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The Validation of an Existing Sleep Apnea Questionnaire and its Application to Clinical Orthodontics

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THE VALIDATION OF AN EXISTING SLEEP APNEA QUESTIONNAIRE AND ITS APPLICATION TO CLINICAL ORTHODONTICS

By

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A Thesis Submitted in Partial Fulfillment of the Requirements of the Degree Master of Science in Orthodontics

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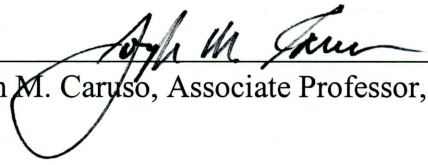
Each person whose signature appears below certifies that this thesis in their opinion is adequate, in scope and quality, as a thesis for the degree Master of Science.



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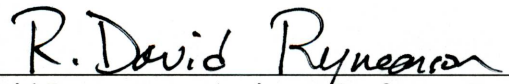

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ABSTRACT

THE VALIDATION OF AN EXISTING SLEEP APNEA QUESTIONNAIRE AND ITS APPLICATION TO CLINICAL ORTHODONTICS

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The purpose of this study was to validate a sleep apnea questionnaire from the Loma Linda University Sleep Disorders Center, and then to utilize the questionnaire in order to identify potential sleep apnea patients from a pre-orthodontic patient sample. Study patients were selected from the Loma Linda University Sleep Disorders Center and the Loma Linda University Orthodontics Clinic, both in Loma Linda, California, USA.

Methods: The study sample represented all of the patients initially examined from 1996 through 1998 who were age 8-15 years old at the time of the initial exam. Validation of the questionnaire was based on responses to questions of these 90 patients from the sleep center, irregardless of the diagnosis, whose parents responded to the questionnaire. To assess potential sleep apnea patients, 65 patients, age 8-15, were selected from the Sleep Center archives, all of whom completed the Loma Linda Sleep Disorders Center Questionnaire (LLUSDCQ). Their responses on the LLUSDCQ, were compared to 65 patients, ages 8-15, from the Loma Linda Orthodontic Clinic, who were under orthodontic evaluation.

From the LLSDCQ, questions of clinical utility were then identified as significant in their ability to predict sleep apnea correctly. These questions were then analyzed statistically for their sensitivity and specificity at uniquely predicting sleep apnea. Pre-treatment orthodontic patients with a strong indication for sleep apnea were also identified and orthodontic records were then assessed, on the basis of the results from the questionnaire.

Results indicated that, neither gender nor age, predicted sleep apnea when using the Normal Approximation to Binomial statistical test. A Logistics Regression Model found that a combination of questions 2 (Snoring or noisy breathing), 3 (Observed times with no breathing), and 8 (Mouth breathes while awake) resulted in a statistically significant ability to predict sleep apnea correctly. (R^2=56.5 ; $p < .05$).
The incidence of sleep apnea predicted by the questionnaire in the pre-treatment orthodontic patients was 4.6%, an incidence roughly double that expected in this age group. Similarities in orthodontic patient records were examined in those patients identified, particularly the dolichofacial pattern characteristics, suggesting an aggregation of craniofacial characteristics which may additionally be important in the identification of sleep apnea patients.

In summary, the LLUSDCQ may assist in the identification of sleep apnea patients in the orthodontic population age range examined. Evaluation of orthodontic records may be coordinated with the LLUSDCQ to help clinicians and other health professionals to make an appropriate referral to sleep centers with confidence.

I. INTRODUCTION

As clinicians, we see there is an ongoing demand to continue to better our diagnostics skills and improve our ability to treat patients. Interaction and exchange of knowledge with other clinicians and medical professionals has allowed us to begin to respond to these demands. Clinicians strive to increase their ability to identify patients with health risks, and subsequently diagnose, treat or appropriately refer that patient. Continual efforts are made to come up with simple, inexpensive diagnostic tools to aid in this process.

Over the past several years, there has been a shift in attention given to patients who suffer from sleep apnea. Much of the attention has been extended to the child and adolescent as more of these younger patients are being identified. Since the majority of an orthodontist's practice is comprised of children and adolescents, it seems imperative that there be some interaction and exchange of knowledge with a sleep disorder specialist to aid in the identification, diagnosis and treatment of such young patients. There has already been an increase in this interaction with sleep disorder specialists with regard to the adult patients.

Orthodontists recognize that there is a rise in the number of oral appliances being developed for patients suffering from excessive snoring and sleep apnea. Often, the orthodontist is being called on for the fabrication and insertion of such oral devices for the treatment of these patients. Because of the increased rate of mortality in patients with sleep apnea, it is important for the orthodontist to identify those potential patients, assist in the diagnosis of all potential patients, and make an appropriate referral.

To do so, it is necessary for the orthodontist to have the appropriate information available to them regarding the number of patients who may potentially suffer from the consequences of sleep apnea. It is desirable to have a simple method of identifying those potential individuals, as well as having identifiable parameters that can be evaluated and subsequently assist in an appropriate referral to a sleep center.

The purpose of this study was to statistically validate the Loma Linda Sleep Disorders Center Questionnaire, and in particular identify specific variables (questions), which may predict patients in an orthodontic population (age 8-15 years.) with a strong indication for a positive diagnosis of sleep apnea. After validating this questionnaire, the goal was then to find the incidence of potential sleep apnea patients in a pre-treatment orthodontic patient sample. Finally, records of orthodontic patients, identified as having the potential for sleep apnea, would be compared to non-sleep apnea orthodontic patients' records.

