Developing an Index for the Orthodontic Treatment Need in Pediatric Patients with Obstructive Sleep Apnea: A Communication Tool between Physicians and Orthodontists

by

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Abstract

Purpose: To develop a physician screening index for the oral and facial assessment of the orthodontic treatment-need in pediatric patients with Obstructive Sleep Apnea (OSA) that may diminish the severity of their OSA symptoms.

Methods: Based on the available literature and WHO guidelines for index development, a draft index was produced, and subjected to multiple iterative revisions based on the feedback from: 1) the Index Development Group; 2) the External Review Group; and 3) the Steering Committee. Once the index was formalized, it was subjected to reliability testing

Results: Based on the feedback from the Index Development Group, the External Review Group and the Steering Committee, an index has been developed that is simple, easy to understand and easy to use. The index also exhibits a fair to substantial inter-rater reliability, and moderate to almost perfect intra-rater reliability. The orthodontic residents scored better than the medical residents in the reliability tests and took less time to use the index. The average time to use the index on 15 cases was 17:14min.

Conclusions: This is a much needed, simple, and easy to use index that is reliable among orthodontic residents and among medical residents.

Dedication

In the name of God The Beneficent, the Merciful.

This thesis is dedicated to my Mom and Dad, my brothers Mohamed, Mahdi, Mosa, sister in-law Fatima and my niece Melak.

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This work is the culmination of a lot of effort and help along the way from my friends and family. A specific thank you goes to Humam Saltaji, my brother and friend for always being there for me. Additional thanks to Genvieve Lemieux Hart, Jabir Devjee, Sajjid,& Arsheen, Natasha Nazarali and Arsalan Lavang. Thank you to my co-residents Stephanie, Ryan and Long for being so awesome. Couldn't ask for better journey men and women to embark on this journey with.

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Chapter 1: Introduction

1.1 Introducing the Problem

Pediatric obstructive sleep apnea (OSA) is one of those conditions that have been shown to be associated with psychological as well as physiologic barriers to "thrive". Among the myriad of psychological symptoms that Pediatric OSA can present with are: abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, and irritability.[1-5] Furthermore, the affected child can suffer from stunted growth, high blood pressure, and damage to the heart: including ventricular hypertrophy, and cor pulmonale.[6-9] With these types of conditions, the affected child requires much more effort just to keep up with their fellow peers in their academic and sporting settings, which puts the child at risk of being left behind.

When these symptoms are recognized the family physician or pediatrician does not have a tool to take into account the way the craniofacial phenotype could be affecting the pediatric OSA symptoms. This tool will allow for better communication between the physicians and the orthodontists.

Moss was the first to postulate that form follows function in 1968. His research focused on the effect soft tissue had on the developing hard tissue, and that the soft tissue was dictated by functional needs.[10] Similarly, Harvold demonstrated in 1981 that blocking the noses of rhesus monkeys would bring about an adaptive malocclusion due to the changed functional demands.[11]

Although research on monkeys doesn't always translate onto humans, it does provide valuable insight into a cause-and-effect relation between the breathing pattern and the craniofacial phenotype. Also, since then much research has confirmed the association between the craniofacial phenotype and pediatric OSA;[12-14] and, much research has shown the beneficial effects of correcting the craniofacial phenotype on pediatric OSA symptoms. It is now recognized that orthodontic intervention on these patients with pediatric OSA symptoms is an effective, viable and important treatment option to offer.[15-27]

1.2 Statement of the Problem

Since orthodontic treatment of the pediatric OSA craniofacial phenotype may be an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, etc.) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dentally trained practitioners to help identify which children with OSA would benefit from orthodontic treatment. Therefore this study aims to develop a screening index for physicians that can summarize the need for orthodontic treatment in children with pediatric OSA that may benefit from relief of their OSA symptoms.

1.3 Significance

This index development accounts for the multi-factorial nature of OSA and the need for multi-disciplinary care. The purpose of this index is to provide a method to assess craniofacial features of children with OSA, as diagnosed by a

physician, to identify those who may benefit from orthodontic intervention. Therefore this index will help physicians and other medical professionals understand and identify which pediatric OSA patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly. This in turn will allow for better communication between the physicians and the orthodontists. It will provide a systematic method to assess craniofacial form for long-term follow up and to compare between centers and across different populations of children with obstructive sleep apnea.

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Chapter 2: Literature Review

2.1 Introduction

Obstructive sleep apnea (OSA) is a complex and multifactorial disorder that requires a multidisciplinary approach for treatment. Therefore one of the keys to being able to plan this multidisciplinary treatment appropriately is efficient communication between the members of the team. In order to ameliorate this communication, this project aims to develop a simple and easy to use index to assess the orthodontic treatment need in pediatric patients with obstructive sleep apnea in order to help non-dentally trained team members identify those malocclusions that may benefit from orthodontic treatment. The index will be a visual representation of the characteristics that an orthodontist would assess in the treatment of the patient, and will assign a score to the patient's craniofacial morphology in order to prioritize and streamline those patients whom are most likely to benefit from orthodontic treatment. In this literature review the craniofacial morphology of children with OSA and the current orthodontic treatment modalities that may help with the symptoms/manifestations of OSA in children are summarized; as well as, the evolution of indices, the nature of them and how they were developed.

2.2 Craniofacial Morphology

A comprehensive search of the literature was conducted on September 25th, 2013 using PubMed, Medline, Embase, Scopus, web of Science and grey literature. The search identified two recently conducted systematic reviews describing craniofacial morphology of pediatric OSA patients. The first review by

Katyal et al[1] had a search that was performed on April 2012, and included non-syndromic children 0-18 years old with sleep disordered breathing or sleep apnea diagnosed by a sleep disorders unit, screening questionnaire, or polysomnography. What they found was that children with OSA had increased weighted mean differences in ANB 1.64 deg (P<0.0001) (Fig 2.1) compared with the controls. Also, children with obstructive sleep apnea had a reduced posterior nasal spine to adenoid tissue space by 4.17mm (weighted mean difference) (P<0.00001) measured along the PNS-basion line (Fig 2.1), compared to controls. Children with obstructive sleep apnea had a reduced posterior nasal spine to adenoid tissue space by 3.12mm (weighted mean difference) (P<0.0001) measured along a line perpendicular to the sella basion line, compared to controls. Finally, SN-MP angle was increased by 2.74 deg (weighted mean difference, P=0.006) indicating a larger lower face height, although there was a lot of heterogeneity in this variable among patients with OSA (Fig 2.1).

The second review by Flores-Mir et al[2] was also performed on April 2012 and included non-syndromic children 0-18 years old, with no previous orthodontic or orthognathic treatment, OSA diagnosed with polysomnography, and lateral cephalometry to assess the craniofacial morphology. What the authors found was that children with OSA had an MP-SN angle 4.20 degrees greater in the OSA group than the control group (P<0.001). Also, SNB was 1.79 degrees less in the OSA group than in the control group (P<0.001), and ANB was 1.38 degrees greater in the OSA group than in the control group (P<0.001) (Fig 2.1).

Both reviews agree that: the ANB is increased, implying that there's an increased discrepancy between the mandible and the maxilla; the SNB is decreased, implying that the mandible is more retrognathic relative to the cranial base; that the MP-SN angle is increased, implying that there is an increased tendency for a divergent pattern of growth comparing the mandible with the cranial base and a longer face height.

These parameters have been shown to be statistically significant, but whether they are clinically significant is still up for debate. Many people would consider 1.64 degrees in ANB to be not of much clinical significance. Furthermore, ANB doesn't distinguish between a discrepancy in jaw sizes and jaw positions, as well as being affected by the position of the maxillary and mandibular incisors and the cranial base. Therefore ANB isn't always a reliable measure of the sagittal relationship of the jaws.[3]

2.3 Rapid Maxillary Expander (RME)

The only systematic review, which we are aware of, with regards to RME in the treatment of OSA has recently been accepted for publication. The review by Major et al. was performed on November 2012 and included studies with non-syndromic subjects 3-16 years old, with results of maxillary expansion treatment that can be distinguished from other treatment modalities, and an initial Apnea/Hypopnea Index (AHI) score of 1-5. The AHI is a score of OSA severity and is measured through a sleep study by measuring the average number of apnic and hypopnic events per hour throughout the patient's sleep. These events are

usually also associated with a diminishment of blood oxygen levels. Based on their results of 8 studies with evidence levels in the fair to poor range; they found that the RME procedure does improve the AHI scores mildly to moderately, in patients with OSA and a constricted maxilla, although not fully resolving them. The authors also suggest that RME combined with adenoidectomy and tonsillectomy (A&T) does resolve the symptoms of the select patients reducing the failure rate (AHI<1) to 3.8-6.7% compared to the 50-75% seen with A&T alone. They concluded that studies of better quality (minimizing bias) are needed and long term studies, and indicated that this treatment modality showed some promise in select cases of mild pediatric OSA.

2.4 Mandibular Advancing Appliance (MAA)

A Cochrane review by Carvalho et al. was found reviewing the literature on MAAs.[4] Their search was performed on February 2007. They evaluated studies with children 0-15 years old, that had adequate control groups, and whom had more than 1 apneic episode per hour measured by polysomnography. Since this Cochrane review is dated, we performed a more up-to date search of the primary literature in this area, through PubMed, Medline, Web of Science and Scopus on September 25th, 2013. The Cochrane Review found only one article[5] that fulfilled its inclusion criteria, while the more recent search revealed three more.[6-8] Villa et al. followed a group of 32 7-year-old children with OSA, given a MAA for 6 months and observed a mean reduction in AHI from 7.1+/- 4.6 events/hr to 2.6+/- 2.2 events/hr, whereas the control group did not show any reduction. A second study by Schutz et al[6] examined sixteen 12-year-olds with OSA treated with a

MAA and a maxillary expander for 12 months, and found that the number of respiratory-effort related arousals decreased from a mean of 7.06+/- 5.37 events/hr to 1.31+/- 1.45 events/hr of sleep. The Cozza et al[7] study examined 4-8 year olds for six months of mainly night-time wear of a modified monobloc; the authors reported a diminishment of the median AHI score from 7.88 to 3.66, along with a subjective improvement in sleep quality and diminishment of daytime sleepiness. Finally, the Zhang et al[8] study took 8-10 year olds and delivered a twin block treatment fulltime for an average of 10.8 months. The results showed an average diminishment of the mean AHI score from 14.08 +\- 4.25 events/hr to 3.39 +\- 1.86 events/hr.

Therefore the studies demonstrate that there is a beneficial effect of using MAAs, particularly in a mandibular retrognathic patient leading some researchers to conclude that MAA is an effective therapy for symptom diminishment, particularly for children with OSA and craniofacial anomalies.[9] There are limitations in drawing conclusions from all these studies because not all of them had control groups (due to ethical concerns), they were all different ages, used different measures of OSA severity, and used different lengths of follow up. Also MAA use may be contra-indicated in children with harmonious craniofacial morphologies due to the dentofacial effects it would have on a growing child; the effects of these types of appliances decrease the patient's overjet.

The Villa et al[5] study's strengths are in its randomized allocation of patients, using an untreated control and in being able to match the patients' weight and height. Also, it is a prospective study. The weaknesses of the study are that it had

a small sample size of 19 and 13 between treated and control respectively, with 5 and 4 lost to follow-up. Also, rather than evaluating a normalized AHI, the criteria for success was deemed to be a 50% decrease in the AHI, which seems like a low threshold for success. Finally it is not clear if these patients were followed long term; therefore we don't know if these results are sustainable.

The Schutz et al[6] and Cozza et al[7] studies' strengths are that they are prospective studies, but also have small sample sizes of 16 and 20, respectively. Moreover, a control group was not included in either study due to ethical concerns over withholding treatment. Therefore there could not be any randomized allocation of treatment. The Zhang et al[8] study employed a larger sample size of 46 children, but they also could not select a control group for ethical reasons.

2.5 Reverse-pull headgear with RME

Although the RME component of the treatment has been shown to help with the symptoms of OSA, there are currently 5 studies that look at the effects of the reverse pull headgear, with all of them having weak designs.[10-14] Most notably, none of the studies used polysomnography in assessing the OSA subjects. That being said, the studies show great promise in that subjective markers reveal a benefit on OSA symptoms with this treatment, and that the dimensions of the upper airway increases in size. There is a lot of anecdotal evidence with regards to this treatment modality, yet until better studies are performed no conclusions can be ascertained.

2.6 Surgical Maxillary Mandibular Advancement (MMA)

Patients in the older pediatric population may also benefit from surgical treatment for their OSA as their craniofacial growth is nearing relatively completion.[3] Generally the MMA treatment is reserved for patients with severe craniofacial anomalies and/or patients with severe OSA. Bear and Preist have shown that surgical mandibular advancement can bring about a reversal of the sleep symptoms and return the EEG readings ack to normal, but on only one patient.[15] Despite it being a case study, much more research in the adult population suggests that surgery may be an option in sever OSA cases. According to Holty et al. after performing a met-analysis on all the studies assessing surgical MMA outcomes, they found the mean AHI decreased from 63.9 events/hr to 9.5 events/hr following surgery, and the surgical success rate was 86% with a cure rate (AHI<5 events/hr) of 43.2%. Therefore they concluded that MMA was a safe and effective treatment for severe OSA. Moreover, the patient pool included 22 unique populations totaling 627 patients with OSA and 320 other OSA subjects. The population of these studies involved a mixture of ages from 35-53.8 years old and since the conclusions are based on an adult population, more studies on the pediatric population are needed.[16]

2.7 Summary

- OSA patients are at risk for an increased ANB, decreased SNB, and an increased MP-SN angle
- MAAs may be beneficial for patients with mild cases of OSA and a concurrent mandibular retrognathia.

- RME combined with tonsillectomy may be beneficial for patients with mild to moderate cases of OSA and a concurrent maxillary constriction.

2.8 Indices

In the orthodontic context an index is used to as a rating or categorizing system that assigns a numeric score or alphanumeric label to a person's occlusion or facial type.

2.8.1 Craniofacial Morphology Assessments

Craniofacial Morphology is best assessed using radiographic imaging, and historically the gold standard were lateral cephalograms, although more recently Cone Beam Computed Tomography (CBCT) imaging is considered better in assessing the craniofacial morphology in three dimensions.[3]

In the quest for establishing norms for occlusion and craniofacial morphology there was a need to establish the criteria for what the ideal is. Shortly after World War II, Downs came up with an analysis to study lateral cephalograms; he based his analysis on 25 untreated Caucasian adolescent males.[3] Despite it being a good initial effort, there were gaping flaws in that it deemed the Caucasian male face to be ideal and left little room for the other gender and other ethnicities to be treated according to their sex-specific and ethno-racial standards; also if we use only the ideal craniofacial morphologies as "normal" we are effectively skewing the interpretation of what is normal to one side of the spectrum of malocclusions. The epitome of this concept was when Steiner subsequently used one Hollywood starlet, whom he thought displayed exceptional

qualities, to establish all of his norms.[17] To his credit his norms have been now shown to be a decent predictor of normal craniofacial phenotype. But since no one person may possess all the attributes that are necessary for the perfect craniofacial form and occlusion, contemporary analyses of the craniofacial form are incorporating mild malocclusions and facial discrepancies into the calculation of the norms in order to shift the norms back to being more relevant to the entire population. Moreover, different sex-specific, and ethnic-specific norms are now available to help make the treatment of different sexes and ethnicities more appropriate.[17] Despite advances there is still no consensus as to the single analysis that is most useful for assessing craniofacial morphology.

2.8.2 Occlusal Assessments

The first index to be developed for normal and malocclusion was in 1899 by Edward Angle.[3] He proposed that the first molars were the anchor teeth, and that all the occlusion could be defined based on the relation of the upper first molar with that of the corresponding lower first molar.[17] Subsequently, Andrews refined this index in 1972 and added his own six keys of occlusion that defined the ideal occlusion.[18] These six keys included all the teeth, and their position with regards to rotation (or lack thereof), no spacing, tip and torque of the teeth. Then in 1981 Roth added some functional parameters, such as centric relation, centric occlusion, lateral rise and incisor guidance as qualities that an ideal occlusion should possess.[19] These characteristics define our understanding of an ideal occlusion, with normality being a slight deviation from the ideal.

Once normal was established there was a need to further classify the abnormal malocclusions into different severities favoring parameters deemed to be more disfiguring. Each of the subsequent developed indices had a slightly different reason for existence and each served a different purpose. For epidemiological data collection in non-syndromic patients Bjork,[20] Baume[21] and Bezroukov[22] each developed an index in 1964, 1974 and 1979 respectively. These indices provided researchers the ability to document malocclusions for research purposes. For clinical purposes, occlusal classifications of malocclusion started with deviations of angle's ideal molar occlusion,[18] then Dewey modified that classification in order to further stratify the Class 1 component of the malocclusion to five types.[23] The British Standard Institute came up with their own version of classification of malocclusions called the incisor classification in 1983 that was further modified in 1992.[24] This form of classification was rooted in the position of the lower incisor relative to the upper incisor, and similar to the other indices in this class were meant to evaluate deviations from normal occlusion in a clinical setting, facilitating communication between clinicians.

