Université de Montréal

A prevalence study of dental malocclusions in children with sleep disorders

by
Jérémie Abikhzer

« Département de santé buccale – Section d'orthodontie »
« Faculté de Médecine Dentaire »

Thesis presented to the « Faculté des études supérieures »
to obtain a Master's degree (M.Sc.)
in dental medicine
orthodontic option

May, 2017

Résumé

Introduction : Les troubles respiratoires du sommeil (TRS) sont un continuum qui va du ronflement à l'apnée du sommeil. Le ronflement est un bruit à l'inspiration, causé par la vibration des tissus mous des voies aériennes supérieures détendus par le sommeil. Le syndrome d'apnée du sommeil est caractérisé par l'arrêt partiel ou complet du flot respiratoire de façon répétitive et transitoire durant le sommeil. Alors que l'hypertrophie des adénoïdes/amygdales est le facteur primaire contribuant aux TRS pédiatriques, il pourrait y avoir d'autres origines à l'obstruction tel que les malformations craniofaciales. Le but de cette étude de prévalence est de faire le compte du nombre de patients qui bénéficieraient d'une évaluation dentaire et orthodontique parmi ceux qui ont des troubles respiratoires de sommeil vus au CHU Sainte-Justine. Notre hypothèse de recherche est que la prévalence de malocclusions et d'anomalies dento-squelettiques serait différente entre les enfants apnéiques et non-apnéiques. **Méthodologie :** Lors de cette étude prospective multicentrique, les patients qui vont compléter un enregistrement de sommeil pour diagnostiquer les troubles respiratoires du sommeil au laboratoire de sommeil du CHU Sainte Justine seront contactés pour participer à cette étude de prévalence (n=100). L'évaluation dentaire se fera durant le rendez-vous. Le questionnaire de dépistage de Gozal et les données polysomnographiques, orthodontiques et craniofaciales seront étudiées. **Résultats :** Un total de 100 patients a été recruté (58 M, 42F). L'âge moyen des patients était de 9.6 ± 4.05 (3-18 ans). Les patients étaient divisés en groupes (n=57) IAH < 2, (n=43) IAH ≥ 2. Le groupe IAH < 2 avait une moyenne 0.79 ± 0.53. Le groupe IAH ≥ 2 avait une moyenne de 7.79 ± 8.03. Aucune différence n'a été trouvé entre les groupes IAH et le IMC (p=0.303). Par contre, le score de Gozal était significatif pour dépister des IAH plus sévères (p=0.011) pour un score ≥ 2.72. Aucune différence significative n'a été trouvée entre les amygdales hypertrophiques (score ≥3) et l'IAH (p=0.426). De plus, aucune différence significative n'a été trouvée entre les groupes IAH pour les caractéristiques craniofaciales et dentaires. Les patients ayant des habitudes orales (morsures des ongles/joues/ lèvres, bruxisme, succion du pouce) avaient une tendance d'avoir un IAH <2 (p = 0.064). La régression logistique a conclu que les garçons sont plus à risque (OR=3.52, 95%CI 1.27-9.77), ceux avec des habitudes orales sont moins à risque (OR=0.33, 95%CI 0.13-0.89) et que le risque d'avoir l'apnée augmente de 1.09 pour chaque unité d'accroissement d'IMC. **Conclusions :** La prévalence des malocclusions dentaires chez les enfants a été jugée non significative entre les groupes de différentes sévérité d'IAH. Aucune corrélation significative n'a été trouvée entre la morphologie craniofaciale et dentaire et les données sur le sommeil. Néanmoins, il s'agit d'une analyse préliminaire. L'objectif de cette étude multicentrique est de recruter jusqu'à 400 enfants et une analyse plus approfondie sera effectuée. D'autres études sont recommandées pour tirer de meilleures conclusions et améliorer le pouvoir statistique dans le rôle de la morphologie craniofaciale et dentaire chez les enfants avec des troubles respiratoires du sommeil.