II. REVIEW OF THE LITERATURE

Over a hundred years ago, Dr. W. Hill, 1889, was probably describing manifestations of Obstructive Sleep Apnea (OSA) when he stated "the stupid looking lazy child who frequently suffers from headaches at school, breathes through his mouth instead of his nose, snores and is restless at night, and wakes up with a dry mouth in the morning, is well worthy of the solicitory attention of the school medical officer."[1] In 1973, sleep apnea as a syndrome (Obstructive Sleep Apnea Syndrome, OSAS), was first described by Guilleminault, et al.[2] OSAS is a syndrome of disordered breathing characterized by a combination of intermittent partial and/or complete airway obstruction occurring during sleep, leading to hyperventilation (hypoxia and hypercapnia) and sleep disturbance. Depending on the degree of hyperventilation and/or sleep disturbance, certain physiologic abnormalities may be seen including failure to thrive, cor pulmonale, and excessive daytime sleepiness (EDS). In addition, learning problems, developmental delay, and hyperactivity may be seen in children and adolescents.[3] It is suggested that OSA is part of a continuum of sleep disorders which also includes snoring and Upper Airway Resistance Syndrome (UARS).[4]

Sleep apnea is a common condition affecting about 2 % to 4 % of the population in the United States.[5] Young et al, reports that sleep apnea remains undiagnosed in at least 82% of men and 93% of women with the condition.[6] Snoring is more common and affects approximately 50% of the males and 30% of females. Snoring probably may be a pre-clinical state for the development of OSAS,[7] and is the most common symptom of childhood OSAS. Snoring occurs in 10-12% of school age children,[8-10] and the incidence of sleep apnea is estimated at 1-3%.[11,12]

Childhood and adolescent OSAS differs from OSAS in adults in several important ways.[13-15] (Table 1)[16]

	Childhood	Adult
Gender	Males equal females	Males more than females
Obesity	+	++++
Common Presentation	Snoring	Hypersomnolence
Arousals	+/-	++++
Respiratory Pattern	Obstructive Hypoventilation	Obstructive apnea
Tonsils/Adenoids	Common	Rare

Table 1. Childhood vs. adult OSAS: clinical features. (Adapted from [Rosen C, 1996])

The peak incidence of sleep apnea between 2-6 years corresponds to the peak for normal lymphoid hyperplasia.[16] In school age children, the male/female ratio is approximately equal. In contrast to the predominance of obesity in adult sleep apnea patients, the majority of children with sleep apnea have a normal weight, however, it has been identified in obese children or even children who fail to thrive. Snoring is the most common presenting symptom in children in contrast to excessive daytime sleepiness in adults.[16]

A. Etiology

Over the last decade, considerable work has focused on various anatomic and physiologic factors in the search for an etiology and pathophysiology of sleep apnea. Anatomic factors such as tonsillar hypertrophy, micrognathia and macroglossia all narrow the pharyngeal airway, which leaves the airway more susceptible to collapse.[17]

Physiologic factors, such as upper airway hypotonia induced by changes in conscious state, alcohol, or sedative hypnotic consumption, may also precipitate upper airway collapse in sleep.[17] Any factor that decreases pharyngeal size or increase pharyngeal compliance might be expected to predispose an individual to sleep apnea. As it has been seen, children with sleep apnea share certain clinical features with adults, but there remain important differences in the clinical presentation, diagnosis, and treatment which suggests that although the pathophysiology is similar, it is probably not the same.[18]

B. Clinical Manifestations

Some clinical manifestations shown to be evident in children with sleep apnea include learning problems, morning headaches, frequent upper airway infections, failure to thrive or obesity.[19] Some nocturnal symptoms include difficulty breathing while asleep, heavy snoring, apneic episodes, restless sleep, heavy sweating, nightmares, night terrors and enuresis.[19] It appears as though the reason for seeking consultation tends to vary with age.

In children younger than five years, difficulty breathing while asleep, nightmares and night terrors are more frequent reasons for consultations than in older children. This may be because parents may check young children's sleep more often, and young children fall asleep earlier than older children, so parents have a greater chance to note abnormal sleep behavior.[19]

In children older than five years, excessive daytime sleepiness (EDS) (associated with complaints of tiredness and daytime fatigue), abnormal daytime behavior, learning

disabilities, frequent morning headaches, nocturnal enuresis, and major discipline problems are more common reasons for consultations.[19] A few children are referred at a late stage of the syndrome; they not only present with significant failure to thrive, but also may have been hospitalized for unexplained acute cardiac failure or unexplained development of systemic hypertension.[19]

C. Sleep Apnea Diagnosis

Sleep apnea diagnosis is based on the clinical symptoms and signs as well as physical examination.[20] A careful history is taken to determine if the sleep apnea is secondary to another syndrome that could be at least partially responsible for the sleep-related upper airway occlusion. Regardless of the combination of symptoms, a patient should be evaluated thoroughly. The following should be investigated in an evaluation of the oronaso-maxillofacial region; the aspects of the nares, collapse of the nostril with inspiration, the size of the nose, the tongue's size and consistency; the length of the tongue's protrusion, the width of the mouth, the length and position of the soft palate, the presence of an abnormal amount of lymphoid tissue, enlargement of the tonsils and adenoids, the presence of retrognathia, micrognathia, cleft palate, or repair of cleft palate, infiltration of oropharyngeal soft tissues by storage disease, and any other areas which may seem pertinent to the individual.[19]

D. Imaging Techniques

Cephalometric radiographs may help distinguish subtle bony abnormalities that may predispose to sleep apnea.[21] CT,[22] MRI[23] and direct visualization by laryngoscopy[24] have also been suggested to image the pharynx, and may be useful in determining the best treatment option for the individual. As an example of imaging the pharynx, Schwab has shown an illustration comparing a mid-sagittal MR image of a normal subject (A) and a patient with sleep apnea (B). Soft palate and tongue area, are larger in the apneic patient.[25] (Figure 1)

