantly greater in the experimental group, an underestimation might be present since children frequently do not report such pains.

Since the cephalometric radiographs used in the study came from multiple sources, they varied in quality. The radiographs of poor diagnostic quality were

eliminated from the study. This was done in approximately fifty cases. In each case, questionnaires were not obtained from the subject's parents or guardians. Although radiographic quality could be controlled, the same could not be said for head position during exposure. Hellsing observed a change in hyoid bone position and increased cross-sectional dimension of the retroglossal pharynx with a change from natural head position to a twenty degree extension (Hellsing 1989). The radiographs used in the present study were all exposed on cephalometers incorporating ear rods and nasion rests to approximate the Frankfort horizontal plane parallel to the floor.

Use of cephalometric radiographs provides only a static two dimensional image in the sagittal plane. Inferences to three dimensional anatomic structures cannot be made based solely upon the cephalometric image. The value of the cephalometric radiograph in the study of the head and neck and associated pathologies, is unsurpassed. It is widely accessible and relatively inexpensive in comparison to alternative imaging procedures. Information obtained from cephalometric assessment has been invaluable in a great number of studies in airway pathology, including snoring and sleep apnea. The present study tests the utilization of the cephalometric radiograph in comparing snoring and non-snoring children.

SUMMARY AND CONCLUSIONS

This cross-sectional study attempted to test the hypothesis that craniofacial differences exist between snoring and non-snoring children. Over a period of ten months, cephalometric radiographs and questionnaire data was collected on a total of sixty-two snoring and non-snoring children. Cephalometric variables and questionnaire responses between the two groups were compared. Statistical significance comparing the equality of the means between the groups was determined.

Analysis of the data yielded the following results:

1. The anterior-posterior measure of the pharyngeal airway was narrower in snoring children at the superior and most narrow widths of the pharynx.
2. The hyoid bone was positioned more inferior in snoring children.
3. The body mass index was greater in snoring children.
4. The airway tended to be extended in snoring children.
5. The craniofacial sagittal dimension tended to be shortened in the snoring group.

The results of the questionnaire data suggest that the most significant differences between snoring and non-snoring children are:

1. Snoring children slept with their heads tipped back.
2. Snoring children breathed through their mouths.

3. Snoring children had enlarged adenoids and problems related to sleeping.

4 There was a tendency for snoring children to·

- have trouble getting to or staying asleep

- get frequent coughs and colds

- sleep restlessly

- get frequent morning headaches

- fall asleep during the day when not active

- have enlarged tonsils, and

- have had tonsils and/or adenoids removed.

In conclusion, findings from the present study indicate that there are cephalometric differences in snoring and non-snoring children. It is hoped that the present study may stimulate further research in the area of childhood respiratory disease.

APPENDEX A

Human Investigation Consent Form

HUMAN INVESTIGATION CONSENT FORM

My child has been asked to participate in a research study entitled: *Cephalometric Assessment of Children With and Without Sleep Related Problems*

This study is being conducted by: Randall W. Kulnis, D.M.D., Suchitra S. Nelson, M.S., Ph. D., Mark G. Hans, D.D.S., M.S.D. and Kingman P. Strohl, M.D. from Case Western Reserve University School of Dentistry.

I understand the purpose of the study is to test if children with sleep related problems have the same facial features on an x-ray as children that do not have sleep related problems. I also understand this study involves a sleep and behavior questionnaire.

I further understand that:

1. The sleep questionnaire will require answers to questions about my child's sleep and other associated behaviors.

2. The x-rays taken at my child's records appointment will be measured by one of the investigators. No x-rays will be taken specifically for this study.

3. The information from the study is confidential and will be contained in a data base for future study.

4. Dr. Kulnis, Dr. Nelson , Dr. Hans and Dr. Strohl will be coordinating this study and will be available to answer any questions about the study. The telephone numbers in the Department of Orthodontics are (216) 368-3469 and 368-4649.

5. I have the right to refuse to participate in this study. In which case, my refusal will not affect any care given at this facility.

I have read and understand the above information and hereby give my consent for my child to participate in the study of children with and without sleep related problems.

_____ _____

Name of Child *Date*

_____ _____

Signature of Parent/Legal Guardian *Relationship to Child*

APPENDEX B

Sleep Behavior Study in Children
Demographics Form

SLEEP BEHAVIOR STUDY IN CHILDREN

ID#:_____ DATE:_____
departmental use

PARENT INFORMATION

NAME:_____
first *middle initial* *last*

OCCUPATION:_____ _____
father *mother*

ADDRESS:_____
street *city*

_____
state *zip* *apt. #*

PHONE:_____home_____work RACE:_____

SMOKERS:
father- ___yes___no
mother- ___yes___no
other- ___yes___no

CHILD INFORMATION

NAME:_____
first *middle initial* *last*

AGE:_____ DOB _____ SEX:_____M_____F

WEIGHT:_____lbs. HEIGHT ____ft.____in.