Mots-clés : troubles respiratoires du sommeil, apnée obstructive du sommeil, enfants, prévalence, craniofacial, malocclusion dentaire, polysomnographie

Abstract

Introduction: Sleep-disordered breathing (SDB) is a continuum that ranges from snoring to sleep apnea. SDB occurs in children of all ages, from neonates to adolescents, and it is characterized by repeated events of snoring, and either partial (i.e. hypopnea) or complete (i.e. apnea) upper airway obstruction during sleep. While hypertrophy of the adenoids / tonsils is the primary factor contributing to pediatric SDB, there may be other origins to obstruction such as craniofacial malformations. The purpose of this prevalence study is to count the number of patients who would benefit from a dental and orthodontic assessment among those with sleeping breathing problems seen at the CHU Sainte-Justine. Our research hypothesis is that the prevalence of malocclusions and dento-skeletal abnormalities would be different between apneic and non-apneic children. **Methods:** In this prospective multicenter study, patients who will complete type 1 polysomnography to diagnose sleep disorders at the CHU Sainte Justine will be contacted to participate in this prevalence study (n=100). Dental and orthodontic evaluation will be done during the appointment. Gozal screening questionnaire, polysomnographic, orthodontic and craniofacial data will be studied. **RESULTS:** A total of 100 patients were recruited (58 M, 42 F). The mean age of the patients was 9.6 ± 4.05 (3-18 years). Patients were divided into groups (n = 57) AHI <2, (n = 43) AHI ≥ 2. The AHI <2 group had a mean AHI of 0.79 ± 0.53. The AHI ≥ 2 group had a mean AHI of 7.79 ± 8.03. No difference was found between AHI groups and BMI (p = 0.303). On the other hand, Gozal score was significant for detecting more severe AHI's (p = 0.011) for a severity score ≥ 2.72. No significant difference was found between hypertrophic tonsils (score ≥3) and AHI (p = 0.426). In addition, no significant difference was found between AHI groups for craniofacial and dental characteristics. Patients with oral habits (nail/cheek/lip biting, bruxism, thumb sucking) tended to have an AHI <2 (p = 0.064). Logistic regression calculations concluded that boys are at higher risk (OR = 3.52, 95% CI 1.27-9.77), those with oral habits are less at risk (OR = 0.33, 95% CI 0.13-0.89) and that odds of having apnea increases by 1.09 for each unit of BMI increase. **CONCLUSIONS:** The prevalence of dental malocclusions in children was found to be insignificant among groups of different AHI severity. No significant correlation was found between craniofacial and dental morphology and sleep data. Nevertheless, this is a preliminary analysis. The objective of this multi-center study is to recruit up to 400 children and further analysis will be carried out. Further studies are recommended to draw better conclusions and improve statistical power in the role of craniofacial and dental morphology in children with sleep disorders.

Keywords: sleep-disordered breathing, obstructive sleep apnea, children, prevalence, craniofacial, dental malocclusions, polysomnography

Table of Contents

Table List

Figures List

Abreviations List

OSAS: obstructive sleep apnea syndrome

OSA: obstructive sleep apnea

SDB: sleep-disordered breathing

CSA: central sleep apnea

AAP: American Academy of Pediatrics

PSG: polysomnography

UARS: upper airway resistance syndrome

PS: primary snoring

RERA: respiratory effort-related arousals

AHI: apnea-hypopnea index

T&A: tonsils & adenoids

CPAP: continuous positive airway pressure

NEPAP: nasal CPAP

RME: rapid maxillary expansion

RDI: respiratory disturbance index

EEG: electroencephalogram

ADHD: attention-deficit hyperactivity disorder

DISE: drug-induced sleep endoscopy

UPPP: uvulopalatopharyngoplasty

MRI: magnetic resonance imaging

REM: rapid-eye movement

SB: sleep bruxism

RMMA: rhythmic masticatory muscle activity

To my wife, parents, siblings,
Thank you for all your support &
encouragement,
I love you all.

Acknowledgements

I would like to thank my research team for guiding and assisting me throughout my research project. Thank you, Dr. Nelly Huynh, for inspiring me in this field since the beginning of my undergraduate studies. I have learned so much throughout this whole journey and enjoyed every step of it. To my co-director, Dr. Andrée Montpetit, thank you for your advice in both the research and clinical aspects. Thanks to Dr. Sophia Laberge, Dr. Sheila Jacobs, and Sylvie Laporte for making this research possible. Finally, I would like to thank all my other team members for their valuable and appreciated help in this project: Dr. Julia Cohen-Levy and Dr. Mathieu Laramée.