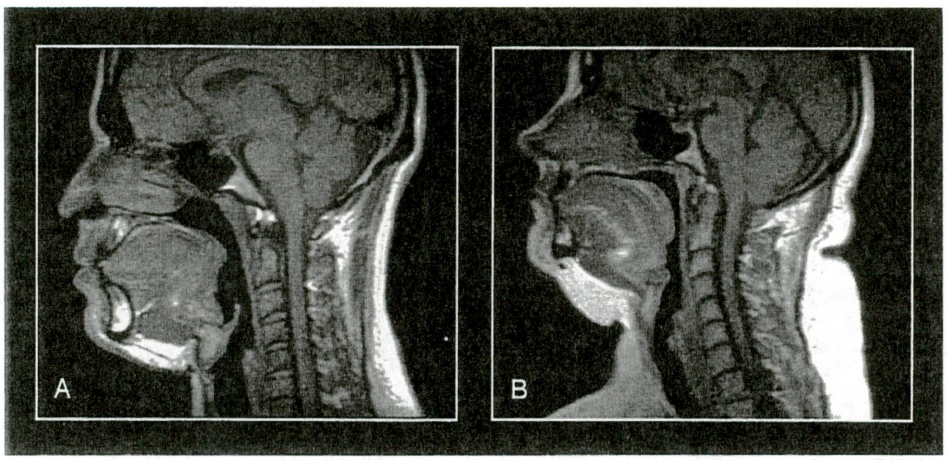

Figure 1. A. MRI of normal airway. B. Airway of patient with sleep apnea. (Adapted from [Schwab, 1998]).

E. Polysomnogram (PSG)

Although the history and physical examination are important in suggesting the diagnosis of sleep apnea, the best test to confirm the diagnosis of sleep apnea is the polysomnogram (PSG).[26] This all-night study usually includes electroencephalography,

electrooculography, and chin electromyography (EMG) to monitor the stages of sleep; measurement of airflow at both the nose and mouth; assessment of chest wall and abdominal motion by inductance plethsymography or strain gauges to assess "paradoxical" breathing and differentiate obstructive from non-obstructive events; electrocardiogram (ECG); oximetry; and pretibial EMG leads to detect motion.[26] The nocturnal polygraphic recording provides information on a number of sleep-related parameters: the number and duration of complete or partial obstructions per hour of sleep, the lowest SaO_2 during the night, the presence and type of cardiac arrhythmia's, and the presence and severity of respiratory disturbances and their impact on the cardiovascular system.[26] An important aspect of a PSG is the assessment of sleep continuity as well as the information it provides regarding the severity of the sleep disruption.[27]

The PSG also measures the number of respiratory events, their duration, and their effect on oxygen saturation. Respiratory events are traditionally defined as apneas (no airflow) and hypopneas (partial airflow). Both can cause arousal from sleep and significant oxygen saturations. A measure commonly used is the Respiratory Distress Index (RDI=apnea+hypopnea), which is the number of respiratory events per hour, a normal RDI is five or less.[28]

Polysomnography is indicated in all children suspected of having a significant disorder of breathing during sleep in order to confirm the clinical impression, determine the severity of the disorder, and establish the need for therapy. Polysomnography can be labor intensive, and often, facilities with expertise in evaluating young children may not

be available.[16] It has been shown that given the incidence of OSAS and the estimated number of children who undergo adenotonsillectomy for OSAS, there simply are not enough sleep laboratory beds for all the patients who would benefit from this assessment. Therefore, various "screening" studies such as home audio or video taping, overnight oximetry, or overnight sonography have been suggested, but these tests are limited for several reasons: 1) failure to detect obstructive hypoventilation; 2) failure to detect hypercapnia or establish the cause of the hypoxia; 3) inability to distinguish between central and obstructive apenea; and 4) they give no information about sleep disruption.[29] On the other hand, missing the diagnosis because of reliance on an insensitive screening tool extends the child's exposure to the consequences of sleep apnea.[30] The role of these less time and resource consuming approaches in children is currently being investigated, but at present, PSG is the diagnostic tool of choice for OSA.[16] The recommendation from the International Classification of Sleep Disorder (ICSD) committee states, "In the young child, the signs and symptoms of obstructive apnea are more subtle than in the adult, therefore, the diagnosis is more difficult and should be confirmed by polysomnography."[31]

F. Therapy

For children and adults with sleep apnea, the approaches to therapies are quite different. However, there are incidences where a type of treatment used for an adult sleep apnea patient may also be used effectively for a child with sleep apnea. Factors to be considered in deciding on the vigor of treatment include polysomnographic measures,

such as the RDI and the extent of the oxyhemoglobin desaturation, as well as clinical factors, such as daytime sleepiness, etc.[32] Intervention may be suggested in borderline cases where the severity of the patient's snoring is interfering with normal family dynamics. Treatment strategies can be divided into behavioral, medical, surgical, and nasal continuous positive airway pressure. Many children simply require behavioral modification including weight loss and avoidance of alcohol or sedatives (commonly found in over-the-counter cold preparations) at bedtime.[32] Respiratory stimulants such as progesterone or nicotine have had limited success in adults, and are rarely indicated in children because of potential side effects.[32] Protriptyline may be helpful in select cases by decreasing REM sleep or by selectively stimulating pharyngeal muscles, however, its side effects limit its usefulness.[32] Possible approaches to therapy in children are listed in Table 2.[3]

The recent focus on oral appliances in mild to moderate sleep apnea is an important treatment option for patients not wanting CPAP or surgery.[33] Recent investigations, using an adjustable mandibular Herbst appliance for treatment of sleep apnea, found that part of the mechanism for patient improvement appeared to be a forward and upward movement of the hyoid bone to the mandibular plane. These appliances have been devised to reposition the mandible, shift the tongue forward, or otherwise enlarge the pharyngeal air space. However, because of the effects these appliances may have on the craniofacial complex, these appliances may not be indicated in children.[33] Cistulli et al, suggest that Rapid Maxillary Expansion (RPE), may be a useful treatment alternative for select patients with sleep apnea.[34] Adenotonsillectomy