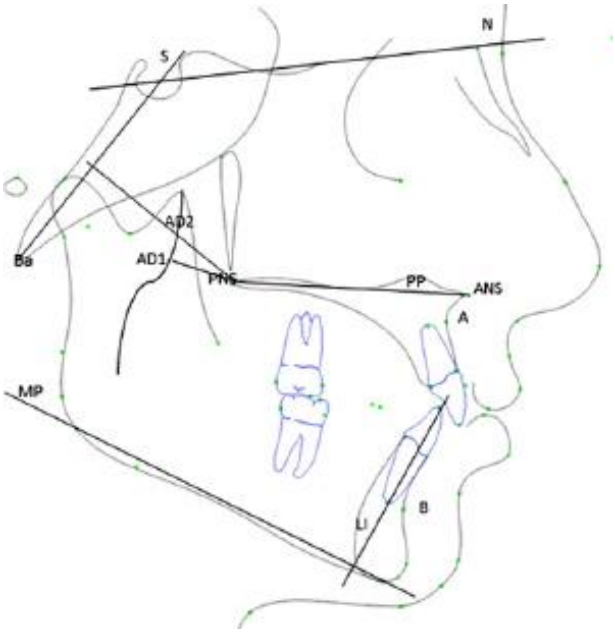
Priority treatment-need was assessed as part of the Handicapping Labiolingual Deviation Index,[25] Occlusal Index,[26] Index of Orthodontic Treatment Need: Dental Health Component, and the Index of Orthodontic Treatment Need: Aesthetic Component.[27] These indices developed between 1960 and 1989 due to a demand by government funding agencies to establish a tool for funding select malocclusions deemed particularly detrimental to function and esthetics. Then the

PAR Index was developed to compare pre and post treatment results, and orthodontic outcomes.[28]

Indices assessing the future need for surgery in cleft lip and palate patients have also been developed. The GOSLON Yardstick was first developed to assess cleft patients in the early mixed dentition,[29] and later modified to the Five-Year-Old Index which can be used in the primary dentition.[30] Also, the Modified Huddart Bodenham can be used reliably on all ages[31] and should be considered the gold standard with regards to the cleft lip and palate occlusal indices.[32]

Most of the initial indices developed have relied on individual expertise or that of a small group of 2-4 individuals; while more contemporary indices that have been developed have relied on the previous indices and modified what they perceived as weaknesses.[33] Of all the developed indices, the Index of Complexity, Outcome and Need (ICON), is the one that stands out as having the most complete and extensive development methodology; it also is an index that is not considered a modification of a previous index. The aim of the ICON is to assess treatment-need and outcomes using one index, and this was first achieved by subjecting a group of orthodontists from many different countries to assess pictures of patients and give a dichotomous yes or no answer as to whether treatment is warranted or not and whether the outcome was acceptable or not, as well as a subjective judgment of the pre-treatment complexity and post-treatment improvement on a scale of 1-5. Using a Stepwise Multiple Logistic Regression the highly predictive variables for pre-treatment complexity and post-treatment results were chosen and used to predict the panelists' dichotomous responses. The

highly predictive variables were: left and right buccal antero-posterior relations, IOTN aesthetic component, upper arch crowding, overbite/open bite, and presence of a crossbite. Weights were then given to each of the predictive variables, and a cut-off value was chosen to maximize the sensitivity and specificity of the index responses.[34] Given the sound methodology of the ICON's development, portions of its methodology have been adapted to the development of our current index.



(Source: Katyal et al.2013[1])

Figure 2.1. A point represents the anterior part of the maxilla. **B** point represents the anterior part of the mandible. **N** (nasion) point represents the junction of the frontal bone with the nasal bone. **S** (sella) point represents the hypophyseal fossa where the pituitary gland is located. **PNS** (posterior nasal spine) point represents the most posterior part of the maxilla. **MP** (mandibular plane) line represents the inferior border of the mandible. **ANB** angle represents the antero-posterior relationship of the maxilla with the mandible relative to the bridge of the nose. **SNB** angle represents the antero-posterior relationship of the mandible relative to the cranial base. **MP-SN** angle represents the vertical direction of the mandible relative to the cranial base.

2.10 Summary

Indices have been used for a long time and help to quantify a malocclusion or a craniofacial phenotype. Despite being around so long, there is no consensus as to how these indices should be developed.

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Chapter 3: Developing an index for the orthodontic treatment need in paediatric patients with obstructive sleep apnoea: a protocol for a novel communication tool between physicians and orthodontists.

Published Article:

Altalibi M, Saltaji H, Roduta Roberts M, Major MP, MacLean J, Major PW. *BMJ Open*. 2014 Sep 18;4(9):e005680

3.1 Abstract

Introduction: Sleep disordered breathing in the pediatric population can manifest as an array of different systemic symptoms; among them is a distinct malocclusion and craniofacial phenotype. Emerging research suggests that the treatment of this malocclusion and/or craniofacial phenotype through orthodontic intervention may help with the symptoms of these patients. Selecting the patients that would benefit from orthodontic treatment can be a difficult task for the physician with minimal dental training. Therefore the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study. Based on the available literature, a draft index will be produced, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group, a group of multidisciplinary and internationally acclaimed experts in the field; the

External Review Group, a group of potential end-users and interested parties; and the Steering Committee. Once the index has been formalized, it will be subjected to a pair of reliability tests using physicians and orthodontists scored 2-weeks apart. Subsequently the index will be validated using dichotomous responses from orthodontists on whether they would treat a patient for OSA symptoms, and comparing the responses to the score of the index on the same patient.

Ethics and dissemination: The index will be translated into French, and will be presented in orthodontic and medical conferences, workshops, seminars, round table discussions and free copies for download will be made available on the website of the University of Alberta Interdisciplinary Airway Research Clinic (iarc.ualberta.ca). Furthermore, the index will be published in a peer-reviewed medical journal to further increase the exposure of the index.

Article Summary

Article focus

- To develop a simple index for medical professionals to identify children and adolescents with obstructive sleep apnea who may experience functional airway benefit from orthodontic treatment.

Strength and limitations of this study

- This index will help physicians and other medical professionals understand and identify which obstructive sleep apnea patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly.
- This index development acknowledges the multi-factorial nature of SDB and the need for multi-disciplinary care. The ideal end result of this index is to facilitate and enhance effective collaboration between invested dental and medical specialties.
- Development of a validated index will facilitate future epidemiology studies, allow for quality assurance, and guide funding allocation. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

3.2 Introduction

Snoring, although ubiquitous in the adult population, is considered abnormal in children and adolescents.[1] More importantly, it may serve as an indicator of a more severe respiratory problem that presents as a continuum, from primary snoring to obstructive sleep apnea (OSA). Reports vary on the prevalence of obstructive sleep apnea ranging from 0.7% to 5% of the population under 18 years old.[2-8] Moreover, breathing induced sleep disorders have been proven to have a profound effect on the child's behavior, growth and development; the myriad of symptoms include: morning tension-type headaches, excessive morning thirst, excessive fatigue and sleepiness, abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, irritability, among many others.[9, 10 11-13] Other physiologic processes that can be affected include stunted growth,[14] high blood pressure,[15] damage to the heart: ventricular hypertrophy,[16] and cor pulminale[17].

In addition to behavioral and systemic health consequences, craniofacial development is also affected. These patients generally have a craniofacial component contributing to their OSA, which would manifest as a retrognathic maxilla[18] or mandible, a long lower face height and restriction in the space of the upper airway[19, 20]. Furthermore, when evaluating the polysomnogrophy of these patients, the evidence suggests that palatal expansion, and mandibular advancement appliances[21-26] can be of benefit at reducing the severity of OSA. Reverse pull headgear[27-32], and maxillary & mandibular advancement surgery[33]

have also been shown to have great promise at helping this group of patients. Since orthodontic treatment of the OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, etc.) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with SDB would benefit from orthodontic treatment.

Therefore this study aims to develop an index that can summarize the need for orthodontic treatment, in select cases of children with OSA, to physicians, and adjunct medical professionals. Once the index is developed, it will be assessed for reliability and will be validated. Upon completion, this index will equip medical professionals with a simple way to assess which patients have a malocclusion that contribute to their OSA and may benefit from orthodontic treatment.

3.3 Methods and Analysis

3.3.1 Initial Development

In accordance with the World Health Organization's recommendations on developing an index,[34] development of the index will be achieved through the following objectives:

1- Establishing a Steering Committee

2- Scoping the index

3- Reviewing the literature

4- Drafting the index

5- Organizing an Index Development Group

6- Organizing an External Review Group

3.3.1.1 Steering Committee

The Steering Committee will be established apriori and is responsible for overseeing every aspect of the study. It will be composed of a representative group of 3 experts in orthodontics, pediatric sleep medicine, and a methodologist specializing in psychometric property analysis. Their responsibilities include: scoping the index, overseeing evidence retrieval, drafting the index, selecting members of the index development group and external review group, finalizing the index.

3.3.1.2 Scoping the Index

Scoping is the process of defining what factors will be investigated in the literature for inclusion in the index. Scoping will be achieved through the combined experience and expertise of the Steering Committee, and each factor that is suggested will be further investigated in the literature to establish an evidence-based approach to the development of the index.

3.3.1.3 Reviewing the Literature

For each of the scoped factors the literature will be reviewed to establish relevance. Specifically, the evidence must demonstrate that appropriate treatment of the craniofacial factor in question will lead to an improvement in the OSA symptoms. Priority will be given to the results of well-conducted and well-reported systematic reviews and randomized control trials. Each of the factors will be assessed on its effect on pediatric OSA. Furthermore, the literature will be

searched for the craniofacial morphology of pediatric OSA patients. All of this information will be brought back to the Steering Committee for discussion of inclusion / elimination of factors in the index.

3.3.1.4 Drafting the Index

Since this index will have visual-rating scales for each of the craniofacial and occlusal factors, the steering committee will devise an outline suitable for displaying each of the factors representing the index. The factors in the drafted index will also have a number of levels that will divide the factor into categories of severity. The number of levels for a given factor will also be determined by the Steering Committee. Details describing the nature of the illustrations, the amount of levels that each of the factors will have, and the general layout of the index will be agreed upon by the steering committee, and the graphic artist will then design a preliminary version of the index based on the relevant factors and the feedback from the steering committee.

3.3.1.5 The Index Development Meeting

The Index Development Group (IDG) is formed by a group of external multidisciplinary experts who provide evidence-based recommendations, on the content, layout and development process of the index. This eclectic group should be small enough to be able to have effective discussions, while large enough to ensure the appropriate representation from all the stakeholders. In this study we plan on including a broad spectrum of medical specialists, family physicians, orthodontists, and methodologists. The goals of this meeting are to gain additional

information to strengthen the index, as well as gaining the professional credibility of a broad range of experts on the topic of pediatric OSA.

Once the members have been chosen by the Steering Committee and accept the invitation to participate, the IDG members will be emailed a document 4 days prior to the meeting, explaining: the purpose of the index and meeting, how the meeting will proceed, what to expect in the meeting, and a brief literature review of the orthodontic techniques currently available to help with the symptoms of OSA. Moreover, every member will be randomly assigned a number in order to maintain anonymity of the responses.

Procedure of the IDG Meeting

The meeting will commence with a brief introduction summarizing the literature review, the purpose of the index and its relevance. The meeting will then proceed to collect feedback (APPENDIX A) on the aforementioned chosen factors using a modified Delphi technique[35, 36] , which is a communication technique that structures the meeting and minimizes bias in responses. Participants' responses will be collected through a web-based response portal. Each factor will be explored through yes or no questions, and yes responses followed-up with a scale of 1-9 based on its importance for decision-making. Once all the feedback is received on a particular factor, the summary of the results will be displayed for everyone to see. Discussion will ensue for a maximum of 5 minutes, with each person talking no more than 1 minute. Then everyone will be asked to re-enter their feedback on the website for the same factor in light of the discussion. This cycle will continue until a consensus of greater than 80% of the

members is reached. Once consensus is reached, then we will move on to the next factor and the participants will be asked to give feedback on this new factor in a similar manner.

In addition, two negative control factors will be used to calibrate the responses. The first negative control will be of orthodontic relevance but have no effect on the amelioration of the symptoms of OSA (ie. crowding, or impacted canine). The second negative control will be of relevance to OSA symptoms, but cannot be changed by orthodontic treatment (ie. BMI or neck circumference).

Once all the factors have been discussed and consensus reached, we will get feedback through the website on the: alignment - importance of the index, and if the development process is appropriate; relevance – content analysis, and whether all the factors identified are important; representation – if there is anything that needs to be added to the index (APPENDIX B). Finally, additional written feedback will be accepted at the very end of the session.

All the feedback from the IDM will be summarized and presented to the Steering Committee, and decisions will be made to remove factors, modify factors, and /or modify the outline and layout of the index. These modifications will subsequently be presented to the graphic artist and a second draft will be procured.

3.3.1.6 The External Review Group

The External Review Group (ERG) is composed of end-users and interested parties. This group is not responsible for any content analysis, instead it

will be responsible for reviewing the layout, simplicity and ease of use. It will also be responsible to assess usefulness of the index in the healthcare setting and give feedback on the feasibility of implementing the index in practice. The goal of this meeting is to gain the end-user approval for the ease and feasibility of administering the index. This group should be large enough to be a representative sample of the population, yet small enough to allow for ease of explanation and healthy discussion. It is not as structured as the IDM, and allows for the participants to freely express their opinions in an open forum.

In this meeting we will explain the theory behind the index by briefly reviewing the literature and then explain the purpose of the index. Subsequently we will show the group a pilot version of the index and a paper will be distributed to receive for their feedback based on the following questions:

1- Do you understand the purpose of the index?
2- Do you understand what each factor is assessing?
    a. If not which one(s) do you not understand and why?
3- On a scale of 1 to 10, how simple would you rate this index to understand and use?
4- Would you use this index in your practice?
5- Other recommendations:

All the feedback from the ERG will be summarized and presented to the Steering Committee, and decisions will be made to modify the outline and layout of the index. These modifications will subsequently be presented to the graphic artist and a third draft will be procured.

3.3.2 Reliability

Reliability of the index will be tested within a group that represents the typical end-user population. This includes physicians of family medicine, pediatricians, pediatric ENT, or pediatric sleep physicians. Reliability will also be assessed within a group of orthodontists, who by their training are experts at assessing malocclusion and craniofacial morphology. Therefore 10-20 physicians and orthodontists will be recruited as examiners in this study.

A pool of 15-40 randomly selected patient charts from the University of Alberta Inter-disciplinary Airway Research Clinic (I-ARC) will be recruited as reliability test subjects, and their intra-oral and extra-oral photographs will be used in the reliability assessment. After a brief explanation about the use and application of the index, the physicians as well as the orthodontists will apply the developed index to the sample patient pool's pictures once. In order to diminish recall bias, application of the index will be repeated 2 weeks later.

Intra-rater reliability and inter-rater reliability between the physicians, between the orthodontists and between the physicians and orthodontists will be compared. The reliability will be assessed using Interclass Correlation coefficients (ICC) and Bland –Altman Plots.

3.3.3 Validation

The Index will be validated using dichotomous responses from orthodontists on whether specific patients would require orthodontic treatment to help their obstructive sleep apnea symptoms and comparing it to the score that the

index gave those same patients. This will be achieved by setting up a website where 30 orthodontists with experience in dealing with pediatric OSA will be recruited to take the assessment. The website will contain extra-oral and intra-oral pictures of patients randomly selected from the Interdisciplinary Airway Research Clinic diagnosed with OSA, and the orthodontists will be asked to rate these patients, using a "yes" or "no" response, whether they would benefit from orthodontic treatment for their OSA symptoms. The index will then be applied on the same patients by the principal investigator and the score of each patient will be recorded. Using a stepwise multiple-logistic regression each of the identified factors will be given a weight; this will represent the relative importance of the factor. Once analyzed if the correlation is high between the expert scores and the cluster groups, then the clusters are meaningful and valid. Furthermore, a cutoff for most efficient score above which to refer will be chosen using a graph and observing the value that optimizes the sensitivity, specificity and overall accuracy of the index. Finally, four grades of treatment-need will be determined using the twenty-five percentile ranges. The grades will be:

1- Minimal Need
2- Mild Need
3- Moderate Need
4- Severe Need

3.4 Discussion

The development plan of this index has been conceived through a modification of the WHO Handbook for guideline development,[34] as well as

reviewing the orthodontic literature for ways indices have been previously developed. The WHO provided an excellent starting point, from there modifications were made to cater to the specifics of this study, given that there are differences between developing a guideline and an index for orthodontic treatment need. The literature was useful, and among the index development protocols reviewed, certain assessed the orthodontic treatment need within the entire population,[37-40] while others assessed it for a given subpopulation[41-46]; each had strengths and weaknesses, and thus we further modified our methods, synthesizing a protocol for our particular needs from the available literature and using the experience and expertise of the authors. Through this protocol we aim to develop an index that fulfills all of the following criteria:[47]

1. Gradient of Numeric Values: The severity of the orthodontic treatment need within the pediatric OSA patients should be defined within a numerical scheme that demonstrates a finite and progressive gradient from low need to high need.
2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
3. Clinical Importance: The numerical scale should correspond with the clinically appraised orthodontic treatment need of pediatric OSA patients.
4. Statistical Ease: should be amendable to statistical analysis.
5. Reliability: Should have a high intra- and inter-rater reliability.
6. Practical: The instruments required to score the index should be

practical to the setting in which it will be administered.