I would also like to thank Mr. Pierre Rompré for his statistical expertise all along my research project. Thank you for your patience. Thank you, Dr. Gilles Lavigne, for accepting to preside my thesis; as well as Dr. Audrey Bellerive for taking the time to review my thesis and for all your valuable comments.

I would like to thank my classmates, Mélanie, Charles, Julien as well as all my other co-residents for making this whole journey so much more enjoyable.

Finally, I would like to show gratitude to our department chair, Dr. Claude Remise, as well as all the professors, clinicians, and staff of the department of Orthodontics for your expertise, teachings and your support throughout these three beautiful years. Your guidance and teachings will guide me throughout my career.

Chapter 1. Introduction

1.1 Sleep

Sleep is a universal biological procedure necessary in maintaining health. Sleep is defined by a physiological state of partial isolation from the environment. The average amount of sleep is between 6-9 hours for an adult and is more variable in children depending on age. Sleep is often described as recuperating when it is continuous and not disturbed. Sleep plays multiple functions: fatigue recuperation, biochemical functioning, immune function aid, memory and well-being. Its role is especially physiological in children. Development of a good night's sleep in children is critical for proper growth mainly because growth hormone is secreted at its peak during nighttime.(1)

1.2 History

Sleep apnea research became much more regular in the 1950s. Around then, sleep apnea has been officially termed as a disorder. Common sleep apnea symptoms were called "Pickwickian syndrome" until the late 19th century originating from Charles Dickens literary contributions "The Pickwick Papers" description of "Fat Joe".(2)

1.3 Obstructive sleep apnea syndrome (OSAS)

Sleep-disordered breathing (SDB) is described by an abnormal respiratory pattern during sleep. It comprises of snoring, mouth breathing, and pauses in breathing.(3) SDB is a continuum that ranges from snoring to sleep apnea. Breathing during sleep can be compromised by increased resistance in the upper airway or partial to complete collapse of the airway. Snoring is a sound at the inspiration caused by upper airway soft tissue vibration

relaxed by sleep. SDB occurs in children of all ages, from neonates to adolescents, and it is characterized by repeated events of snoring, and either partial (i.e. hypopnea) or complete (i.e. apnea) upper airway obstruction during sleep.

The Internal Classification of Sleep Disorders classifies SDB into five principal categories, two of which, are OSA and central sleep apnea (CSA). CSA is characterized by repeated episodes of absence or diminution of respiratory effort due to primary idiopathic reasons or secondary to a pathology.(1) The American Academy of Pediatrics (AAP) defines childhood OSA as a disorder in breathing during sleep with prolonged partial upper airway obstruction and/or intermittent complete obstruction with its associated signs and symptoms. However, the American Academy of Otolaryngology Head and Neck Surgery defines childhood OSAS when clinically SDB is supported by an abnormal polysomnography (PSG) with obstructive events before tonsillectomy.(3)

Chapter 2. Literature Review

2.1 Prevalence

The prevalence of habitual snoring in children, which is considered pathological, is currently estimated as high as 27% (4-6) and approximately 2% to 3% of children have clinical relevant sleep apnea.(7) Approximately 20% of snoring children who would undergo polysomnography would be diagnosed with OSA.(8) According to Marcus et al., prevalence of childhood OSAS can range between 1.2%-5.7% (9) and between 1%-10% according to Alexander et al.(3) Huynh et al. have reported primary snoring in children to be between 3.1%-12.1% and of OSAS to be between 0.7%-10.3%.(10) The peak incidence of pediatric OSAS is between 2 to 8 years old.(11)

2.1.1 Sex

Males have been shown to have a predominant ratio of 2:1 to females in adult sleep apnea. However, recent pediatric studies studying gender differences are limited and the results are inconclusive. In a review conducted by Lumeng et al. on gender differences in pediatric sleep apnea, fifteen studies showed a male predominance while 19 studies showed no sex difference. However, population samples were significantly higher in the studies who showed a higher prevalence in boys.(4) Only one study shows a higher girl predominance.(12) Gender differences become clearer when children enter puberty where hormonal differences play a role. Clearly, pubertal hormonal and physiologic changes potentiate the outcome of sex difference among many factors in SDB prevalence.(4)