I. Medical Management
   A. Nasopharyngeal airway
      1. Acute
      2. Temporary
   B. Continuous positive airway pressure (CPAP)
   C. Weight loss
   D. Pharmacologic treatment
      1. Respiratory stimulants
      2. Antihistamines
      3. Steroids
         a. Oral
         b. Nasal
      4. α-Agonists
II. Surgical management
   A. Adenotonsillectomy
   B. Tracheostomy
   C. Maxillofacial plastic surgery
   D. Uvulopalatopharyngoplasty (UPPP)
III. Oral Appliance Therapy
   A. Mandibular Repositioning Devices (MRD)
   B. Herbst Appliances
   C. Rapid Maxillary Expansion (RPE)

Table 2. Therapy of obstructive sleep apnea in children. (Adapted from [Loughlin GM, 1992])

remains the most common treatment for sleep apnea in children. In children, selection of the procedure must take into account the child's potential for growth. Even though the craniofacial skeleton has attained 60% of its adult size by age 4 years, facial growth continues until about age 15 in girls and age 19 in boys.[35] The risk of altering facial growth by surgery must be considered, especially since changes in facial structure with growth may lesson the severity of the syndrome. Procedures such as mandibular advancement and hyoid resuspension have been successful in select adults with sleep

apnea, but many surgeons are reluctant to perform these procedures on children whose facial structures are still growing and changing.[36]

Uvulopalatopharyngoplasty (UPPP) is an excellent surgical treatment for snoring, however, only about half the patients have significant improvement in apnea,[37] and only 9% have complete resolution of sleep apnea.[33] Adenotonsillectomy remains the oldest and most common means of treating sleep apnea in children, and is effective in as many as 90% of the patients without craniofacial or neurological abnormalities.[38] It can result in prompt remission of cor pulmonale[39] and improvement in school performance.[1] However, it does not affect central apneas or hypopneas, and it appears that patients with the most severe disease do not respond as well as patients with a more mild condition. A baseline PSG is essential to determine the severity of the individual patient and the likelihood of a good response to surgery. Children with severe sleep apnea at baseline, as well as those with persistent symptoms, should have overnight polysomnography about one month after adenotonsillectomy to insure resolution of their apneas.[18] Lymphoid tissue changes with age, and mild sleep apnea may resolve spontaneously with normal regression of the tonsils and adenoids. Conversely, sleep apnea may reappear several years following apparently successful adenotonsillectomy.[40] It is also wise to observe the patient closely in the immediate postoperative period, because the combination of sedation and surgical edema may exacerbate obstructive events.[32]

The most common non-surgical treatment for sleep apnea is CPAP. This "pneumatic splint" results in prompt improvement in cardiac function,[41] hypersomnolence, and in adults, results in superior cure rates to surgery.[42] CPAP has been

employed successfully in children with craniofacial abnormalities as well as normal children as young as 15 months.[43] Children under 2 years of age accept and tolerate CPAP well.[44] With proper education of the patients and their families, excellent long-term compliance can be achieved. CPAP may be particularly useful, while other long term treatments such as weight loss are being implemented, for patients who decline surgery or for patients with an unacceptably high surgical risk.[32]

G. The orthodontist, sleep apnea, and cephalometric analysis

Recently, sleep apnea in the juvenile population has received attention[35,19] It has been suggested that upper airway patency while asleep is relevant to the craniofacial growth pattern.[19] Orthodontists may be the first specialist to detect some of the clinical symptoms of sleep apnea when children visit them for orthodontic treatment, and orthodontist can evaluate the trend of the craniofacial development as the child grows. Evaluating cephalometric radiographs may be useful for selecting which patients with sleep apnea are potentially suitable for oral appliance therapy and which appliance to use.[45] It is suggested that the Mandibular Repositioning Device (MRD) is useful in the long-term treatment of patients with OSA of mild to moderate severity for adults,[46] but there has been no consensus on its use in children.

Cephalometrics have been used to assess OSA patients, however, it is important to note that two-dimensional measurements do not necessarily correlate with three-dimensional measurements of the craniofacial and upper airway structure.[47] In cephalometric measurements of the hard tissue, the following morphologic characteristics

have been found to be true of patients with sleep apnea.[48] The maxilla[53,49] and the smaller[50,51] mandible[52,53,54] are retropositioned; the anterior face height [50,53,55,49,54,56] increased; the occlusal[56,57] and the mandibular[53,54,56] plane angles are enlarged; the maxillary and mandibular molars are overerupted[53] and incisors are proclined[53]; and the overbite is decreased.[53] In measurements of the soft tissue, the following morphologic characteristics have been found in patients with sleep apnea: The tongue,[53,58] the soft palate,[52,59,60,56,61,58] and the pharyngeal length,[60,58] are elongated and the soft palate is thickened;[60,58] the anteroposterior pharyngeal space is reduced[53] at the superior, middle and inferior[52,59,49,60,58] levels; the cross sectional areas of the tongue[49] and soft palate[49,58] are enlarged; and those of the oropharynx[49] and hypopharynx[49,58] are reduced. Moreover, an inferior position of the hyoid bone has frequently been documented.[62,52,59,49,54,63,61,58]

III. METHODS AND MATERIALS

The design of this investigation was divided into four sections: (1) Validation of the Loma Linda Sleep Disorders Center Questionnaire; (2) distribution of the same questionnaire to pre-treatment orthodontic patients in order to identify potential sleep apnea patients; (3) based on the results of the validation and identification of pre-treatment orthodontic patients with potential sleep apnea , an incidence of pre-treatment orthodontic patients with potential sleep apnea would be determined; (4) evaluation and comparison of orthodontic patient records, which were determined to be potential sleep apnea patients, with "normal" pre-treatment orthodontic patient records.