APPENDEX C

Sleep Behavior Questionnaire

Circle *one* for each question.
PLEASE ANSWER ALL QUESTIONS IN REGARD TO YOUR CHILD

I.D.#_____

1. Does your child have a problem related to sleeping? Yes No
   If yes, please specify_____
2. Is your child a restless sleeper? Never Rarely Sometimes Often Unsure
3. Does he/she have trouble getting to or staying asleep at night? Never Rarely Sometimes Often Unsure
4. Does he/she fall asleep during the day when not active? (for example watching TV or reading) Never Rarely Sometimes Often Unsure
5. Is he or she considered to be hyperactive? Never Rarely Sometimes Often Unsure
6. Does he/she get frequent coughs and/or colds? Never Rarely Sometimes Often Unsure
7. Does he/she snore at night? (even without a cold?) Never Rarely Sometimes Often Unsure
   If "often", at what age did snoring start? _____
8. Do you consider the volume of the snoring to be... Quiet Moderate Loud Very Loud Unsure
9. Does your child sleep with their head tipped back? Never Rarely Sometimes Often Unsure
10. Does your child tend to breath through their mouth rather than their nose? Never Rarely Sometimes Often Unsure
11. Has your child had his/her tonsils or adenoids taken out? Yes No
12. Have you ever been told that your child has enlarged tonsils? Yes No Unsure
    enlarged adenoids? Yes No Unsure
13. Does your child get frequent morning headaches? Never Rarely Sometimes Often Unsure
14. Does your child have any problems with their hearing? Yes No Unsure
15. Does your child have difficulty paying attention in class? Never Rarely Sometimes Often Unsure
16. Is your child currently taking any medications? Yes No
    If yes, please specify_____
17. Is your child currently wearing braces/retainer? Yes No

APPENDEX D

Orthodontist's Agreement to Participate in the Study

Dear parent,

A study investigating sleep behavior in children is being conducted by Dr. Randall W. Kulnis, an orthodontic resident at Case Western Reserve University's School of Dentistry. Dr. Kulnis has been an outstanding student of mine for the past 12 months; and has asked for my support in completing his Master's Thesis. He is a fine young man and I am happy to help him attain his goals.

Dr. Kulnis has been given my permission to send you a copy of the sleep questionnaire enclosed with this letter. Please do the best you can and circle the most appropriate answers regarding the sleep behavior of your child. Since some families have multiple children under treatment at the same time, please complete the survey only on the child named at the top of the questionnaire.

Within the next few days you can anticipate a phone call from Dr. Kulnis or his wife about this sleep questionnaire. You will also be asked the approximate height and weight of your child.

Your cooperation is greatly appreciated. If you have questions please call Dr. Kulnis at Case Western Reserve University or at home. You may also contact my office during regular office hours.

Thanks again for your help.

APPENDIX E

Participating Private Practice Orthodontists

OFFICE	ADDRESS
Dr. L. Alvetro	Sidney, Ohio
Dr. B.D. Amberman (1)	Vermillion, Ohio
(2)	North Ridgeville, Ohio
Dr. N. DeLoach, Jr.	Cleveland Heights, Ohio
Dr. J. B. Dworkin	University Heights, Ohio
Dr. H.E. Kossoff (1)	Solon, Ohio
(2)	Mentor, Ohio
Dr. S. Simpson	Hudson, Ohio
Dr. G. A. Souris	North Olmsted, Ohio
Dr. T. L. Wenger	Mayfield Heights, Ohio
Dr. J. White (1)	Aurora, Ohio
(2)	Independence, Ohio

APPENDEX F

Approval of University Review Committee on Human Studies

CASE WESTERN RESERVE UNIVERSITY
REVIEW COMMITTEE FOR HUMAN STUDIES
NOTICE OF APPROVAL

TO: **Nelson, Suchitra**
Kulnis, Randall W.

Dept/School: **Dentistry**

Subject: Notice of (X) Review and approval
( ) Exemption

The committee has reviewed and approved your proposal entitled: **Cephalometric Assessment of Children with and without Sleep Related Problems**

Please be advised that with respect to: (1) the rights and welfare of the individual(s) involved, (2) the appropriateness of the methods to be used to secure informed consent, and (3) the risks and potential benefits of the investigation, the Committee considers your project:

( ) Exempt
(X) Fully acceptable
( ) Acceptable with reservations noted
( ) Not acceptable for reasons noted

Follow-up: The Committee wishes to have a status report on this project on **9/11/1998**

SIGNED FOR THE COMMITTEE BY:

Approval Date:
9/25/1997

Charlotte L. Horton
Director
Office of Research Administration

CZ

ORA USE ONLY**********

Type Project: (X) NEW ( ) RENEWAL
Human Risk: ( ) Yes ( ) No
Source of Support: ( ) Outside Funding (X) Department/other
Agency (Potential): Agency No.

Are any of the following involved: (X) Yes ( ) No
If yes, which category(ies):

(X) Minors () Fetuses () Abortuses () Pregnant Women
() Prisoners () Mentally Retarded () Mentally Disabled

cc: Dean
Department Chairman
Follow-up (Book)
File

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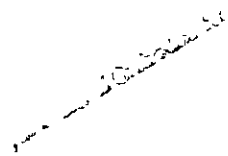
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