2.1.2 Age

Numerous papers studied within their own population pool SDB variations with age. Most studies have shown no difference in age windows with parental-reported SDB symptoms. Only four studies have demonstrated an age difference in children with SDB.(4) One of those showed a significant decrease in parent-reported snoring between 4-12 years old.(13) Another study reported no statistically significant increase in snoring prevalence between 9-15 years old, but a marked age prevalence was seen after 15 years old.(14) Moreover, Ersu's study reported a higher snoring prevalence amid 5-8 years old, then a prevalence decrease in 9-10 years old, and once again an increase with pubertal changes at around 11-13 years old.(15) However, data in children is insufficient to attest an SDB prevalence which differs analytically by age. Also, since parent-reported snoring alone has been used to screen children for PSG, additional underestimation of OSA prevalence is possible. Parent-reported snoring may be useful but not sufficient enough to differentiate pediatric primary snoring from OSA, and therefore further diagnostic tools such as PSG are recommended.(4)

2.1.3 Race

Race differences in pediatric SDB have also been reported controversial. African American's have shown a higher potential association between race and prevalence of SDB amongst children in comparison to Caucasian's.(16, 17) Also, subjectively, Hispanic parents have reported more SDB symptoms than Caucasian parents.(18) Yet, more extensive research is needed to come to better conclusions in regards to race differences in SDB prevalence.

2.2 Signs & Symptoms

Pediatric SDB has been associated with numerous daytime and nighttime symptoms which vary by age (Table 1). Daytime symptoms are generally seen in older children while nighttime symptoms are reported by parents instigating an initial consultation. Snoring is the most reported symptom in children.(3)

Table 1: Sign & Symptoms of SDB in children(10)

Table I. Symptoms of sleep-disordered breathing in children and adolescents[6-8,12,13]

Nighttime	*Daytime*
• Abnormal sleeping positions	• Morning tension-type headache
• Chronic, heavy snoring	• Mouth breathing
• Confused arousal	• Excessive morning thirst
• Delayed sleep onset	• Excessive fatigue and sleepiness
• Difficulty breathing during sleep	• Abnormal shyness, withdrawn and depressive presentation
• Difficulty waking up in the morning	• Behavioral problems
• Drooling	• Pattern of attention-deficit/ hyperactivity disorder (ADHD)
• Enuresis	• Aggressiveness
• Frequent awakenings	• Irritability
• Insomnia	• Poor concentration
• Mouth breathing	• Learning difficulties
• Nocturnal migraine	• Memory impairment
• Nocturnal sweating	• Poor academic performance
• Periodic limb movement	
• Restless sleep	
• Sleep talking	
• Sleep terror	
• Sleepwalking	
• Witnessed breathing pauses during sleep	

(Table adapted from Huynh et al. Associations between sleep-disordered breathing symptoms and facial and dental morphometry, assessed with screening examinations. 2011)

2.3 Diagnosis

SDB is primarily diagnosed by a clinical perspective. Presence of common relevant clinical observations such as chronic snoring, excessive fatigue and sleepiness, attention-deficit hyperactivity disorder, and learning difficulties may be helpful in guiding the clinician. SDB ranges from primary snoring (PS) to upper airway resistance syndrome (UARS) to obstructive sleep apnea (OSA). PS is defined by snoring without apneas, arousals on polysomnography (PSG) and gas exchange abnormalities. UARS is described by snoring with repetitive cycles of respiratory effort-related arousals (RERAs) without oxygen desaturation. UARS day-time symptoms can often resemble those of OSAS. UARS is diagnosed through an esophageal pressure monitor during overnight PSG (Figure 1).(3)