A. Validation of the Loma Linda Sleep Disorders Center Questionnaire: (LLSDCQ)

The LLSDCQ is a diagnostic screening tool utilized at the Loma Linda University Children's Hospital Sleep Disorders Center. This questionnaire is given to each parent of the patient, at the patients' initial visit. The questionnaires are filled out by the parents, in which they are asked to score the following 13 questions from 1 to 10, where lower scores indicate "never," middle scores indicate "sometimes" and higher scores indicate "always." (See Appendix A)

Variable 1 - Difficulty Breathing

Variable 2 - Snoring or Noisy Breathing

Variable 3 - Observed Times with no Breathing

Variable 4 - Irritability (fussy)

Variable 5 - Sleepiness During the Day or Excessive Napping

Variable 6 - Poorer School Performance than Expected

Variable 7 - Restlessness, Tossing , Turning

Variable 8 - Mouth Breathes While Awake

Variable 9 - Tonsil infections or Sore Throats

Variable 10- Sweating During Sleep

Variable 11- Gasps for air During Sleep

Variable 12- Choking During Sleep

Variable 13- Turns Pale or Blue

The patients in this study were selected at random from patients who were referred to the Loma Linda University Children's Hospital Sleep Disorders Center, due to some sort of underlying signs or symptoms. Initially, 90 subjects were selected at random with the patients' diagnosis unknown at the time of selection. The scores to the thirteen questions from each patients questionnaire were recorded. Each patient was then diagnosed as either positive or negative for sleep apnea, regardless of the severity, by Dr. Ralph Downey III, director of the Sleep Disorders Center. Age and gender of each patient was recorded. The purpose of this portion of the investigation was to determine which questions were statistically significant in their ability to predict a positive or negative diagnosis for sleep apnea, as well as to determine if gender or age have any significance on the outcome of the diagnosis.

Statistically significant questions, as well as clinically significant questions, were identified. Clinically significant questions were identified by Dr. Ralph Downey III,

based on his years of clinical experience diagnosing patients with sleep disorders. Statistically significant questions were identified using Logistic Model for Analysis. Logistic Model for Analysis was used to determine which combination of clinically and statistically significant questions would result in a statistically significant combination of questions, in their ability to predict the correct diagnosis for sleep apneic patients. Next, a random sample of 65 patients, all confirmed positive for sleep apnea, were identified for the purpose of determining a baseline score for the identified significant questions. The patients in this sample were selected if their ages were between 8-15 years old. This range was selected because the baseline score would be compared to a pre-treatment orthodontic sample and because this age range represents a common age range of patients in an orthodontic practice. The subjects in this sample were selected from patients who were initially examined between 1996 and 1999.

B. Distribution of the LLSDCQ to pre-treatment orthodontic patients parents

The same questionnaire, (LLSDCQ), was distributed to parents of pre-treatment orthodontic patients at the Loma Linda University Orthodontics Department. The parents completed the questionnaire at the patients' initial visit for orthodontic treatment. Sixty five patients were selected for this sample to coordinate with the 65 positive apnea patient sample. The patients selected were also between the age of 8-15 years old. Scores for the statistically significant question were determined, for the purpose of comparison to the positive sleep apnea sample baseline scores. Pre-treatment orthodontic patients, whose response scores to the significant questions were greater than or equal to

the positive sleep apnea patients response scores, were identified. For this investigation, these patients were considered as positive for sleep apnea, based on their response scores to the statistically significant questions.

C. Incidence of sleep apnea patients in the pre-treatment orthodontic sample

The incidence of patients with sleep apnea in the pre-treatment orthodontic patient sample was determined. Incidence was based on the number of pre-treatment orthodontic patients identified with sleep apnea, via the LLSDCQ, out of the total number of pre-treatment orthodontic patients.

D. Evaluation and comparison of pre-treatment orthodontic patient records

Pre-treatment orthodontic patient records were taken at the Loma Linda University Orthodontic Department. Orthodontic records were obtained from pre-treatment orthodontic patients identified as positive for sleep apnea, as well as from an equal number of patients identified as negative for sleep apnea. The records obtained included: Lateral and frontal cephalograms, panorex, full mouth series, maxillary and mandibular occlusal radiographs, intraoral and extraoral imaging, 2 view temporal mandibular joint radiographs, and maxillary and mandibular study models. The same qualified operator made all the tracings and measurements on the patients' records. The lateral and frontal cephs were traced and digitized into the "Quick Ceph" software program for cephalometric analysis. Cephalometric measurements were based on the Ricketts' Analysis and the Jarabak Analysis.

Those patients identified as positive for sleep apnea, based on the results of the questionnaire, were compared with "normal" patients identified as negative for sleep apnea. Several areas of evaluation were compared, and listed in Table 3.

Area of Evaluation	**Determination of Measurement**
Facial Type	Dolichofacial, Mesofacial, or Brachyfacial
Angle Classification	Class I, Class II Div.1, Class II Div. 2, or Class III
Maxillary Arch Form	From Ricketts' Pentamorphic Arches
Mandibular Arch Form	From Ricketts' Pentamorphic Arches
Maxillary Inter-molar Width	Mesial pit to mesial pit.
Maxillary Inter-canine Width	Cusp tip to cusp tip
Maxillary Depth	Angle between FH plane and Na-A-point plane.
Facial Depth	Angle between FH plane and Facial plane
Facial Axis Angle	Angle between Facial Axis and Na-Ba plane
Saddle Angle	Angle between Ar-Sella and Sella-Na
Mandibular Plane Angle	Angle between FH plane and Mandibular plane
Lower Facial Height	Angle from ANS to Xi to Po
Total Facial Height	Angle between Corpus Axis and Na-Ba plane
Nasal Cavity Width	The widest aspect of Nasal cavity
Maxillary Width	The distance between "J" points
Tonsils and Adenoids	Present or absent

Table 3. Orthodontic records measurements.