7. Minimal Judgment: Applying the index should require minimal judgment.
8. Simple: The index shouldn't have a high financial or time cost, therefore should be simple enough to administer to many patients.
9. Detect Change: The index should be able to detect changes in orthodontic treatment need in pediatric OSA patients.
10. Validity: should be valid over time.

Validity can be characterized into different types: Face Validity, Content Validity, Construct Validity, and Criterion Validity.[48] In this study, we will examine these kinds of validity at different stages of development. The first draft of the index will focus on establishing face validity. Feedback from the steering committee and IDG will assist in establishing content and construct validity. Assuming that the "gold standard" in assessing the orthodontic treatment-need in pediatric patients with OSA is an orthodontist with experience in dealing with pediatric OSA patients, then the subsequent modification of the index based on the reliability tests and the dichotomous responses from the orthodontists provide the index with the necessary criterion-related validity evidence through statistical means.[49]

3.5 Significance

This index will help physicians and other medical professionals identify which craniofacial phenotypes may benefit from orthodontic treatment as part of their multi-disciplinary OSA management. Furthermore, due to the diverse

medical effects of sleep deprivation, there will be a trend to make sleep apnea into a centralized service, where the main focus is for a highly trained multidisciplinary team to treat a high volume of patients to a standardized protocol, where meticulous documentation is exercised. This index is part of that documentation process. It will allow for quality assurance, funding allocation and epidemiologic studies to be performed. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

3.6 Dissemination Plan

The dissemination of this index will be done through a variety of ways in order to maximize its reach. Primarily, it will be published in a peer-reviewed journal, which will allow its introduction to the scientific literature. The journal should be a respected medical journal with broad reach, in order to allow the greatest number of physicians to be exposed to the index. Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian and American population of medical professionals. Moreover, the index will be presented at national and international conferences to increase the awareness of the index among the scientific community. Finally, the index will be used at the University of Alberta's Interdisciplinary Airway Research Center, and more research, so that future research in this center will incorporate it. It will also be placed on the University of Alberta's Interdisciplinary Airway Research Center's website under the physician section, to further educate the doctors who visit the site on the index.

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Chapter 4: The Development of the Alberta Pediatric Obstructive Sleep Apnea (APOSA) Index: a Novel Communication Tool between Physicians and Orthodontists

4.1 Abstract

Introduction: Many aetiologies have been associated with pediatric obstructive sleep apnea (OSA). In patients with a craniofacial phenotype contributing to their symptoms, orthodontic intervention may be warranted, yet there is no guideline as to which patients would be candidates for orthodontic treatment. Therefore, the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study and the recommendations of the steering committee. Based on the available literature, a draft index was developed, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group (IDG), a group of multidisciplinary and internationally acclaimed experts in the field; the External Review Group (ERG), a group of potential end-users and interested parties. Once the index was formalized, it was subjected to a pair of reliability tests using medical residents and orthodontic residents scored 1-2 weeks apart.

Results: The finalized index showed fair to substantial inter-rater reliability, and moderate to almost perfect intra-rater reliability. The average time to use the index on 15 cases of patient photos was 17:14min.

Conclusion: Selecting the candidates for orthodontic treatment has been simplified by developing the Alberta Pediatric Obstructive Sleep Apnea (APOSA) Index.

4.2 Introduction

Pediatric Obstructive Sleep Apnea (OSA) is a disease requiring a multidisciplinary treatment approach. Within the growing evidence of treatment modalities for OSA are orthodontic treatments that focus on changing the craniofacial morphology of the young patient. Craniofacial features of pediatric OSA include: a retrognathic maxilla[1] or mandible, a long lower face height and restriction in the space of the upper airway.[2, 3] Furthermore, orthodontic interventions aimed at correcting these craniofacial phenotypes, such as palatal expansion, and mandibular advancement appliances[4-9] have been shown to be of benefit to reduce the severity of the pediatric OSA especially in the mild to moderate cases. Reverse pull headgear[10-15], and maxillary & mandibular advancement surgery[16] also show some promise as being effective treatment modalities for pediatric OSA, although more research is needed.

Since orthodontic treatment of the pediatric OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, etc.) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with pediatric OSA would benefit from orthodontic treatment. Therefore this study aims to develop an screening index that can summarize the need for orthodontic treatment to physicians in select cases of children aged 7-18 years old with OSA. Upon completion, this index will equip medical professionals with a simple way to

assess which patients have a craniofacial phenotype associated with their pediatric OSA amenable to orthodontic treatment that can help ameliorate their symptoms.

4.3 Methods

4.3.1 Initial Development

In accordance with the World Health Organization's recommendations on developing an index,[17] development of the Alberta Pediatric Obstructive Sleep Apnea (APOSA) Index was achieved through the following objectives:

1- *Establishing a Steering Committee*

2- *Scoping the index*

3- *Reviewing the literature*

4- *Drafting the index*

5- *Organizing an Index Development Group*

6- *Organizing an External Review Group*

4.3.2 Steering Committee

The Steering Committee was established on March 11th, 2013 and was responsible for overseeing every aspect of this study. It was composed of a representative group of 3 experts and the principal investigator. The 3 experts comprised: an orthodontics professor with special interest and expertise in sleep related issues, a professor in pediatric sleep medicine, and a professor specializing in methodologies related to measurement evaluation and analysis of psychometric properties. Their responsibilities included: scoping the index, overseeing evidence

retrieval, drafting the index, selecting members of the index development group and external review group, and finalizing the index. The steering committee organized 5 formal meetings as a collective group and 21 formal meetings between individual members of the committee. With the newly established Steering Committee, the index was scoped for pertinent factors on August 21st, 2013 and a literature review was conducted on September 25th, 2013; based on these results and the aid of a graphic artist, the index was drafted for use at the Index Development Meeting on November 13th, 2013.

4.3.3 The Index Development Group Meeting

The Index Development Group (IDG) was formed by a group of external multidisciplinary experts who provide expert opinion-based recommendations on the content, layout and development process of the index. This group was small enough to be able to have effective discussions, while large enough to ensure that the appropriate representation from all the stakeholders was present. In this study we included: 5 orthodontists, 2 pediatric otolaryngologist, 2 pediatric respirologists, 1 pediatric psychiatry and sleep medicine physician, 1 pediatric otolaryngology and facial plastic and reconstructive surgeon, 1 nurse practitioner and 1 methodologist.

The IDG members were emailed a document 4 days prior to the meeting explaining: the purpose of the index and meeting, the proceedings, what to expect, and a brief literature review of the orthodontic techniques currently available to help with the symptoms of OSA. Every member was randomly assigned a number in order to maintain anonymity of his or her responses.

The meeting commenced with a brief introduction summarizing the literature review, the purpose of the index and its relevance. The meeting then proceeded to collect feedback (APPENDIX A) on the aforementioned chosen factors using a modified Delphi technique,[18, 19] to structure the meeting and minimizes bias in responses. Due to time constraints some of the factors had to be grouped together in the discussions. Responses were collected through a web-based response portal. Each factor was explored through yes or no questions, and yes responses were followed-up with a scale of 1-9 based on its importance for decision-making.

Two negative control factors were used to calibrate the responses. The first negative control, crowding, was of orthodontic relevance but has no effect on the amelioration of the symptoms of OSA. The second negative control, neck circumference, was of relevance to OSA symptoms, but cannot be changed by orthodontic treatment.

Also, feedback was retrieved on the: 1) alignment - importance of the index, and if the development process is appropriate; 2) relevance – content analysis, and whether all the factors identified are important; and 3) representation – if there is anything that needs to be added to the index (APPENDIX B). Finally, additional unstructured written feedback was accepted at the very end of the session.

Based on the feedback, a second draft of the APOSA index was constructed on December 4th, 2013.

4.3.4 The External Review Group Meeting

On March 12th, 2014, the External Review Group (ERG) meeting was conducted. The ERG was composed of end-users in the form of 11 family medicine residents from 1st and 2nd year. This group was not responsible for any content analysis per-se; instead it was responsible for reviewing the layout, simplicity and ease of use. It was also responsible for assessing the usefulness of the index in the healthcare setting and giving feedback on the feasibility of implementing the index in practice. In this meeting we explained the theory behind the index by briefly reviewing the literature and then explaining the purpose of the index. Subsequently we showed the group a pilot version of the index and requested feedback based on the following questions:

1- Do you understand the purpose of the index?

2- Do you understand what each factor is assessing?

    a. If not which one(s) do you not understand and why?

3- On a scale of 1 to 10, how simple would you rate this index to understand and use?

4- Would you use this index in your practice?

5- Other recommendations:

4.3.5 Reliability of the APOSA Index

The reliability of the APOSA index was tested from June 4th to June 23rd, 2014, within a group of 10 pediatric medicine residents from 1st to 3rd year, as well as among 10 orthodontic residents in their 1st to 3rd year.

A group of 15 patient charts ranging from 8-16 years old from the University of Alberta Inter-disciplinary Airway Research Clinic (I-ARC) with a spectrum of craniofacial patterns was selected as reliability test subjects, and their intra-oral and extra-oral photographs were used in the reliability assessment. A matrix was used to assess the patients' scores and making sure that none of the factor/levels were being expressed less than twice throughout the 15 patients selected (Fig 4.1). Instructions were given before the first reliability test for both the orthodontic and pediatric residents. In order to diminish recall bias, application of the index was repeated 1-2 weeks later. The residents were also timed to see the speed at which the APOSA index can be reliably conducted.

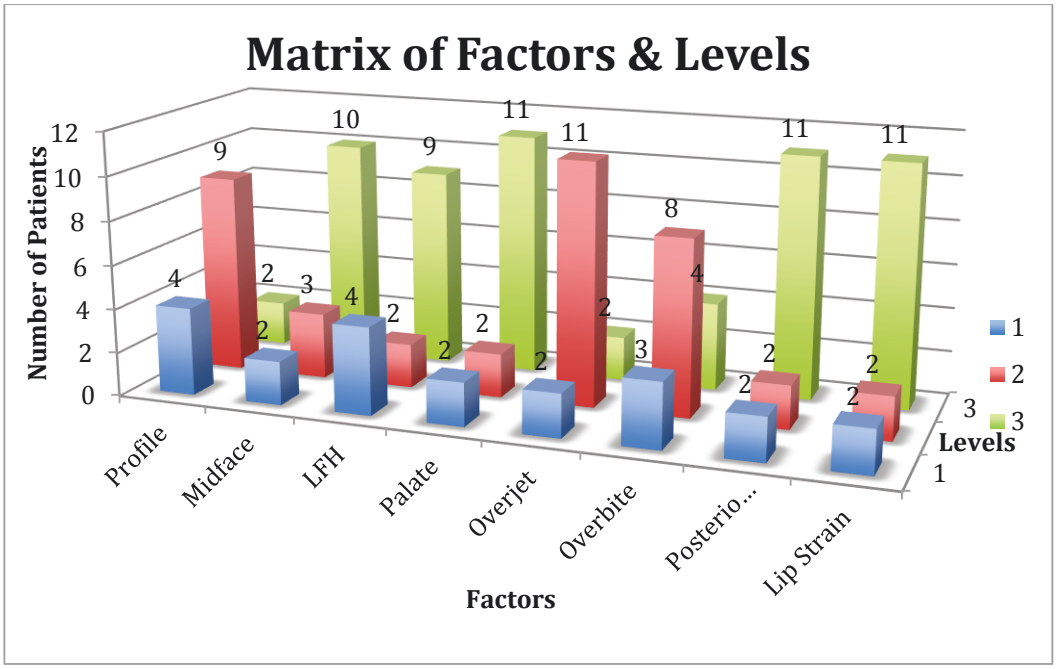


Figure 4.1.

The horizontal x-axis represents the different factors evaluated within the APOSA Index. The depth z-axis represents the various levels within each factor. Since each factor is divided into 3 levels on the APOSA Index, the levels from the left to the right of the index have been designated a number from 1 to 3 (ie, Profile: 1= Severely convex; 2=

Normal; 3= Concave). The vertical y-axis represents the amount of patients expressing that level within its respective factor.

For each factor of the APOSA index intra-rater agreement was evaluated using weighted Cohen Kappa statistics[20] and inter-rater reliability between the pediatric and orthodontic residents was evaluated using Fleiss' Kappa statistics.[21] The degree of agreement was categorized as poor ($k<0$), slight ($k = 0.0\text{-}0.20$), fair ($k = 0.21\text{-}0.40$), moderate ($k = 0.41\text{-}0.60$), substantial ($k = 0.61\text{-}0.80$), or almost perfect ($k = 0.81\text{-}1.0$) using accepted norms.[22]

Through this protocol we aimed to develop an index that fulfills all of the following criteria:[23]

1. Gradient of Numeric Values: The severity of the orthodontic treatment need within the pediatric OSA patients should be defined within a numerical scheme that demonstrates a finite and progressive gradient from low need to high need.
2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
3. Clinical Importance: The numerical scale should correspond with the clinically appraised orthodontic treatment need of pediatric OSA patients.
4. Statistical Ease: should be amendable to statistical analysis.
5. Reliability: Should have a high intra- and inter-rater reliability.

6. Practical: The instruments required to score the index should be practical to the setting in which it will be administered.
7. Minimal Judgment: Applying the index should require minimal judgment.
8. Simple: The index shouldn't have a high financial or time cost, therefore should be simple enough to administer to many patients.
9. Detect Change: The index should be able to detect changes in orthodontic treatment need in pediatric OSA patients.
10. Validity: should be valid over time.

4.4 Results

4.4.1 Scoping the Index

Scoping the results yielded 11 factors to be included in the initial stages of development: profile, midface, lower face height, face width, chin-throat depth, chin, lip incompetence, overjet, overbite, posterior bite, and palate.

4.4.2 Reviewing the Literature

After a thorough review of the literature it was found that the craniofacial morphology of patients with POSA patients are at risk for an increased ANB, decreased SNB, and an increased MP-SN angle on cephalometric analysis. Moreover, the orthodontic interventions identified to help with symptom relief of POSA were: mandibular advancing appliances for mild POSA cases associated with mandibular retrognathia, and rapid maxillary expander treatment combined with tonsillectomy in mild to moderate cases of POSA with a concurrent

maxillary constriction. Also, reverse pull headgear treatment and surgical treatment showed promising results.[1]

4.4.3 Drafting the Index

Many intra-oral and extra-oral factors were used in order to be able to assess the many different aspects of the craniofacial phenotype, and redundancy in the factors was purposely done in order to potentially increase the robustness of the index. Appendix C demonstrates the initially drafted index that was used in the IDG meeting. Each of the 11 included factors were further divided into 5 levels in order to maximize the inclusions in the initial draft of the index, allowing for subsequent removal of unnecessary items. Additionally 2 control factors were included which were crowding and neck circumference. Descriptions were also given for each of the factors in order to aid the medical professionals in knowing how to assess the specific factor. Profile was divided into severely convex, mildly convex, normal, mildly concave, and severely concave. Midface deficiency was divided into substantial loss of fullness, moderate loss of fullness, slightly below normal, normal, and overly full. Lower face height was divided into severely excessive, moderately excessive, mildly excessive, normal, and decreased. Face width was divided into severely narrow, narrow, moderately narrow, normal, and wide. Chin throat depth was divided into no chin-throat depth, minimal chin-throat depth, slightly below normal, normal, and excessive chin throat depth. Chin was divided into no chin prominence, minimal prominence, normal, strong prominence, and very strong prominence. Lip incompetence was divided into can't close lips, very strained closing lips, mildly strained closing lips, lips

slightly apart at rest, and no strain. Overjet was divided into excessive, increased, normal, edge-to-edge, and reverse. Overbite was divided into severely openbite, minimal openbite, normal, deep, and severely deep bite. Posterior bite was divided into bilateral crossbite, unilateral crossbite, cusp-to-cusp, normal, and telescopic bite. Palate was divided into very high arched palate, moderately high arched palate, minimally high arched palate, normal, and flat palate.