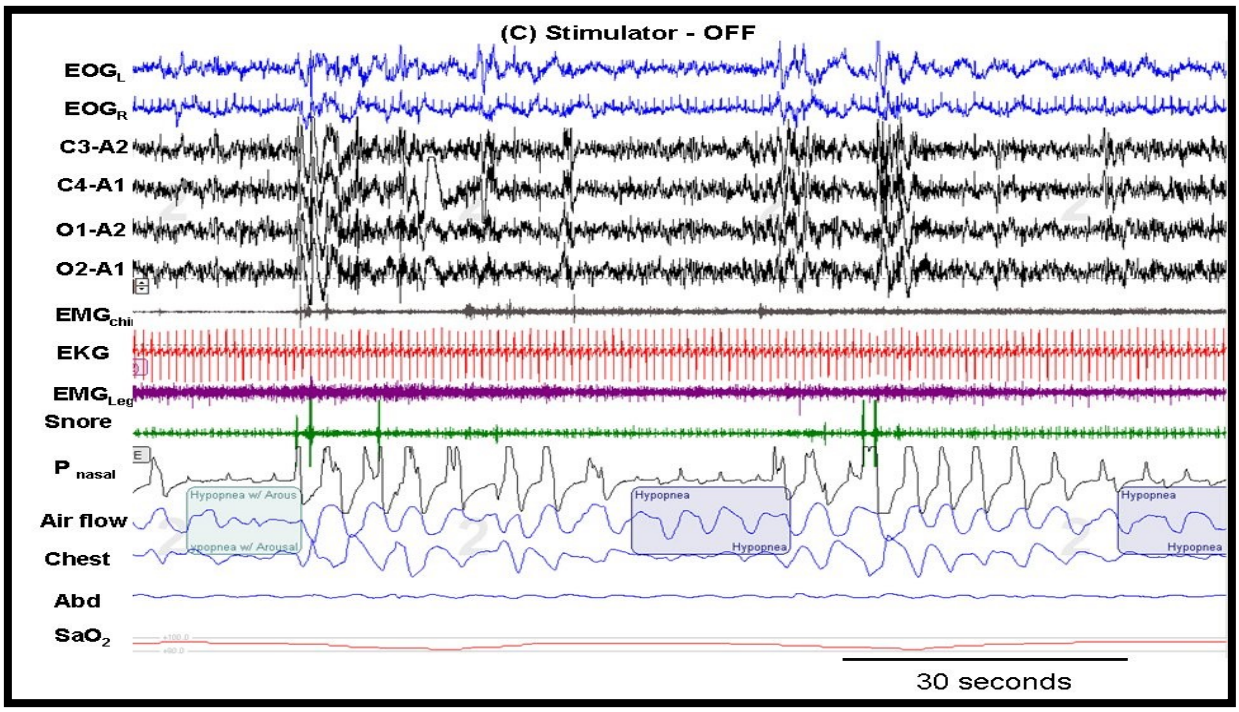


Figure 1: Polysomnography(19)

(Figure adapted from: https://www.thoracic.org/professionals/clinical-resources/sleep/sleep-fragments/images/slide3.jpg)

Per the 2012 AAP updated guidelines, PSG is the gold standard tool to diagnose childhood OSAS. However, due to different limitations, other accessory tools which come in aid to clinical evaluation are used today such as sleep videotaping, daytime nap PSG and nocturnal pulse oximetry. The limits of those tools include poorer sensitivity. PSG became the gold standard in diagnosing childhood OSAS due to poor sensitivity in differentiating OSAS and PS by clinical and physical evaluation alone (Figure 2).(3)

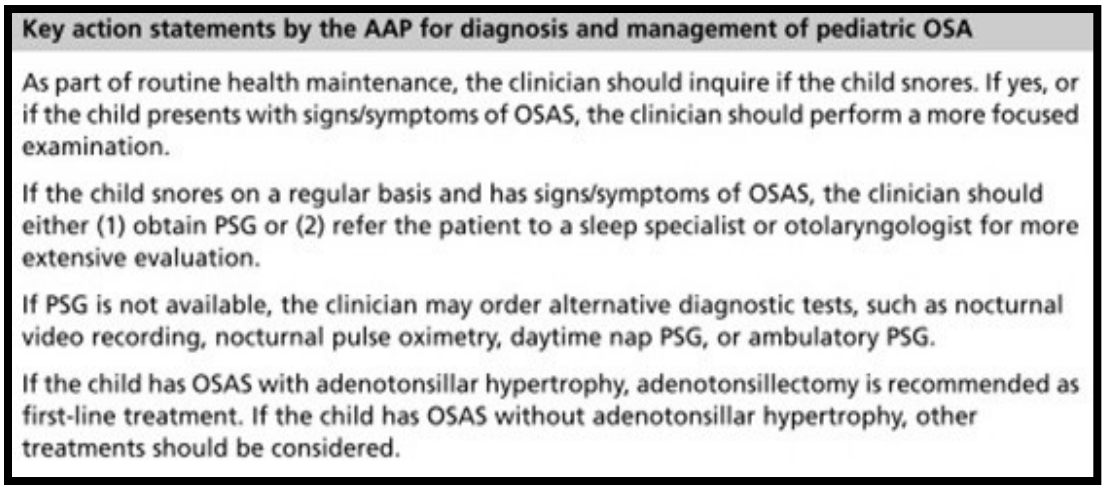


Figure 2: Diagnosis & management of pediatric OSA(3)
(Figure adapted from: Alexander et al. Pediatric obstructive sleep apnea syndrome. 2013)