IV. RESULTS

A. Validation of the Loma Linda Sleep Disorders Center Questionnaire: (LLSDCQ)

For the purpose of maintaining confidentiality of the subjects throughout this investigation, each patient was given a subject number. Gender and age of each patient was recorded and labeled such that males were identified as "1," and females were identified as "2." The initial sample of 90 patients were identified and given subject numbers. Then, each question listed was identified as a variable, where for example, question 1 would equal variable 1. The response to each variable was recorded as it was answered on the questionnaire. Dr. Ralph Downey III, then blindly confirmed the diagnosis for all 90 subjects, and the diagnosis was recorded. Those subjects determined as being positive for sleep apnea were identified as "1" and those diagnosed as negative for sleep apnea were identified as "2." Of the 90 subjects included, 37 out of 90 subjects (41.1%) were female, and 53 out of 90 of the subjects (58.9%) were male. (See Table 4) There was, 59 out of 90 of the subjects (65.6%) diagnosed as positive for sleep apnea, and 31 out of 90 of the subjects (34.4%) diagnosed as negative for sleep apnea. (See Table 5)

Gender	Frequency	Percent
Female	37	41.1
Male	53	58.9

Table 4. Gender. (N=90)

Diagnosis	Frequency	Percent
0	31	34.4
1	59	65.6

Table 5. OSA Diagnosis: "0"=Neg, "1"=Pos. (N=90)

To determine if gender or age of the patient had any significance on the outcome of the diagnosis, the Normal Approximation to Binomial Test was performed. Based on the binomial experiment, there is no statistically significant difference between males and females with regard to the outcome of the diagnosis. It was also determined that the age of the patient in this sample had no statistically significant affect on the outcome of the diagnosis.

The next portion of the investigation was to identify which questions were significant in their ability to predict the correct diagnosis. The questions identified as clinically significant, were determined by Dr. Ralph Downey III. Based on experience, Dr. Downey has identified several of the questions as clinically significant in their ability to identify potentially positive patients. Dr. Downey has generally considered anything higher than a 6 on those questions to indicate a patient as potential for sleep apnea. He has identified questions 1,2,3,5,6 and 8 as clinically significant questions in their ability to identify potential sleep apnea patients.

Statistically significant questions were determined using a Logistic Model for Analysis. Based on this analysis, it was determined that the combination of question 2 (Snoring or noisy breathing), question 3 (Observed times with no breathing), and

question 8 (Mouth breathes while awake), would result in a statistically significant ability to predict the diagnosis correctly. ($R^2 = 56.5$).

The literature reports a prevalence rate of sleep apnea, for patients in our samples age range, to be between 1-3%.[11,12] Therefore, a prevalence rate of 2% was utilized in the analysis for determining sensitivity and specificity for question 2, question 3, and Question 8. The sensitivity and specificity percentages are based on a Likelihood Ratio (LR) of approximately 4, and are shown in table 6.

		Percentages		
Variable	Prevalence Rate	Sensitivity	Specificity	LR ≅ 4
Question 2	0.020	73.4	81.5	
Question 3	0.020	65.6	84.6	
Question 8	0.020	45.3	87.7	

Table 6. Sensitivity/specificity.

The above findings, that questions 2, 3, and 8 are statistically significant in their ability to correctly diagnose patients with sleep apnea, were then utilized to compare a sample of positive patients for sleep apnea with a sample of pre-treatment orthodontic patients.

As mentioned earlier, a second sample of patients was selected from the Loma Linda University Children's Hospital Sleep Disorders Center. There were 65 patients in this sample, and the patients selected into this sample must have met certain criteria to be included. All of the patients selected must have been between the age of 8-15 years old at the time the questionnaire was distributed and filled out. There must have been a

response to all 13 questions, and all patients in the sample must have had a positive diagnosis for obstructive sleep apnea, as diagnosed by Dr. Ralph Downey III.

The response to questions 2, 3, and 8 were recorded for each of the subjects in the sample. A sum of the scores to these three questions, were figured for each subject. For the 65 subjects, the mean score for the sum of these three questions was figured to be 19.9. The frequency of responses to questions 2, 3, and 8 were also analyzed for each of the patients in this sample. Positive patients for sleep apnea frequently scored 6 or above on their responses to the three questions. Dr. Downey has also used a score of 6 or greater for the questions as an indication of a positive diagnosis. Based on the frequency of responses to questions 2, 3, and 8, it was determined that a combined score of 18 or greater would be indicative of a positive diagnosis for sleep apnea for this study. A sum of the scores for the three questions was utilized, because of the fact that questions 2, 3, and 8, were determined to be statistically significant when combined. Due to the closeness of the frequency of responses (18 or greater) to that of the mean score for the sum of the three questions (19.9), a minimum score of 18 or greater was selected to be used as the score for comparison with the pre-treatment orthodontic patient sample.

B. Distribution of the LLSDCQ to pre-treatment orthodontic patients parents

In order to identify pre-treatment orthodontic patients with potential for sleep apnea, responses to LLSDCQ were obtained from 65 pre-treatment orthodontic patients from the Loma Linda University Orthodontic Department. All patients in this sample

were also between the ages of 8-15 years old at the time the questionnaire was completed. Responses to all 13 questions were required.

The sum of questions 2, 3, and 8, were figured for all 65 patients in this sample. Patients with a combined score of 18 or greater were identified and determined as positive for sleep apnea, based on their score.

C. Incidence of sleep apnea patients in the pre-treatment orthodontic sample

Based on a LLSDCQ score of 18 or greater for questions 2, 3, and 8, the results indicate that three patients were identified as positive for OSA. This indicates that the incidence of pre-treatment orthodontic patients in this sample positive for sleep apnea, is 3 out of 65, or 4.6 %. This results in an incidence rate almost two times as high as the general population.