4.4.4 The Index Development Group Meeting

The results of the IDG Meeting are summarized in Table 1. Both negative controls were answered as anticipated, and helped verify the trustworthiness of the other responses, since it demonstrates that the participants understood the task at hand. When asked if crowding contributed to the orthodontic treatment-need the response was “yes” and had a median score of 9, which showed the highest level of importance. In this case because of the small sample size, the median of the responses is more meaningful. When asked if correcting this factor would help with the pediatric OSA symptoms the participants responded “no” with a median of 0. Neck circumference produced the opposite responses having no orthodontic relevance, a median score of 0, while correcting this factor was thought to improve OSA symptoms with a median score of 7 in the importance scale. The participants rated profile as a very important factor, with suggestions for the normal picture to be changed to slightly convex, which is the norm in younger children. Midface deficiency had moderate importance, although the participants did not like the proposed images that portray this factor. Lower face height was rated to have moderate importance. Face width had low importance, and the

participants wanted it removed. Chin-throat depth elicited mixed responses, some participants liked it while others severely disliked it, and ultimately the participants rated the factor to be of low importance. Similarly chin prominence, had mixed responses but was finally found to be of low importance. Lip incompetence was shown to have moderate importance, although the comments indicated that the pictures needed to be improved. Overjet was a very important factor, while overbite was of moderate importance. Posterior bite was of moderate importance, but the picture was deemed to be too complicated, and the palate was seen as being very important. Moreover, the consensus in the IDG meeting was that it was important to reduce the complexity of the index by removing factors and reducing the levels to 2-3 per factor. The drawings on the index were also deemed to look too old for the target population, which was the 7-13 year-old range. The participants also reiterated the need for such an index and its importance. The results from the post meeting survey are also summarized in figures 4.1 to 4.5. Additional non-structured feedback was requested and summarized below.

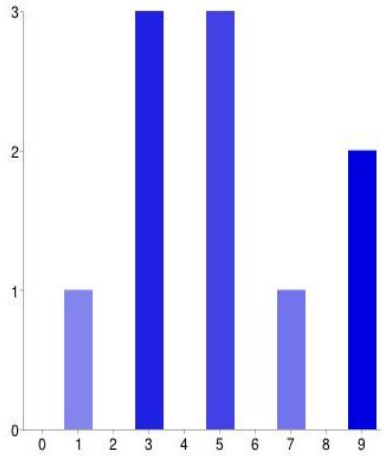
Table 4.1: Results of the Index Development Group Meeting

		C	NC	Pro	MD	LFH	FW	CTD	Chin	LI	OJ	OB	PB	Pal
1- Is this factor commonly observed in Pediatric OSA patients?	Median (Mean)	3 (2.91)	8 (6.55)	6 (5.64)	3 (3)	3 (3.43)	1.5 (2)	2 (3.33)	3.5 (3.5)	7 (6.08)	5.5 (5.2)	4 (3.72)	2 (2.7)	7 (6.27)
2- Does this factor contribute to Pediatric Obstructive Sleep Apnea symptoms?	Median (Mean)	2 (2.55)	8 (6.55)	7 (6.27)	4 (4.62)	3 (3.57)	1.5 (1.83)	2 (3.58)	2.5 (3.75)	4 (3.75)	5 (4.4)	3 (3.36)	2 (3.72)	7 (5.55)
3- Would correcting this factor help diminish the symptoms of Pediatric Obstructive Sleep Apnea?	Median (Mean)	0 (2.64)	7 (5.73)	7 (6.27)	5 (4.43)	4.5 (4.21)	0 (1.33)	2.5 (3.83)	2 (3.67)	4 (3.83)	5 (5.1)	3 (3.18)	2 (3.45)	8 (6.1)
4- Does this factor contribute to orthodontic treatment need?	Median (Mean)	9 (8.1)	0 (0.64)	7 (6.82)	6 (5.33)	6 (5.29)	1 (1.5)	3.5 (3.67)	4.5 (4.25)	6 (5.33)	8 (7.2)	7 (6.45)	8 (6.36)	8 (6.55)
5- Is the 5-point scale appropriate for this factor?	Median (Mean)	0 (1.82)	0 (1)	7 (6.45)	0 (1.29)	0 (0.86)	0 (1.83)	2 (3)	2 (3.25)	0 (2)	4 (4.1)	0 (2.45)	1 (2.64)	0 (2.55)
6- Are the points on the scale attributed correctly for this factor?	Median (Mean)	0 (2.45)	0 (2.09)	7 (5.91)	2 (3.48)	3 (3.76)	4 (3.17)	4.5 (3.83)	5 (4.25)	2 (3.33)	5.5 (4.5)	4 (4)	1 (2.82)	3 (3.55)
7- Are the pictures appropriate for this factor?	Median (Mean)	0 (1.45)	2 (1.82)	6 (4.64)	0 (1.14)	4 (3.5)	0 (1.25)	5 (3.92)	5 (4.83)	2.5 (3.25)	7 (6.1)	5 (4.54)	0 (1.73)	0 (1.45)

C= Crowding; NC= Neck Circumference; Pro= Profile; MD= Midface Deficiency; LFH= Lower Face Height; FW= Face Width; CTD= Chin-Throat Depth; LI= Lip Incompetence; OJ= Overjet; OB= Overbite; PB= Posterior Bite; Pal= Palate
*Based on the responses of the 13 participants

1. Do you think that this index is useful in fulfilling the goals of having an easy to use index that helps non-dentally trained professionals assess the orthodontic treatment need to help the symptoms in pediatric patients with OSA?

If "No", then mark 0. If "Yes" rate the importance from 1-9 by marking the appropriate number



0	0	0%
1	1	10%
2	0	0%
3	3	30%
4	0	0%
5	3	30%
6	0	0%
7	1	10%
8	0	0%
9	2	20%

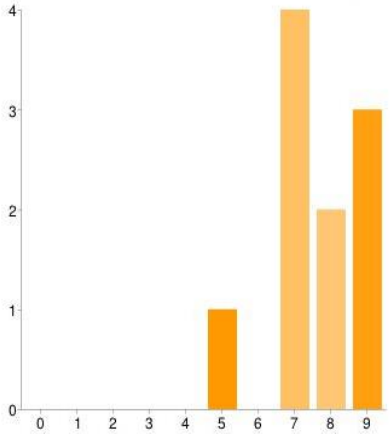
If "No", how would you improve it?

Good Start - Needs to be narrowed down and focused | Eliminate the questions that do not very specifically contribute to the stated goal of the index: helping physicians identify which patients could have a functional airway benefit from interceptive orthodontics. | Needs modification for sure. The idea is great, | Some portions were clearly outside the scope of practice of a gp or peditrician. I would. Select 3-4 scales for implementation. | Implement all the suggestions provided in previous feedback. Filter down questions to the one most relevant to the target audience. | Improve photos to more accurately represent reality.

Figure 4.1

2. Do you believe that the organizing committee is developing this index in the appropriate way? (i.e. Reviewing the recommendations in the literature, seeking expert opinions, gaining end-user approval and testing its reliability)

If "No", then mark 0. If "Yes" rate the importance from 1-9 by marking the appropriate number



0	0	0%
1	0	0%
2	0	0%
3	0	0%
4	0	0%
5	1	10%
6	0	0%
7	4	40%
8	2	20%
9	3	30%

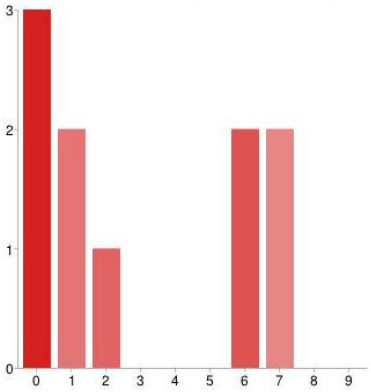
If "No", how would you improve it?

Good use of focus group | It is a step in the right direction. Good Job Mustafa!

Figure 4.2

3. In your opinion were all the factors, identified in this meeting, important factors for this index?

If "No", then mark 0. If "Yes" rate the importance from 1-9 by marking the appropriate number



0	3	30%
1	2	20%
2	1	10%
3	0	0%
4	0	0%
5	0	0%
6	2	20%
7	2	20%
8	0	0%
9	0	0%

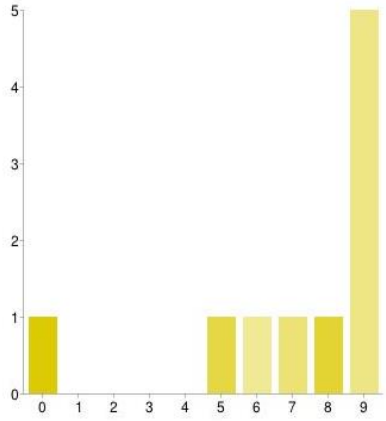
If "No", which factor(s) was/were not important?

Some more than others. Chin, and neck size, and crowding are not important | The less relevant factors have been discussed verbally. | For Orthodontic manipulation crowding and neck circumference not needed. Need to look at what family docs will actually do. | Yes, I do think all the important factors were identified. However, some of the important factors were poorly depicted in the examples (ie. mid face deficiency). Furthermore, I think there are too many questions. The index should be distilled.

Figure 4.3

4. In your opinion, have all the important areas concerning orthodontic treatment need to help symptoms of OSA been identified?

If "No", then mark 0. If "Yes" rate the importance from 1-9 by marking the appropriate number



0	1	10%
1	0	0%
2	0	0%
3	0	0%
4	0	0%
5	1	10%
6	1	10%
7	1	10%
8	1	10%
9	5	50%

If "No", which area(s) was/were missed?

as non orthodontic I do not have the knowledge to answer it | Again, all the important factors have been identified, although substantial improvements in the pictures are needed, and many images should be removed. | Chin position is not a factor, transverse issues would be not productive for MDs

Figure 4.4

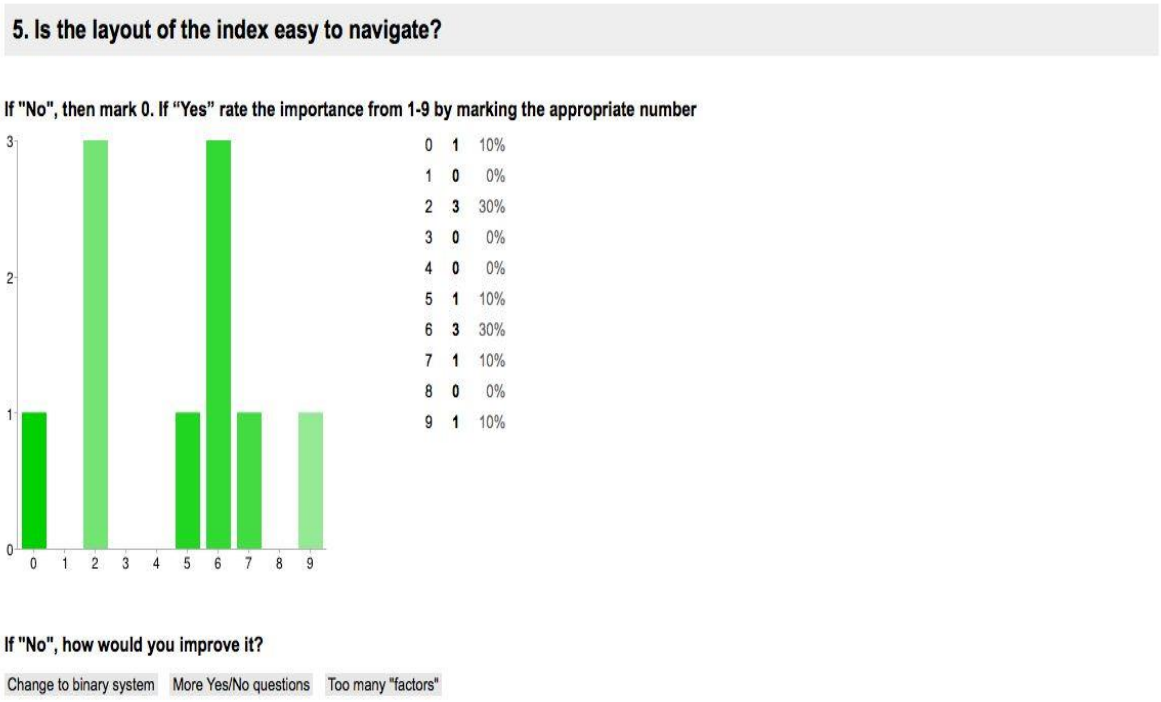


Figure 4.5

Additional feedback that may help improve the index:

- "Good start, need to focus and narrow"
- "Give physicians a list of orthodontic variables that contribute to OSA and under Yes/No"
- "Involve pediatricians and family physician focus groups"
- "My opinion would be to use age appropriate norms. For example most young kids are slightly convex in profile. Also as I've written in previous factor-specific comments, some of the pictures were not clear. Thanks!"

Email Feedback:

- "Should you move to clinical validation of the index let me know"
- "Excellent effort"

- "Great ideas will come from this project, for dentists, dental hygienists and MDs, but mostly patients will benefit if we can get the education of the professionals improved."
- "It is a very cool concept"
- "I applaud you for gathering all of us and for the idea of gathering various opinion from stakeholders."

All the recommendations were summarized and the index was modified to take into account the suggestions. Appendix D shows the revised version of the index.

4.4.5 The External Review Group Meeting

After a brief introduction and explaining the goal and uses of the APOSA index, 11 family medicine residents were shown the index and a brief review of what the available orthodontic interventions are for POSA. The index used in the ERG meeting is found in Appendix D, and the results of the ensuing questions for feedback are summarized in Figures 6-10. The results of this meeting were very encouraging by demonstrating that the family residents unanimously liked the idea of the index and would use it in their future practices.

Do you understand the point of the index?
The point of the index was clear to me. It would make a useful tool for family medicine.
Yes
To ID children presenting with symptoms of OSA who have craniofacial abnormalities who may benefit from orthodontic intervention
Yes
Yes
Purpose of the index to assess facial anomalies as contributory to OSA
Yes
Yes
Yes
To assess facial deformities and their contributions to OSA

Figure 4.6

Do you understand the factors of the index?
The handout helps to understand the factors of the index. It would take some practice on my part.
Yes
I understand most of the factors
Most of the imaging is intuitive but some of the parameters may benefit from a short written explanation
Yes
I don't think I would know how to quantify the midface deficiency
Yes
Is there a measurement of palate height we should look for? Or just appearance?
Yes I understand
The midface deficiency is hard to tell apart

Figure 4.7

On a scale of 1 to 10, how simple would it be to understand and use the index?
8/10: I'm not entirely sure what the total number of the index means. Once we count out the points what does the total score mean?
6/10: The question is how sensitive or specific would I be able to make the right assessment
7/10
10/10: Likely would get easier to use as time goes on
10/10: Once taught about it a bit
8/10: I'm not used to looking at these factors
10/10
9/10: the midface deficiency is difficult to understand
8/10: I would change the order of the pictures to go from the face to the teeth

Figure 4.8

Would you use the index in your practice?
If I have a child diagnosed with sleep apnea, I would most definitely consider using this tool
I would now look for these features quickly on physical exam
If I had the index available I would use it. To date I have not had a child who I suspected had OSA
Yes, if my patient population includes enough children with OSA
Yes
Sure. Don't suspect OSA in children too much (have actually never done it). So looking into our screening tools (e.g., Routine well child visits) may be another approach to increase uptake of your tool.
Yes

Figure 4.9

Other Recommendations
A short written explanation of each parameter
All good. Very interesting. Thanks
Thanks you

Figure 4.10

4.4.6 Reliability

Reliability tests were completed using the final version of the index included in Appendix E. The group of 10 orthodontic residents and 10 pediatric residents, each had a brief explanation of the index and how to score the index. The overall average time to score 15 patient photos using the APOSA index was 17:14 min, with the pediatric medicine residents seemingly taking a little longer to score the patient photos. The detailed results are summarized in Table 2. The agreement between the residents is also summarized in Tables 3 and 4. As expected the orthodontic residents had a higher level of inter-rater and intra-rater agreement, conceivably due to an increased level of training in the field of craniofacial and occlusal characterization. Although all the reliability scores show

an acceptable level of reliability, the pediatric residents seemed to have the most trouble assessing the midface deficiency, lower face height and lip strain on closing when comparing the ratings with that of the orthodontic residents. When comparing the ratings of the first and second trials, intra-rater, the precision of the index is demonstrated in that none of the factors are below a moderate level of agreement.