Childhood OSAS diagnosis is different than adult OSAS. In adult OSAS, apnea is defined as a 10 second or more respiratory pause. In children, shorter respiratory pauses are clinically significant. Childhood apnea is defined by a complete air flow interruption of at least 2 breath periods, and hypopnea is defined by a 50% air flow reduction associated with awakening, arousal or desaturation of 3% or more for the same period.(3) Furthermore, apnea-hypopnea index (AHI) and minimum oxygen saturation readings differ between children and

adults. The values for minimum oxygen saturation readings for children are >1 and <92 respectively. For adults, those values are >5 and <85 respectively.(20)

Pediatric OSA polysomnographic interpretations and performance have not been well defined. There exists controversy between different academies. The 2007 American Academy of Sleep Medicine guidelines defines as abnormal any of those signs; an AHI of 1 or more per hour, common arousals from sleep associated with increased respiratory effort, and arterial oxygen desaturation in association with apneic episodes. Abnormal AHI recordings also differ between adults and children (Table 2). Several studies consider an AHI of 1 or more as abnormal while some set the threshold at an AHI of 5 and more.(3)

Table 2: AHI severity for children & adults

	Children	Adults
Mild	1-5	5-15
Moderate	5-10	15-30
Severe	10+	30+

2.3.1 Pediatric sleep questionnaires

Questionnaires have been used in most domains as a predictive and clinically useful tool for both the parent and clinician. Sleep questionnaires have been useful in sleep research since the late 1980s. Due to their increasing popularity, sleep questionnaires targeting pediatric

SDB have greatly increased and their heterogeneity has led to the need of better tools for more accurate diagnosis.(21) Therefore, Spruyt and Gozal have reviewed an extensive list of published and unpublished instruments and came up with a standardized 11 step instrument development tool based on proper psychometric norms.(22) In a recent publication, they arranged a set of six ordered questions from a wide-ranging list of questions that permits reasonable discernment along the SDB spectrum. Its high negative predictive value suggests that it will rarely misclassify a child with SDB as not presenting with SDB. It was also found that parent-reported snoring was found to be a relevant discriminant symptom factor in sleep questionnaires for screening apneic vs non-apneic snorers. The six questions used in our study are based on these subjective respiratory symptoms. Refer to annex 3 for full detail of sleep questionnaire.(23-26)

2.4 Treatment

2.4.1 Adenotonsillectomy (T&A)

The first-line and most common medical procedure for SDB in children is T&A (Figure 4). In the United States alone, 530,000 tonsillectomies are performed annually on children. The other frequent indication of T&A is for recurrent throat infections.(27) Anatomically, tonsils and adenoids occupy a large volume in the respiratory airways due to their frequent hypertrophy in children.(3) A meta-analysis conducted by Brietzke and Gallagher studied PSG data pre and post-T&A in children. The overall treatment success was found to be 82% with an average AHI reduction of 13 events/hr.(28) This data is more representative in a healthy non-obese children population. According to Friedman,

improvement in SDB after T&A is correlated to the degree of obesity.(29) A meta-analysis of 4 studies show an improvement of 10-25% in SDB after T&A in obese children.(30)