D. Evaluation and comparison of pre-treatment orthodontic patient records

Pre-treatment orthodontic records were obtained from the three patients identified as positive for sleep apnea. Pre-treatment records were also obtained from three patients who scored the minimum score possible for questions 2, 3, and 8. These patients were identified as negative for sleep apnea based on their responses. The measurements for the areas of evaluation for both the positive and negative group of patients are presented in Tables 7 & 8 respectively.

Area of Evaluation	Patient #1	Patient # 2	Patient # 3
Facial Type	Dolichofacial	Dolichofacial	Dolichofacial
Angle Classification	Class II Div. 1	Class II Div. 1	Class II Div. 1
Maxillary Arch Form	Narrow Tapered	Narrow Tapered	Narrow Tapered
Mandibular Arch Form	Narrow Ovoid	Narrow Tapered	Narrow Tapered
Maxillary Inter-molar Width	44 mm	39 mm	44 mm
Maxillary Inter-canine Width	31 mm	26 mm	31 mm
Maxillary Depth	90.3° (Norm 90°)	89.0° (Norm 90°)	87.5° (Norm 90°)
Facial Depth	85.8° (Norm 88.8°)	85.9° (Norm 87.5°)	85.4° (Norm 87.7°)
Facial Axis Angle	85.9° (Norm 90°)	85.9° (Norm 90°)	82.3° (Norm 90°)
Saddle Angle	130.1° (Norm 123°)	129.0° (Norm 123°)	129.5° (Norm 123°)
Mandibular Plane Angle	27.1° (Norm 23.7°)	25.1° (Norm 25°)	28.4° (Norm 24.8°)
Lower Facial Height	47.5° (Norm 45°)	52.4° (Norm 45°)	51.2° (Norm 45°)
Total Facial Height	61.4° (Norm 60°)	63.7° (Norm 60°)	64.6° (Norm 60°)
Nasal Cavity Width	27.4mm (Norm 30)	26.0mm (Norm 27.2)	26.7mm (Norm 27.7)
Maxillary Width	63.5mm (Norm 65.8)	54.7mm (Norm 63.5)	62.8mm (Norm 63.7)
Tonsils and Adenoids	Present	Present	Present

Table 7. Positive patients for sleep apnea. (Score = 18 or >)

Area of Evaluation	Patient #1	Patient # 2	Patient # 3
Facial Type	Brachyfacial	Brachyfacial	Mesofacial
Angle Classification	Class II Div. 1	Class II Div. 1	Class I crowding
Maxillary Arch Form	Tapered	Narrow Ovoid	Normal
Mandibular Arch Form	Tapered	Normal	Narrow Ovoid
Maxillary Inter-molar Width	43 mm	40mm	45 mm
Maxillary Inter-canine Width	33 mm	34 mm	34.5 mm
Maxillary Depth	87.3° (Norm 90°)	93.7° (Norm 90°)	90.4° (Norm 90°)
Facial Depth	87.1° (Norm 87.6°)	90.7° (Norm 88.3°)	90.2° (Norm 88.3°)
Facial Axis Angle	88.6° (Norm 90°)	90.0° (Norm 90°)	89.1° (Norm 90°)
Saddle Angle	128° (Norm 123°)	122.0° (Norm 123°)	116.9° (Norm 123°)
Mandibular Plane Angle	24.1° (Norm 24.9°)	20.9° (Norm 24.2°)	26.6° (Norm 24.2°)
Lower Facial Height	41.6° (Norm 45°)	42.8° (Norm 45°)	46.4° (Norm 45°)
Total Facial Height	59.1° (Norm 60°)	54.6° (Norm 60°)	60.0° (Norm 60°)
Nasal Cavity Width	29.1mm (Norm 27.5)	30.2mm (Norm 28.9)	29.8mm (Norm28.9)
Maxillary Width	62.4mm (Norm 63.7)	63.2mm (Norm 65)	62.4 mm (Norm65)
Tonsils and Adenoids	Present	Present	Present

Table 8. Negative patients for sleep apnea. (Score = 3)

V. DISCUSSION

Just as Temporomandibular Dysfunction once went unnoticed, unidentified, undiagnosed, and untreated in orthodontic patients, sleep apnea has also gone unnoticed because of the lack of information and diagnostic tools available to the orthodontist. Orthodontists have been given the information necessary to help understand the importance of properly identifying and diagnosing TMD in orthodontic patients. Orthodontists need additional information to help understand the potential implications of improperly identifying and misdiagnosing potential sleep apnea patients in their practice.

This investigation has given the orthodontists both information, and a diagnostic tool, (LLSDCQ), to assist in the identification and diagnosis of potential sleep apnea patients. Orthodontists can also utilize this information and diagnostic tool to make a referral to the appropriate health professional with confidence. It has been shown that the responsibility of the orthodontist has been expanded to assisting in the treatment of sleep apnea patients, via Mandibular Repositioning Devices, Herbst appliances, and Rapid Palatal Expansion. Because of this, it is important that orthodontists continue to communicate and exchange knowledge with other health professionals regarding patients with sleep disorders. Since the orthodontist may be one of the few, or only, health professionals seen by the patients over an extended period of time, it seems we may become required legally, and ethically to attempt to identify these patients.

The importance of identifying children and adolescents with potential sleep apnea has been shown due to some of the potentially significant sequelae such as medical complications, growth and development disturbances, and poor educational development.

Valid, reliable, simple and inexpensive clinically diagnostic tools need to be made available to aid in this identification and referral process. This valuable questionnaire has been shown to be important in its ability to identify potential sleep apnea patients. It can help identify individuals needing further sleep studies that may not have been identified in the past.