Table 4.2. Time Required to Score 15 Patients Using the APOSA Index

	Average	Range
Pediatric Residents Trial 1	21:44 min	17:04 min – 30:23 min
Pediatric Residents Trial 2	18:02 min	13:57 min – 23:11min
Orthodontic Residents Trial 1	14:51 min	12:20 min – 18:45 min
Orthodontic Residents Trial 2	14:19 min	12:42 min -17:59 min

Table 4.3. Inter-Rater Reliability

Factor	Inter-rater reliability Within Both Pedo and Ortho Residents (N=20)		Inter-rater reliability Within Pedo Residents (N=10)		Inter-rater reliability Within Ortho Residents (N=10)	
	Kappa (95%CI)	Classification	Kappa (95%CI)	Classification	Kappa (95%CI)	Classification
Profile	0.493 (0.463, 0.523)	Moderate	0.432 (0.376, 0.489)	Moderate	0.561 (0.499, 0.623)	Moderate
Midface Deficiency	0.324 (0.294, 0.354)	Fair	0.227 (0.167, 0.288)	Fair	0.416 (0.352, 0.480)	Moderate
Lower Face Height	0.337 (0.308, 0.366)	Fair	0.247 (0.188, 0.307)	Fair	0.479 (0.420, 0.540)	Moderate
Lip Strain ,Close	0.403 (0.375, 0.431)	Fair	0.260 (0.204, 0.317)	Fair	0.567 (0.509, 0.626)	Moderate
Palate	0.405 (0.376, 0.433)	Moderate	0.375 (0.316, 0.434)	Fair	0.416 (0.363, 0.469)	Moderate
Overjet	0.536 (0.499, 0.573)	Moderate	0.482 (0.405, 0.560)	Moderate	0.643 (0.586, 0.670)	Substantial
Overbite	0.532 (0.506, 0.559)	Moderate	0.416 (0.362, 0.471)	Moderate	0.764 (0.709, 0.819)	Substantial
Posterior Bite	0.587 (0.558, 0.616)	Moderate	0.437 (0.377, 0.497)	Moderate	0.743 (0.685, 0.801)	Substantial

Table 4: Intra-Rater Reliability

	Profile Overall	Midface Deficiency	Lower Face Height	Lip Strain, Close	Palate	Overjet	Overbite	Posterior Bite
Medical Pediatric Resident, Kappa (95%CI)								
Rater 1	0.762 (0.311,1.213)	0.646 (0.287, 1.004)	0.482 (0.049,0.917)	0.815 (0.464,1.166)	0.802 (0.548,1.057)	0.835 (0.523,1.147)	0.839 (0.533,1.144)	0.769 (0.472,1.067)
Rater 2	0.602 (0.199, 1.005)	0.839 (0.631,1.047)	0.229 (0.090,0.547)	0.662 (0.380,0.946)	0.746 (0.488,1.003)	0.769 (0.471,1.067)	0.734 (0.464,1.003)	0.749 (0.404,1.093)
Rater 3	0.672 (0.339, 1.004)	0.576 (0.273, 0.879)	0.326 (0.007 ,0.645)	0.767 (0.531,1.003)	0.472 (0.187, 0.758)	0.516 (0.147,1.179)	0.786 (0.509,1.062)	0.643 (0.254,1.032)
Rater 4	0.722 (0.364,1.081)	0.393 (0.0647,0.721)	0.294 (-0.120 ,0.708)	0.359 (-0.099,0.818)	0.630 (0.366,0.895)	0.600 (0.264,0.936)	0.663 (0.380,0.946)	0.869 (0.623,1.117)
Rater 5	0.589 (0.244,0.934)	0.634 (0.327,0.941)	0.444 (0.100, 0.789)	0.659 (0.373, 0.945)	0.502 (0.169,0.836)	0.839 (0.533,1.144)	0.610 (0.283,0.937)	1.000 (1.000, 1.000)
Rater 6	0.762 (0.311,1.213)	0.743 (0.483,1.003)	0.546 (0.220,0.871)	0.578 (0.223,0.932)	0.561 (0.248,0.875)	0.685 (0.367,1.004)	0.724 (0.445,1.003)	0.803 (0.548,1.057)
Rater 7	0.706 (0.326,1.085)	0.539 (0.151,0.926)	0.305 (-0.162,0.771)	0.460 (0.074 ,0.847)	0.477 (0.072,0.881)	0.746 (0.418,1.074)	0.494 (-0.017,1.006)	0.835 (0.623,1.048)
Rater 8	0.671 (0.339,1.004)	0.400 (-0.104,0.904)	0.500 (0.114,0.886)	0.688 (0.425,0.950)	0.667 (0.387, 0.946)	0.766 (0.463,1.068)	0.815 (0.576,1.054)	0.667 (0.237,1.097)
Rater 9	0.722 (0.364,1.081)	0.494 (-0.017,1.006)	0.390 (-0.046,0.827)	0.620 (0.302,0.939)	0.583 (0.234,0.933)	0.727 (0.451,1.003)	0.794 (0.529,1.059)	1.000 (1.000, 1.000)
Rater 10	0.845 (0.646,1.045)	0.727 (0.451,1.003)	0.539 (0.151,0.926)	0.639 (0.335,0.942)	0.757 (0.511,1.003)	0.565 (0.200,0.930)	0.764 (0.526,1.003)	0.857 (0.587,1.128)
Median (Mean±SD)	**0.714 (0.705±0.08)**	**0.605 (0.599±0.15)**	**0.417 (0.405±0.11)**	**0.649 (0.624±0.13)**	**0.606 (0.619±0.12)**	**0.736 (0.704±0.11)**	**0.749 (0.722±0.11)**	**0.819 (0.819±0.12)**
Classification [22]	**Substantial**	**Moderate**	**Moderate**	**Substantial**	**Substantial**	**Substantial**	**Substantial**	**Almost perfect**
Dental Orthodontic Resident, Kappa (95%CI)								
Rater 1	1.000 (1.000, 1.000)	0.887 (0.674,1.101)	0.689 (0.376,1.004)	0.805 (0.554,1.057)	0.839 (0.631,1.047)	1.000 (1.000, 1.000)	1.000 (1.000, 1.000)	1.000 (1.000, 1.000)
Rater 2	0.750 (0.427,1.073)	0.857 (0.587,1.128)	0.640 (0.276,1.004)	0.681 (0.358,1.004)	0.734 (0.464,1.003)	0.869 (0.623,1.116)	0.789 (0.516,1.061)	1.000 (1.000, 1.000)
Rater 3	0.892 (0.688,1.096)	0.878 (0.647,1.109)	0.803 (0.548,1.057)	0.727 (0.4512,1.003)	0.615 (0.226,1.005)	0.902 (0.716,1.088)	1.000 (1.000, 1.000)	0.794 (0.529,1.060)
Rater 4	0.886 (0.669,1.102)	0.835 (0.523,1.147)	0.634 (0.327 ,0.941)	0.731 (0.458,1.003)	0.835 (0.523,1.147)	0.712 (0.339,1.084)	0.910 (0.740,1.080)	0.878 (0.647,1.109)
Rater 5	0.795 (0.529,1.059)	0.762 (0.455,1.069)	0.803 (0.548,1.057)	0.802 (0.548,1.057)	0.805 (0.554,1.057)	0.862 (0.602,1.123)	0.869 (0.623,1.117)	1.000 (1.000, 1.000)
Rater 6	0.878 (0.647,1.109)	0.559 (0.079,1.039)	0.694 (384,1.004)	0.516 (0.216, 0.816	0.831 (0.614,1.049)	1.000 (1.000, 1.000)	0.754 (0.417,1.091)	0.898 (0.705,1.091)
Rater 7	0.887 (0.674,1.101)	0.657 (0.186,1.127)	0.500 (0.080,0.919)	0.609 (0.213,1.005)	0.706 (0.303,1.109)	0.779 (0.495,1.064)	1.000 (1.000, 1.000)	1.000 (1.000, 1.000)
Rater 8	1.000 (1.000, 1.000)	0.757 (0.511,1.003)	0.717 (0.431,1.003)	0.6457 (0.287,1.004)	0.602 (0.199,1.005)	1.000 (1.000, 1.000)	0.901 (0.713, 1.089)	1.000 (1.000, 1.000)
Rater 9	0.899 (0.709,1.090)	0.609 (0.072,1.145)	0.640 (0.276,1.004)	0.819 (0.586,1.053)	0.615 (0.119,1.112)	0.789 (0.516,1.061)	0.911 (0.743, 1.079)	1.000 (1.000, 1.000)
Rater 10	0.878 (0.647,1.109)	0.468 (-0.013 ,0.949)	0.810 (0.565,1.055)	0.769 (0.536,1.003)	0.792 (0.523,1.061)	0.901 (0.713,1.089)	0.911 (0.743,1.079)	0.795 (0.529,1.060)
Median (Mean±SD)	**0.886 (0.886±0.07)**	**0.759 (0.726±0.14)**	**0.691 (0.693±0.09)**	**0.729 (0.710±0.09)**	**0.763 (0.737±0.10)**	**0.885 (0.881±0.10)**	**0.910 (0.904±0.08)**	**1.000 (0.936±0.09)**
Classification [22]	**Almost perfect**	**Substantial**	**Substantial**	**Substantial**	**Substantial**	**Almost perfect**	**Almost perfect**	**Almost perfect**

4.5 Discussion

No prior index has tackled the issue of the orthodontic-need among POSA patients with the aim of helping alleviate their symptoms, The APOSA is a first step to allow better communication between physicians and orthodontists in establishing the most appropriate treatment. Previously, most orthodontic indices that have been developed have relied on individual expertise or that of a small group of 2-4 individuals. More contemporary indices that have been developed have relied on the previous indices and modified what they perceived as weaknesses.[24] The only orthodontic malocclusion index that has a different method of development is the Index of Complexity, Outcome and Need (ICON), and much of the strengths of its methodology have been adapted into the development of the APOSA index.

The reliability of the index shows great promise. In comparison, the reliability of the different components of the ICON range from 0.3032 -0.8420 in the treatment need part, and from 0.2519 - 0.5914 in the treatment outcome part. Since the ICON is an accepted and commonly used and given that the APOSA Index's scores are similar it is reasonable to conclude that the reliability scores of the APOSA Index are acceptable. Limitations of the reliability test can be divided into factors that are index specific, rater specific and context specific. The index specific factors pertain to the amount of factors, and the amount of levels. These were reduced iteratively. The rater specific factors pertain to the skill and experience of the raters. That is why there was a difference between the responses of the orthodontic residents and the pediatric residents. Finally the context

specific factors pertain to the explanation given to the residents, the level of comfort of the participants, and the ease which the patient information is displayed. In all cases, the residents were solicited during their lunch break and may have either been fatigued from a long morning or felt pressured to finish the index in order to enjoy their lunch. Also, the index was applied on 2 dimensional photos which differs from the application of the index in a clinical setting. A more controlled repeated reliability study may show improved results.

The time required to apply the index (1:09 min) could be further reduced with experience as seen by the second trial having a shorter average time than the first in both resident groups. Also, with increased education and awareness of the factor groups used in this index, physicians will be able to conduct the index much more accurately, and much quicker.

Although craniofacial morphology does change with age and differs ethnicity, the index attempts to accommodate for this fact by marking extreme deviations from the norm. That way the natural variance in normal craniofacial phenotype across, age, gender, and ethnicity can be mitigated. Furthermore the index is intended for use on patients of 7-18 years old, thus eliminating the craniofacial features of the young children in the normal pool.

Color-coding was attempted to represent a specific marking scheme, although for the purposes of this stage of the index development it was irrelevant.

Further research still needs to be done to validate the APOSA index. Assuming that the “gold standard” in assessing the orthodontic treatment-need in pediatric patients with OSA is an orthodontist with experience in dealing with

pediatric OSA patients, then the APOSA index results need to be compared to that of the orthodontists' responses in a separate study.

4.5.1 Significance

The APOSA index will help physicians identify which craniofacial phenotypes may benefit from orthodontic treatment as part of their multi-disciplinary OSA management. Furthermore, due to the diverse medical effects of sleep deprivation, it would be desirable to make sleep apnea into a centralized service, where the main focus is for a highly trained multidisciplinary team to treat a high volume of patients to a standardized protocol, where meticulous documentation is exercised. This index may form part of that documentation process. It will allow for quality assurance, funding allocation and epidemiologic studies to be performed. It will also for audits and allow comparisons with other centers.

4.5.2 Dissemination Plan

The dissemination of the APOSA index will be done through a variety of ways in order to maximize its reach. This index can be used within the medical curriculum, as a learning opportunity for the students, to teach them about facial and oral assessment of OSA and the application of the index. Judging by the feedback from the residents during the developmental stages, the index will serve as an opportunity for inter-professional collaborative education. Also, the index will be published in a peer-reviewed journal, which will allow its introduction to the scientific literature. The journal should be a respected medical journal with broad reach, in order to allow the greatest number of physicians to be exposed to

the index. Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian and American population of medical professionals. Moreover, the index will be presented at national and international conferences to increase the awareness of the index among the scientific community. Finally, the APOSA index will be used at the University of Alberta's Interdisciplinary Airway Research Center, so that future research in this center will incorporate it. It will also be placed on the University of Alberta's Interdisciplinary Airway Research Center's website under the physician section, to further educate the doctors who visit the website on the index and allow easy download.

4.5.3 Ethics Approval

The proposed research has received the ethical approval numbered Pro00045067 from the University of Alberta Ethics Board.

4.5.4 CONCLUSION

Identifying candidates for orthodontic treatment has been simplified for physicians by developing the Alberta Obstructive Sleep Apnea (APOSA) Index.

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Chapter 5: Discussion and Conclusion

5.1 Discussion

Throughout the development process of the Alberta Pediatric Obstructive Sleep Apnea (APOSA) Index, medical professionals from several specialties have praised both the process by which this development has taken place and reaffirmed the need for the index. Although the development phase is now complete, the index is far from complete. The index now needs to be validated against a group of expert orthodontists to attribute meaning to the numeric scores of the index. Once this next phase is complete, the index will be ready for general use as a validated and reliable index.

The shortcomings of this research centered on the principal investigator's inability to maintain the invited group of participants within the meeting until the meeting was completed. This happened on a number of occasions and for different reasons. The participants in all the meetings were not paid and volunteered their time. In the Index Development Group (IDG) meeting the participants were highly trained individuals with very strict time constraints and using the delphi approach for the meeting proved extremely difficult to implement. Due to the structure of the meeting that encouraged feedback and the willingness of the participants to give ample feedback, the meeting quickly went over the expected time limit. Modification of the Delphi procedure by grouping factors together for discussion was implemented in order to allow for proper feedback, yet allow the meeting to complete not too much later than anticipated. This sudden change was made in consultation with the methodologist present in

the meeting in order to allow for all the participants to give in their feedback before they left the meeting, and in order to diminish the rising level of stress associated with being significantly behind schedule. The diminishment of the tension in the room subsequently allowed for more relaxed, natural and appropriate responses.

The External Review Group (ERG) meeting with the family medicine residents took place prior to their scheduled classes, and the time was also constrained by the subsequent instructor wanting to start prior to finishing the meeting agenda. After pleading with the instructor they allowed us additional time to complete the meeting, although the meeting was completed at a quicker pace than anticipated.

Reliability is the overall consistency and reproducibility of a measurement. The conditions under which the reliability tests were conducted were difficult to control since it required taking time from the participants during their lunchtime. Despite the circumstances, the results still showed an acceptable overall reliability, and although these results may represent an under-estimation of the actual reliability of the APOSA Index, a more dedicated group of participants would likely be required in subsequent reliability tests. Further limitations of this study were that the reliability was assessed using students rather than licensed clinicians, which would also give a more conservative estimate of the actual reliability of the index. Also, 2 dimensional images were used rather than actual patients, which could be particularly problematic for assessing the factors of midface deficiency, lip strain on closing, and palate. These 3 factors all had

poorer reliability scores than the rest of the factors. Moreover, just because a tool is consistent at measuring something, doesn't mean that it's consistently measuring what it's intended to measure, therefore it is important to follow this study with a validation study. The validation study will assess if the index is measuring the orthodontic treatment need in pediatric patients with OSA that may get a functional benefit from orthodontic treatment. Since this tool will serve as a screening tool of patients diagnosed with OSA, it is necessary that the tool have a really high sensitivity, in order to capture as many of the patients that may benefit from orthodontic treatment as possible. Since this is not a diagnostic or prognostic index, it will then be up to the orthodontist to make the final diagnosis and treatment plan for the patient.

Since this index is intended for use amongst physicians, future research is also needed to evaluate the reliability of the APOSA Index amongst nurses, in order to assess whether conducting the index is a delegate-able task, which may increase the uptake of the index. Moreover, profession-specific modifications of the index may be indicated to cater to different healthcare professionals, such as dentists, nurses, etc. Examples include: simplifying the index to dichotomous responses only, or increasing the complexity of the intra-oral measurements in the case of dentists because they are more experienced in oral measurements.

5.2 Conclusion

The development of the APOSA Index represents a peer-reviewed and accepted, user-friendly index that reliably allows medical professionals to assess

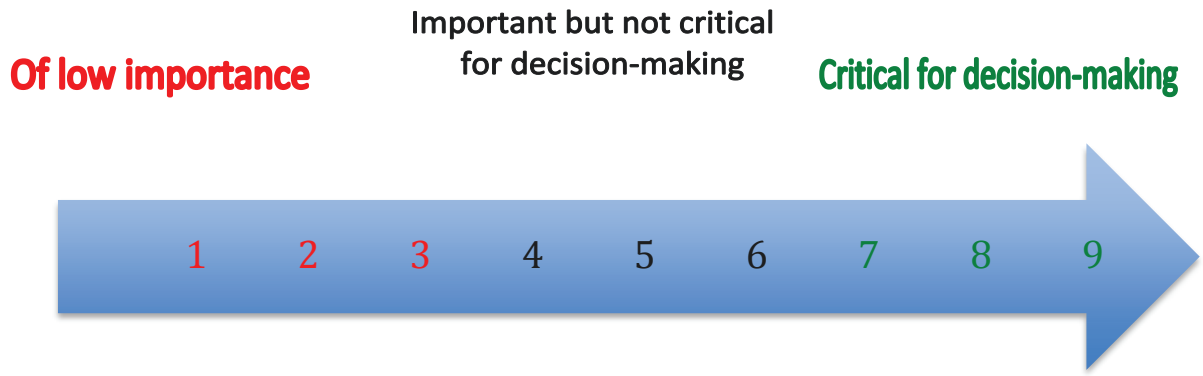
the orthodontic treatment-need in patients with pediatric obstructive sleep apnea that may benefit from the orthodontic treatment.

APPENDICES

APPENDIX A

Assigned Number_____

Factor Being Evaluated:_____



Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.

1- In your opinion, is this factor commonly observed in Pediatric Obstructive Sleep Apnea patients?

Yes **1 2 3 4 5 6 7 8 9**

No

2- In your opinion, does this factor contribute to Pediatric Obstructive Sleep Apnea symptoms?

Yes **1 2 3 4 5 6 7 8 9**

No

3- In your opinion, would correcting this factor help diminish the symptoms of Pediatric Obstructive Sleep Apnea?

Yes **1 2 3 4 5 6 7 8 9**

No

4- In your opinion, does this factor contribute to orthodontic treatment need?

Yes **1 2 3 4 5 6 7 8 9**

No

5- In your opinion, is the 5-point scale appropriate for this factor?

Yes **1 2 3 4 5 6 7 8 9**

No; How would you correct it?

6- Are the points on the scale attributed correctly for this factor?

Yes **1** **2** **3** **4** **5** **6** **7** **8** **9**

No; How would you correct it?

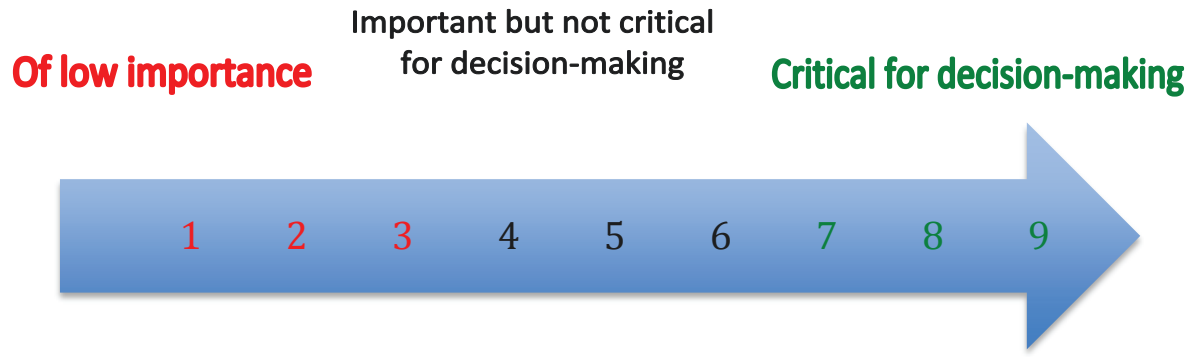
7- In your opinion, are the pictures appropriate for this factor?

Yes **1** **2** **3** **4** **5** **6** **7** **8** **9**

No; How would you correct it?

APPENDIX B

Assigned Number_____



Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.

1. Do you think that this index is useful in fulfilling the goals of having an easy to use index that helps non-dentally trained professionals assess the orthodontic treatment need to help the symptoms in pediatric patients with OSA?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

_____

_____

2. Do you believe that the organizing committee is developing this index in the appropriate way? (i.e. Reviewing the recommendations in the literature, seeking expert opinions, gaining end-user approval and testing its reliability)

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

_____

_____

3. In your opinion were all the factors, identified in this meeting, important for this index?

Yes 1 2 3 4 5 6 7 8 9

No; Which factors were not important?

_____

_____

4. In your opinion, have all the important areas concerning orthodontic treatment need to help symptoms of OSA been identified?

Yes **1** **2** **3** **4** **5** **6** **7** **8** **9**

No; Which ones were missed?

_____

_____

5. Is the layout of the index easy to navigate?

Yes **1** **2** **3** **4** **5** **6** **7** **8** **9**

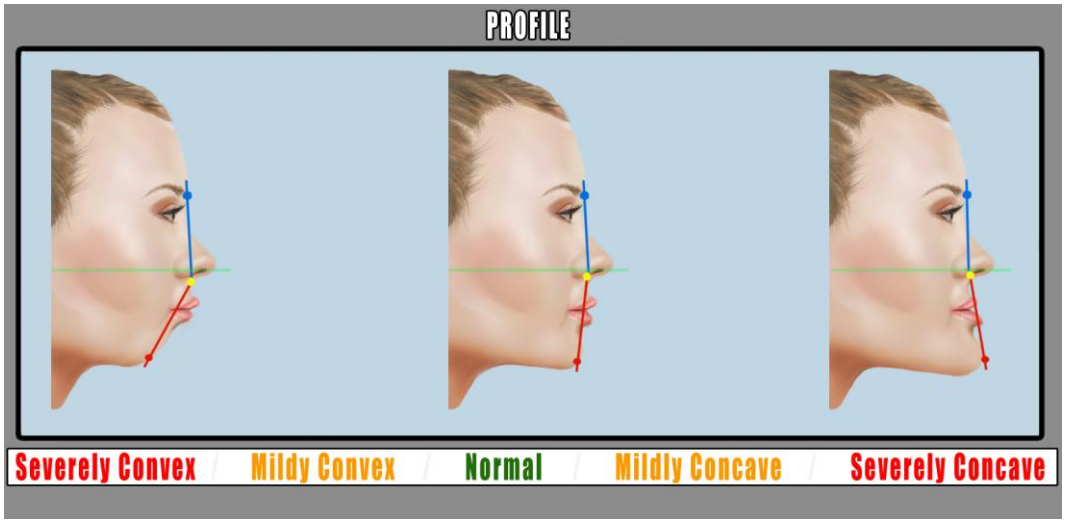
No; How would you improve it?

_____

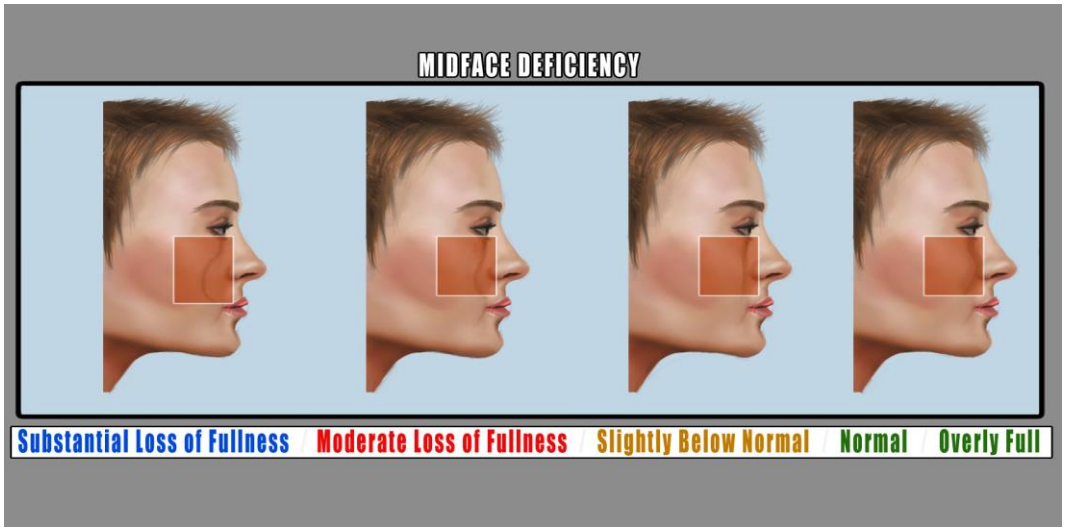
_____

Please provide any other feedback below that may help improve the index:

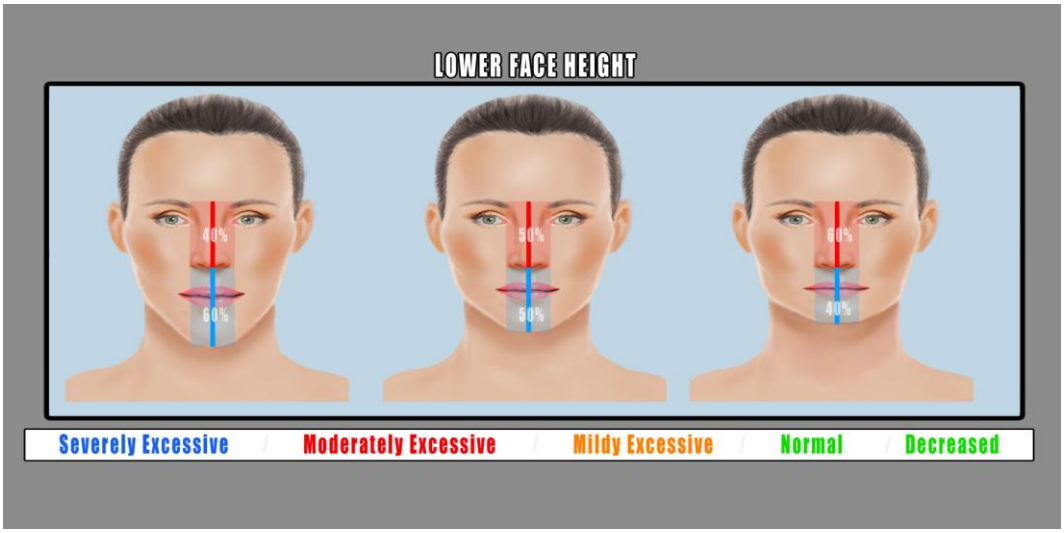
APPENDIX C



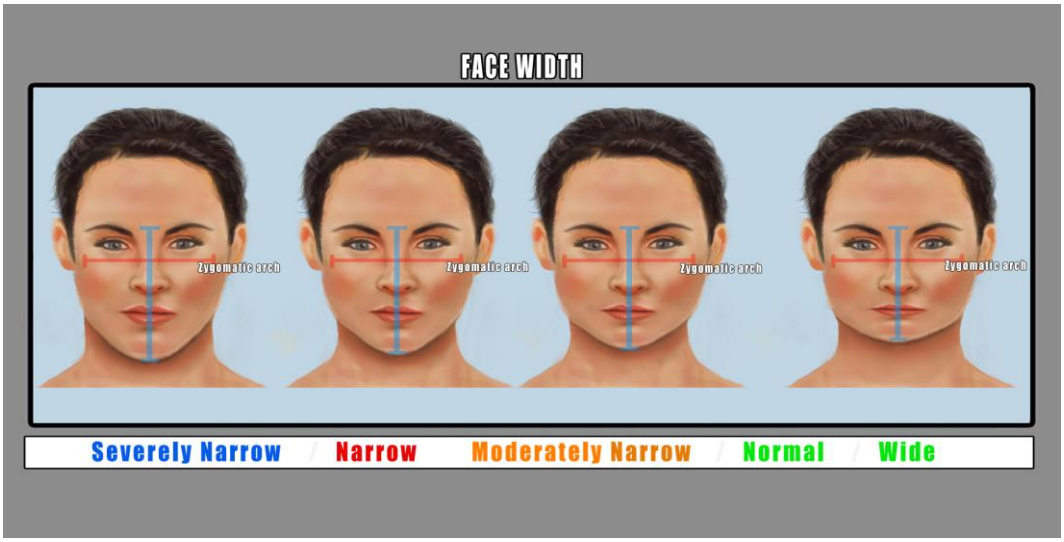
Description: While observing the patient from side view, consider a line from in between the eyebrows to the base of the nose and then to the chin. Evaluate the angle formed between the points.



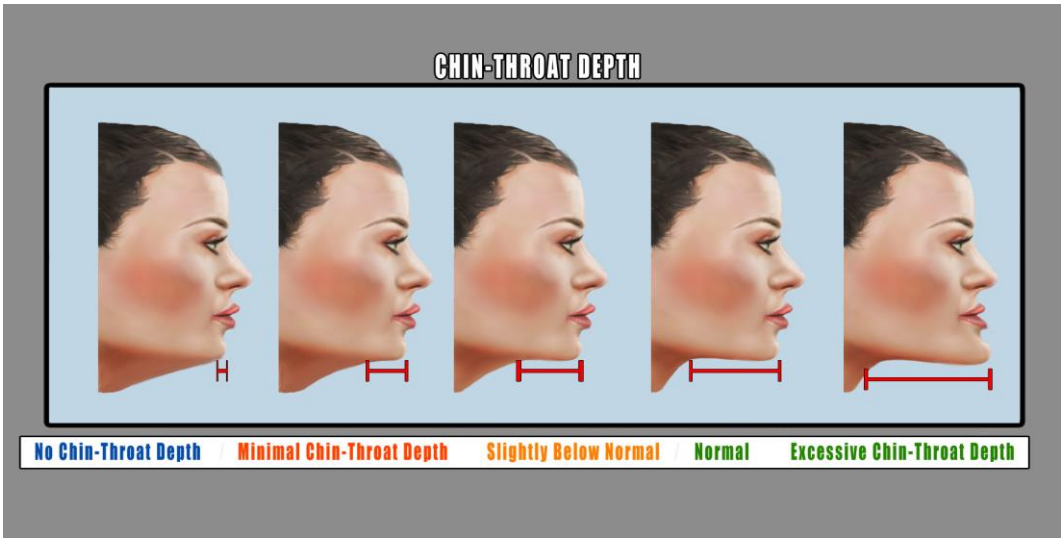
Description: While observing the patient from the left and right side views, consider the projection of the malar area below the eyes.



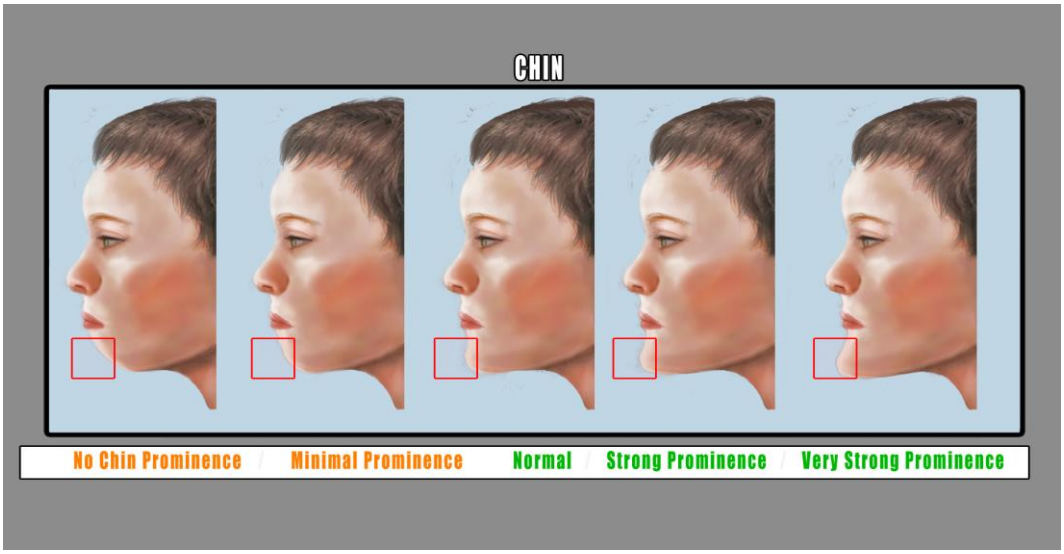
Description: While observing the patient from front view, consider the distance from between the eyebrows to the base of the nose, compared to the distance from the base of the nose to the chin. The normal proportion is approximately 50:50.