It has been shown, based on the results of this investigation and patient sample, that the incidence of sleep apnea patients in this orthodontic sample is approximately 4.6% of the patients. Since the estimated prevalence rate of sleep apnea in school aged children is between 1-3%, this investigation indicates that the number of potential sleep apnea patients in an orthodontic office may be greater than that of the general population. This would indicate that a potentially large number of orthodontic patients are going undiagnosed for sleep apnea. As mentioned earlier, a large number of undiagnosed patients have been seen in adults, where sleep apnea remains undiagnosed in at least 82% of men and 93% of women with the condition.[6]

Due to this investigation, it has been possible to identify positive and negative pre-treatment orthodontic patients for sleep apnea, and compare their orthodontic records. The results of these comparisons suggest several areas, which may be more consistently related to positively identified patients. Although not statistically analyzed, the following general comparisons can be assessed and evaluated, which may help to strengthen the evidence in identifying potentially undiagnosed sleep apnea patients.

With regard to facial type, the positive patients tend to show more of a vertical growth pattern, dolichofacial, where as the negative patients, indicate more of a neutral or

horizontal growth pattern. The positive patients tended to be in more of a class II relationship, both dentally and skeletally, whereas the negative group was more class I skeletally, and less class II dentally than the positive patients. All three of the positive patients exhibit a narrow tapered arch form, whereas the negative patients arch forms varied. These arch forms are consistent with the differences in maxillary inter-molar width and maxillary inter-canine width. Although the inter-molar widths don't appear to be significantly different between the two groups, there does appear to be a consistently narrower inter-canine width for the positive group as compared to the negative patients. All of the patients in the positive group favor a more retrognathic position of the mandible with respect to their individual norms, whereas the negative patients favor a normal or slightly prognathic position with respect to their norms. The facial axis of the positive patients tends to be more open when compared to the negative patients, which are more within normal limits. The saddle angles of the positive patients are consistently more obtuse than the norms, whereas the negative patients varied, but favored a more acute relationship when compared to norms. The positive group tends to have a normal to steeper than normal mandibular plane angle, whereas the negative group tends to have a normal to flatter than normal mandibular plane angle. Lower face height and total face heights appear to be consistently greater than normal for the positive patients, whereas the negative patients face heights tend to be normal or less than normal. Nasal cavity widths for the positive group tended to be narrower than normal in comparison to the negative patients, who tend to have wider nasal cavity widths than normal. Both groups of

patients tended to have maxillary widths narrower than normal. All patients had tonsils and adenoids present.

The results of these comparisons tend to be fairly consistent with those findings as reported in the review of the literature regarding cephalometric analysis. Attention should be given to these areas when the results of the questionnaire suggests a positive diagnosis for sleep apnea, which should then result in the referral of that patient to sleep disorder specialists. With the use of the LLSDCQ and the knowledge obtained from the orthodontic patient records evaluation, identification and appropriate referral of potential sleep apnea patients in an orthodontic practice should be included in the overall diagnosis and treatment process of the orthodontic patient.

VI. SUMMARY

The outcome of this investigation has resulted in the validation of a sleep apnea questionnaire, and has shown its application to clinical orthodontics. Validation of the Loma Linda Sleep Disorders Center Questionnaire has identified statistically significant questions, and can be utilized as a simple, inexpensive, screening tool to aid in the identification and diagnosis of patients with sleep apnea. It is suggested that the questionnaire be given with the all 13 questions since that is how the questionnaire was distributed and validated in this study. To my knowledge, this is the first questionnaire of its kind to be validated. Based on the suggested incidence of potential sleep apnea patients in this orthodontic sample, attention should be given toward reducing the number of unidentified and undiagnosed orthodontic patients with sleep apnea. Further investigation should be directed toward identifying additional practical clinical indicators for sleep apnea.

It is recommended that further research be done to determine additional correlation between confirmed positive sleep apnea patients and pre-treatment orthodontic patients. Suggestions might include, performing sleep studies on those pre-treatment orthodontic patients identified as positive, via the LLSDCQ. Additionally, patients identified via the LLSDCQ could be evaluated for successful sleep apnea treatment as the result of orthodontic treatment. Questionnaires and sleep studies could be performed both pre- and post-orthodontic treatment.

APPENDIX A

LOMA LINDA UNIVERSITY CHILDREN'S HOSPITAL
SLEEP DISORDERS CENTER

PARENT QUESTIONNAIRE

Please answer the following questions about your child's sleep, when he/she DOES NOT HAVE A COLD. On a scale of 1 to 10:

1	2	3	4	5	6	7	8	9	10
	NEVER			SOMETIMES				ALWAYS	

To what extent does your child have:

(1) Difficulty breathing
1 2 3 4 5 6 7 8 9 10

*(2) Snoring or noisy breathing
1 2 3 4 5 6 7 8 9 10

*(3) Observed times with no breathing
1 2 3 4 5 6 7 8 9 10

(4) Irritability (fussy)
1 2 3 4 5 6 7 8 9 10

(5) Sleepiness during the day or excessive napping
1 2 3 4 5 6 7 8 9 10

(6) Poorer school performance than expected
1 2 3 4 5 6 7 8 9 10

(7) Restlessness, tossing, turning
1 2 3 4 5 6 7 8 9 10

*(8) Mouth breathes while awake
1 2 3 4 5 6 7 8 9 10

(9) Tonsil infections or sore throats
1 2 3 4 5 6 7 8 9 10

(10) Sweating during sleep
1 2 3 4 5 6 7 8 9 10

(11) Gasps for air during sleep
1 2 3 4 5 6 7 8 9 10

(12) Choking during sleep
1 2 3 4 5 6 7 8 9 10

(13) Turns pale or blue
1 2 3 4 5 6 7 8 9 10

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* Indicates statistically significant questions in predicting diagnosis. ($R^2 \cong 56.5$)

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