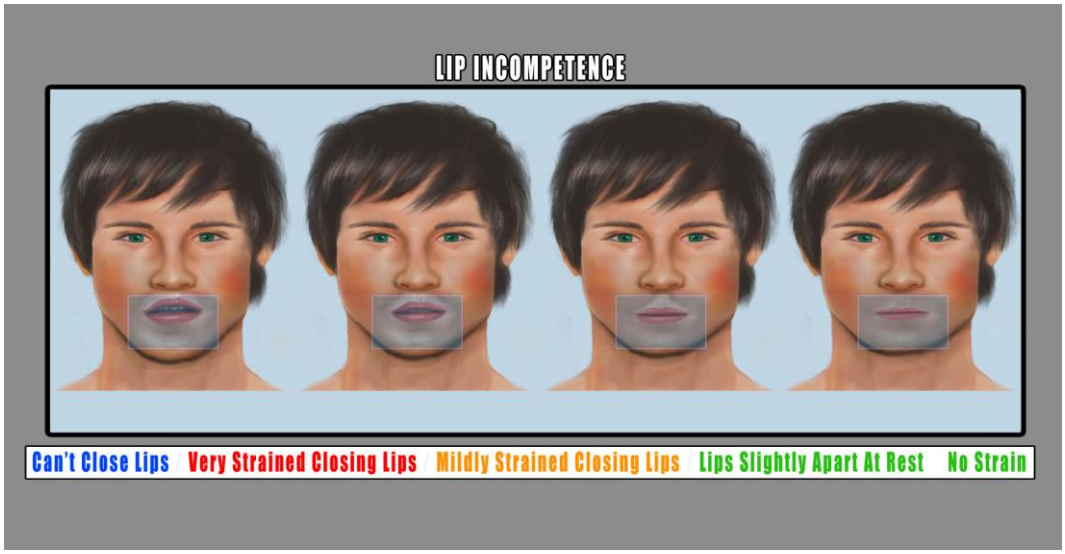
Description: While observing the patient from front view, consider the distance from between the zygomatic arches, compared to the distance from between the eyebrows to the bottom of the chin. The normal proportion is the width should be 81-93%.



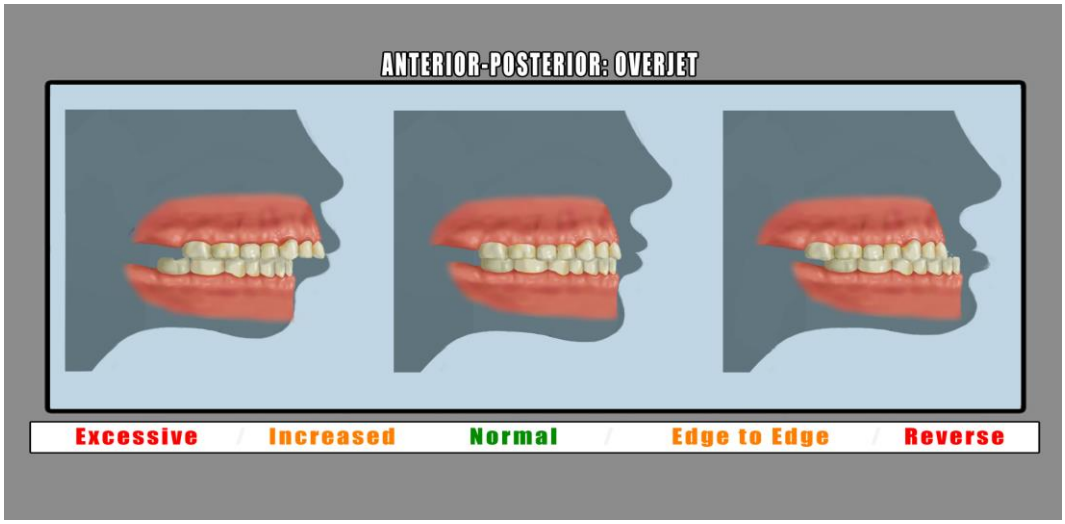
Description: While observing the patient from the side view, consider the distance from the most inferior and anterior portion of the chin to the beginning of the curvature of the throat.



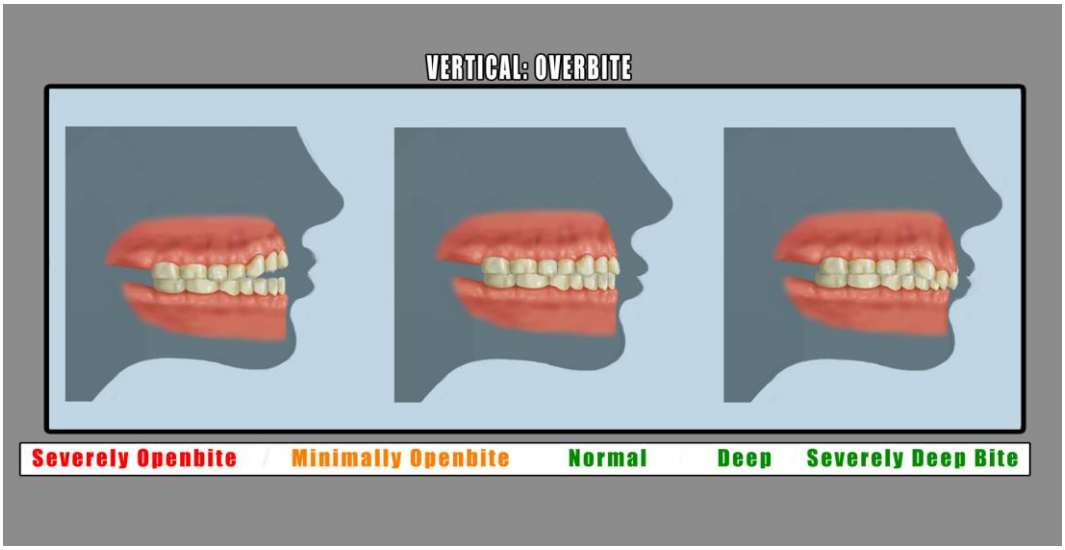
Description: While observing the patient from side view, consider the chin projection relative to the rest of the face.



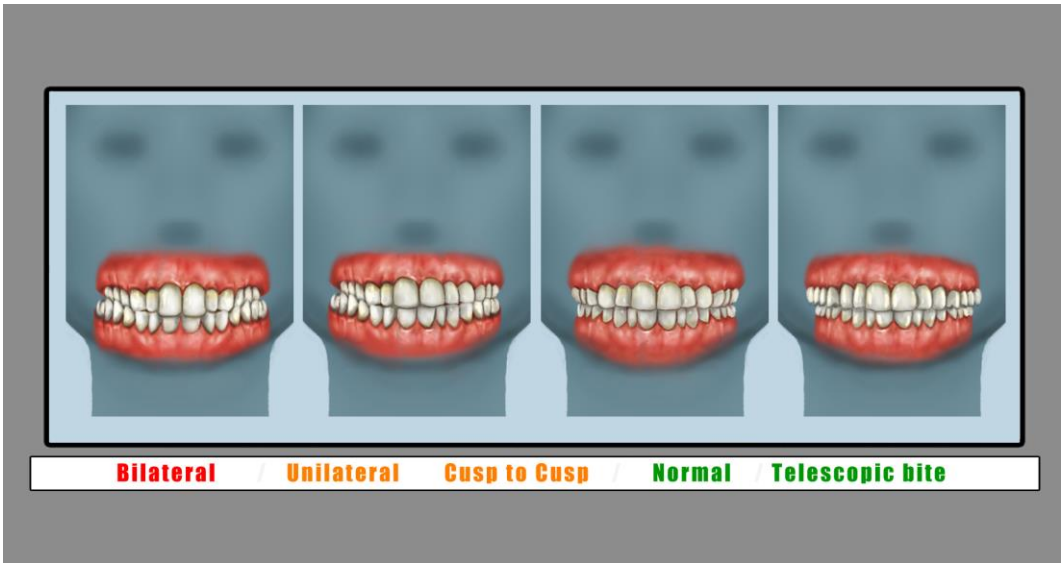
Description: While observing the patient from front view, consider the amount of lip closure the patient has on demand.



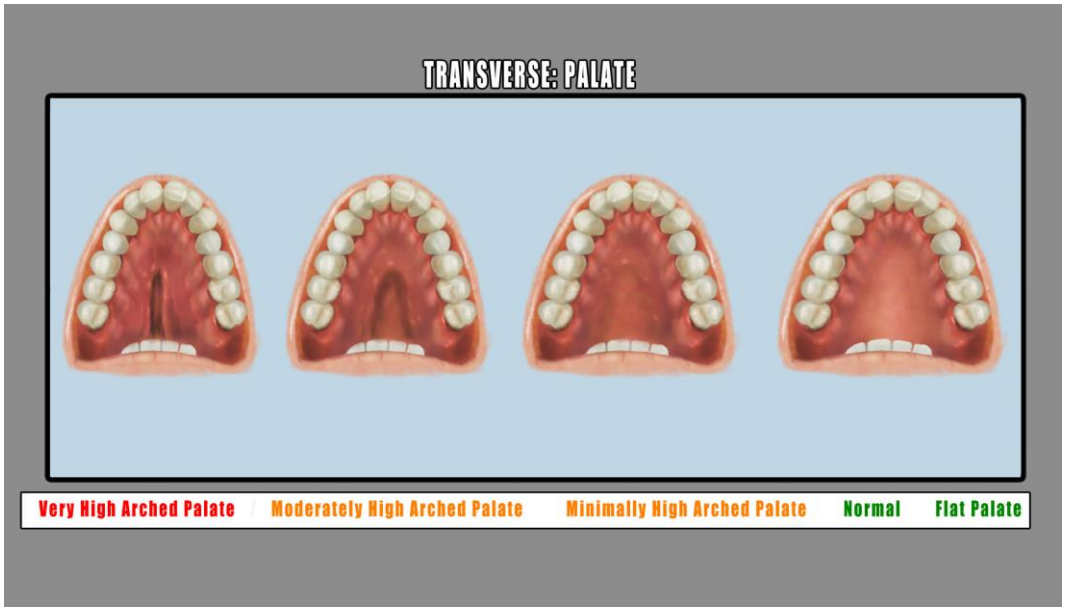
Description: This is the horizontal distance between the buccal surfaces of the lower incisors to the buccal surfaces of the upper incisors. An excessive overjet is greater than 9mm.



Description: This is the vertical overlap between the upper and lower incisors.



Description: This is the transverse relationship of the molars and premolars.



Description: View the depth of the palate relative to the teeth.

Crowded Maxillary and Mandibular Teeth

Severely Crowded Moderately Crowded Mildly Crowded No Crowding/Spacing Spacing

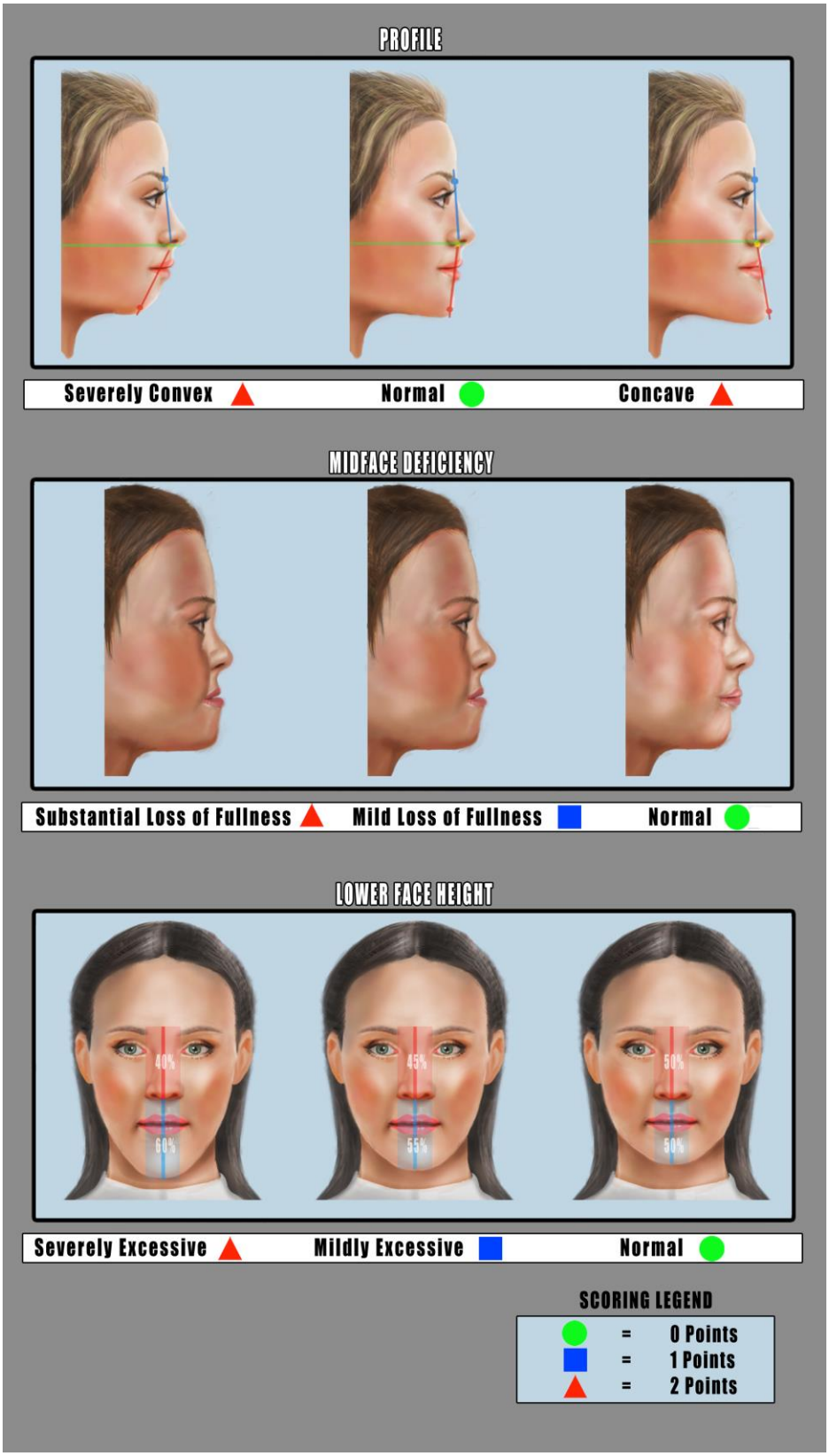
Description: The sum of the crowding in the upper jaw and lower jaw.

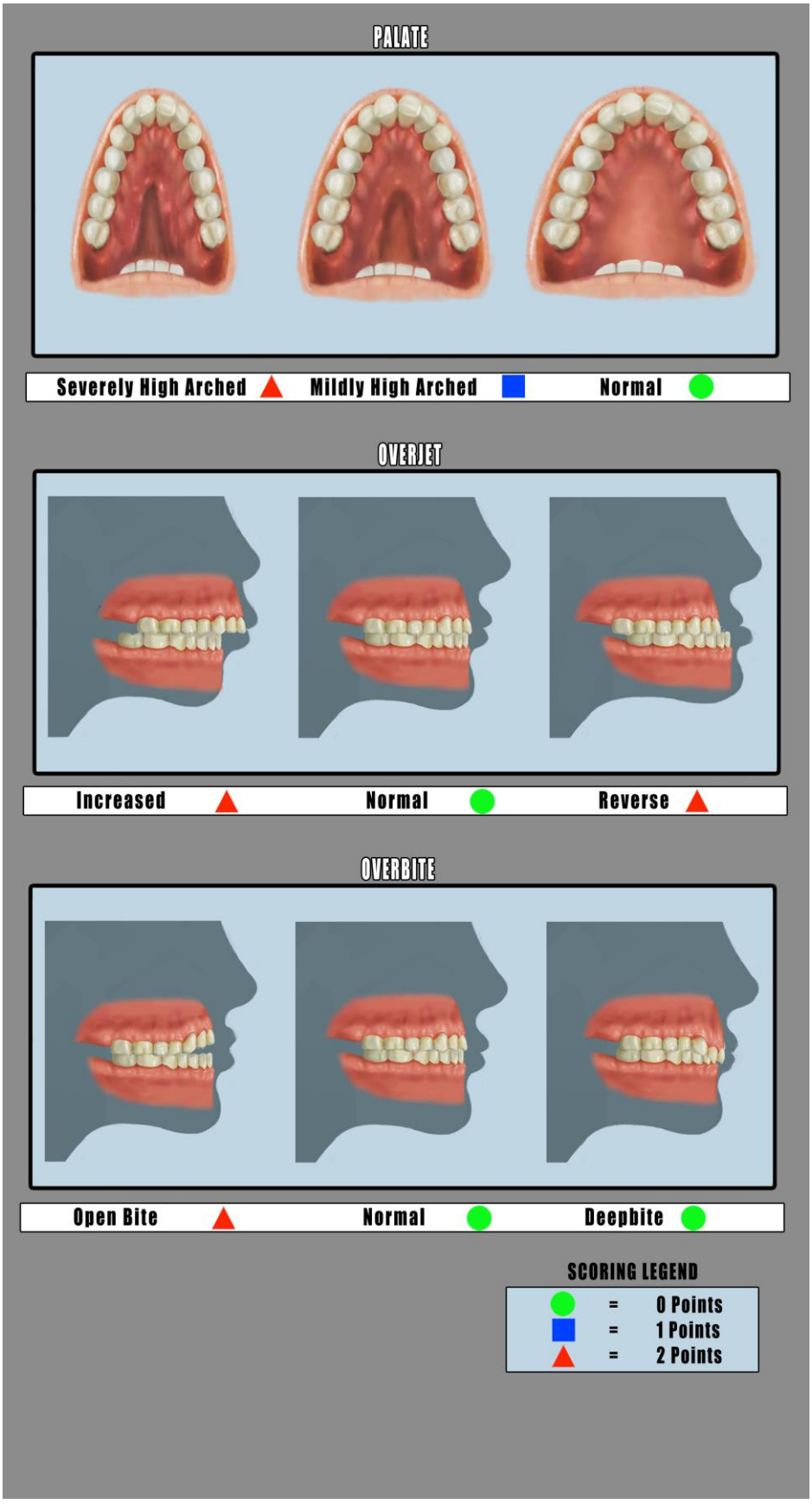
Neck Circumference

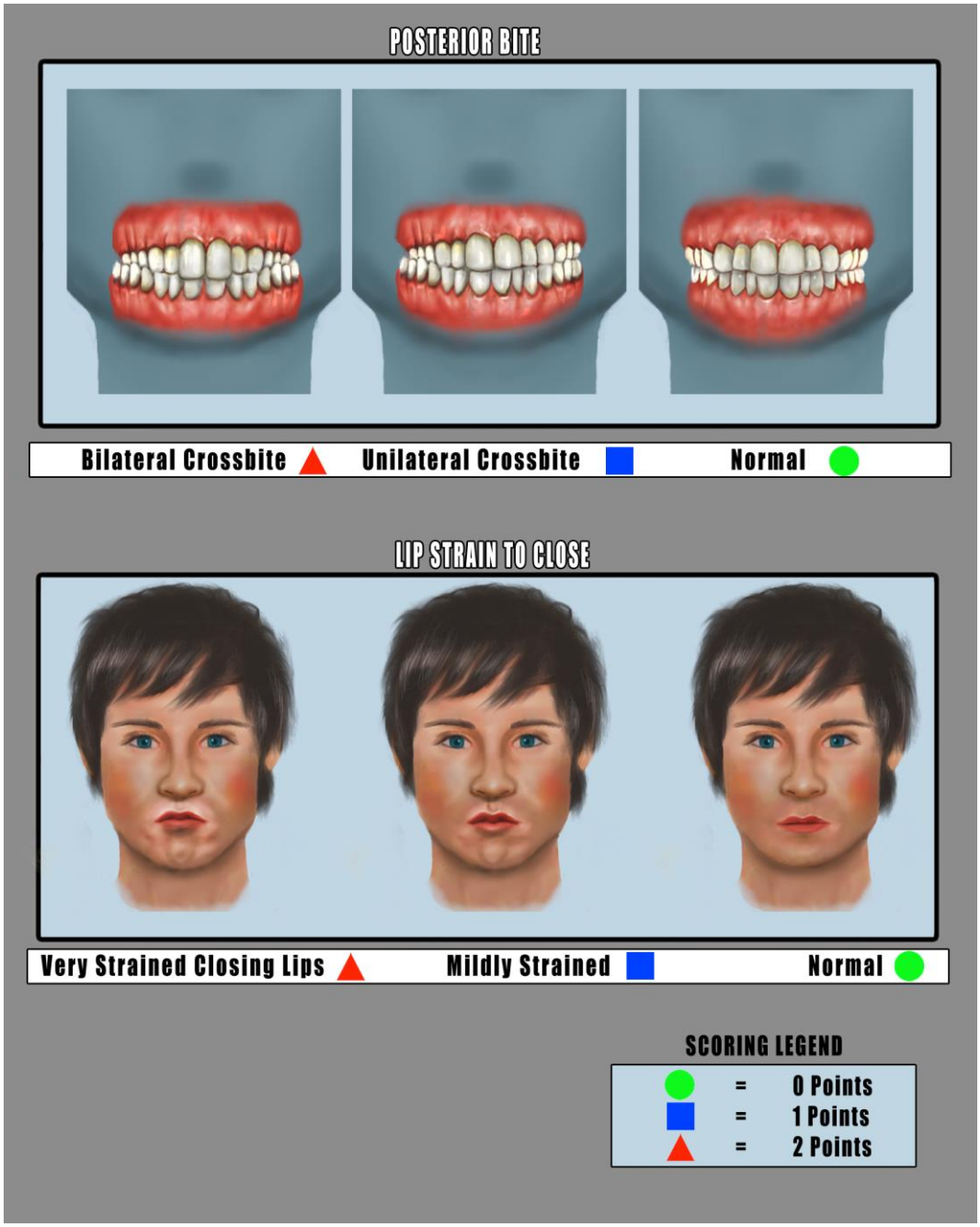
Severely Large Neck **Moderately Large Neck** **Mildly Large Neck** **Normal Neck** **Thin Neck**

Description: The circumference of the neck approximately below the thyroid cartilage

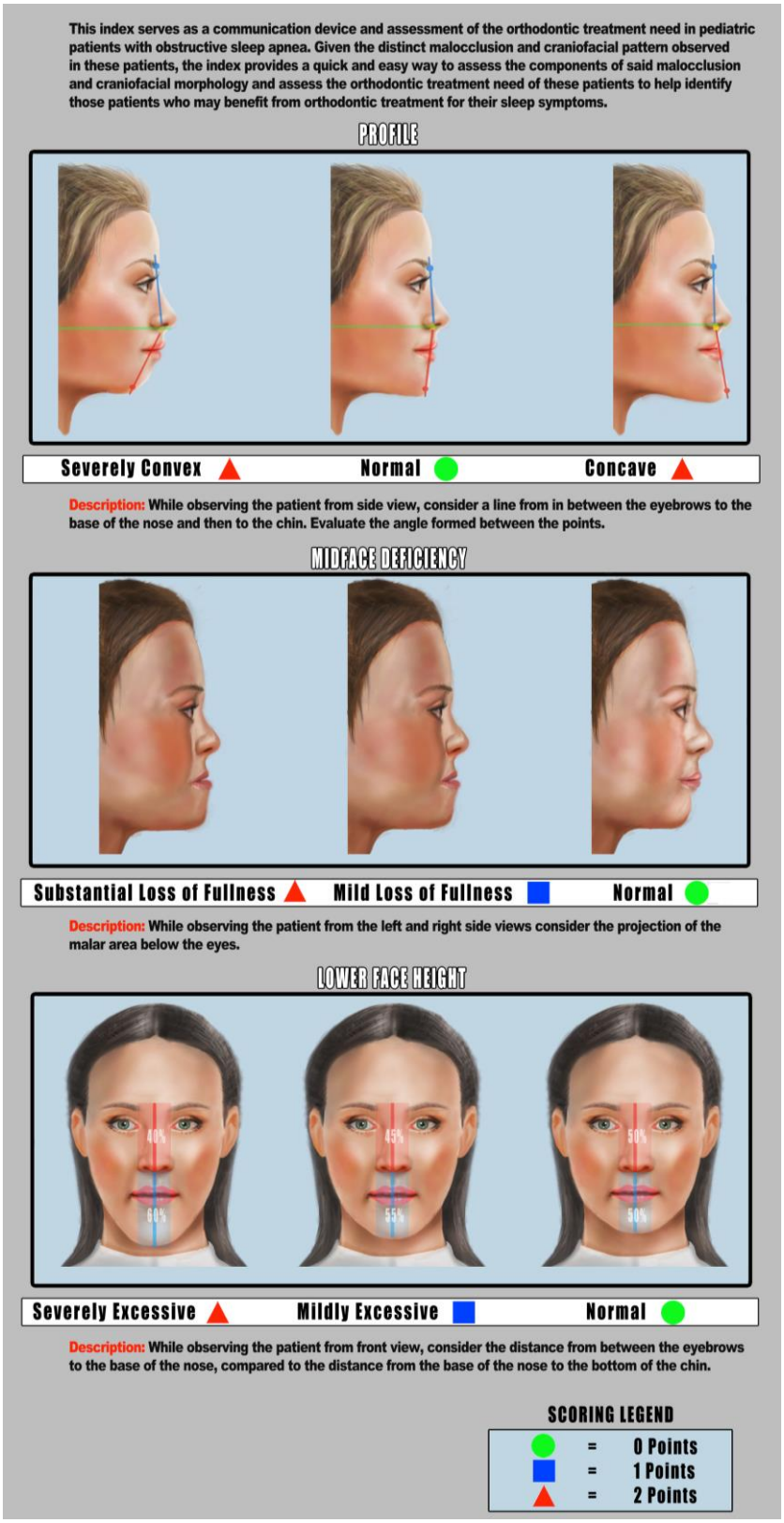
APPENDIX D


PROFILE
Severely Convex
Normal
Concave
MIDFACE DEFICIENCY
Substantial Loss of Fullness
Mild Loss of Fullness
Normal
LOWER FACE HEIGHT
40%
60%
45%
55%
50%
50%
Severely Excessive
Mildly Excessive
Normal
SCORING LEGEND
= 0 Points
= 1 Points
= 2 Points


PALATE
Severely High Arched
Mildly High Arched
Normal
OVERJET
Increased
Normal
Reverse
OVERBITE
Open Bite
Normal
Deepbite
SCORING LEGEND
= 0 Points
= 1 Points
= 2 Points


POSTERIOR BITE
Bilateral Crossbite
Unilateral Crossbite
Normal
LIP STRAIN TO CLOSE
Very Strained Closing Lips
Mildly Strained
Normal
SCORING LEGEND
= 0 Points
= 1 Points
= 2 Points

APPENDIX E


This index serves as a communication device and assessment of the orthodontic treatment need in pediatric patients with obstructive sleep apnea. Given the distinct malocclusion and craniofacial pattern observed in these patients, the index provides a quick and easy way to assess the components of said malocclusion and craniofacial morphology and assess the orthodontic treatment need of these patients to help identify those patients who may benefit from orthodontic treatment for their sleep symptoms.
PROFILE
Severely Convex
Normal
Concave
Description: While observing the patient from side view, consider a line from in between the eyebrows to the base of the nose and then to the chin. Evaluate the angle formed between the points.
MIDFACE DEFICIENCY
Substantial Loss of Fullness
Mild Loss of Fullness
Normal
Description: While observing the patient from the left and right side views consider the projection of the malar area below the eyes.
LOWER FACE HEIGHT
40%
60%
45%
55%
50%
50%
Severely Excessive
Mildly Excessive
Normal
Description: While observing the patient from front view, consider the distance from between the eyebrows to the base of the nose, compared to the distance from the base of the nose to the bottom of the chin.
SCORING LEGEND
= 0 Points
= 1 Points
= 2 Points

LIP STRAIN TO CLOSE

Very Strained Closing Lips ▲ Mildly Strained ■ Normal ●

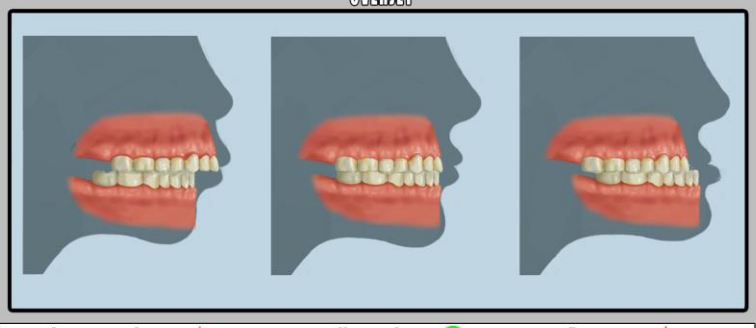
Description: While observing the patient from front view, and asking the patient to close their lips together, consider the amount of strain on the lip closing muscles around the mouth.

PALATE

Severely High Arched ▲ Mildly High Arched ■ Normal ●

Description: When viewing the palate inside the mouth, consider the depth of the palate, and the magnitude of the arch of the palate.

OVERJET



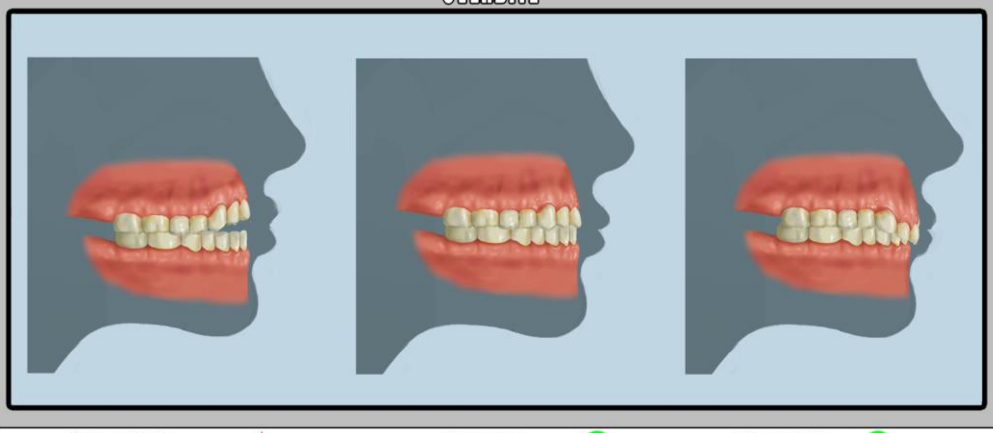
Increased ▲ Normal ● Reverse ▲

Description: Looking inside the mouth, this is the horizontal distance between the outside surface of upper and lower incisors. An excessive overjet is greater than 5mm.

SCORING LEGEND

●	=	0 Points
■	=	1 Points
▲	=	2 Points

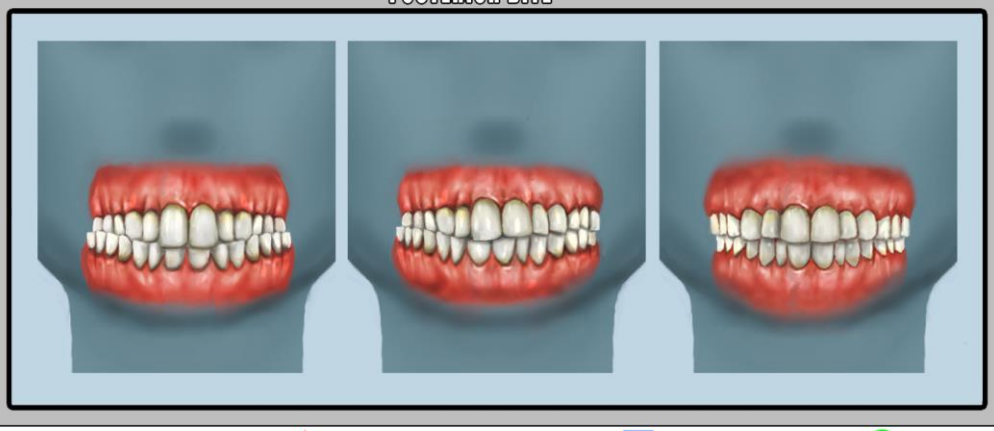
OVERBITE



Open Bite ▲ Normal ● Deepbite ●

Description: When looking inside the mouth, this is the vertical overlap between the upper and lower incisors.

POSTERIOR BITE



Bilateral Crossbite ▲ Unilateral Crossbite ■ Normal ●

Description: When looking this is the transverse relationship of the molars and premolars. When assessing this factor, consider the relationship of the upper posterior teeth to the lower ones on each side.

SCORING LEGEND

●	=	0 Points
■	=	1 Points
▲	=	2 Points