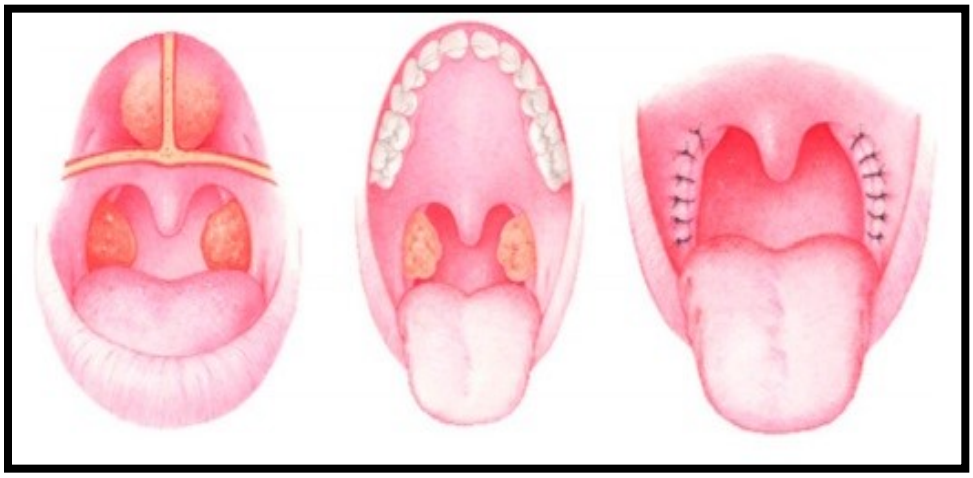


Figure 4: Adenotonsillectomy(31)
(Figure adapted from: Won et al. Surgical treatment of obstructive sleep apnea: upper airway and maxillomandibular surgery. 2008)

2.4.2 Continuous positive airway pressure (CPAP)

Although less successful, other adjunct medical therapies can be offered to children for SDB treatment. CPAP is considered the first-line treatment in adults. However, due to a higher success rate of surgery in pediatric OSA, CPAP is the second-line treatment in children (Figure 5). Home nasal CPAPs (NEPAP) are the most common CPAPs used in pediatrics.(32) A pilot study done by Kureshi et al. showed a 64% AHI improvement in children aged 8-16 years old. However, an improvement was not seen in 21% of children, and the AHI worsened in 14% of children. Better results were seen in older children and those with less hypercapnia. NEPAPs is therefore a potential alternative therapy in pediatric OSA when efficiency is confirmed with a polysomnographic study.(33) Family training with NEPAPs as well as

appropriate nasal cannula usage is important to prevent any craniofacial disturbance growth. Regular follow-ups can prevent complications such as local discomfort, skin ulceration, eye irritation as well as conjunctivitis.(34)

Figure 5: CPAP(35)

(Figure adapted from: http://sleepapneadisorder.info/wp-content/uploads/2012/05/sleep-apnea-treatment-in-children.jpg)

2.4.3 Rapid maxillary expansion (RME)

Numerous studies have examined the effects of RME in pediatric OSA in children who present that indication (Figure 6).(10, 11, 36-40) The benefits of this treatment will be further developed below in the craniofacial anatomy section.

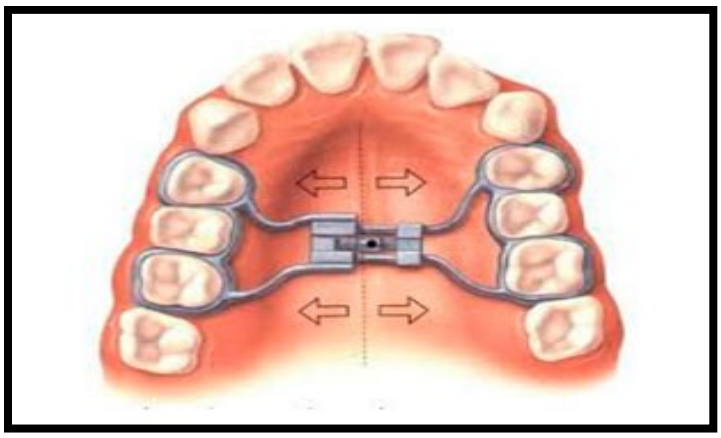


Figure 6 : RME(41)

(Figure adapted from: http://facialsurg.cc/images/expander.jpg?)

2.4.4 Weight loss

Other adjunct therapies can include weight loss in obese children. Only a couple studies show an improvement in pediatric OSA with weight loss but the degree of weight loss required in children has not been well studied. Weight loss and OSAS are more frequently studied in the adult population.(3) One of them, conducted by Verhulst et al. on 21 obese teenagers in a residential facility showed that with a median weight loss of 24 kg (11–48), the AHI decreased from 3.8 (2.2–58.3)/hour to 1.9 (0.6–27.7)/hour ($p = 0.002$). The authors calculated a significant decrease in incidence of moderate to severe OSAS from 33 to 9% ($p = 0.05$).(42) Furthermore, Kalra et al. studied the effect of bariatric surgery on 10 obese adolescents presenting with OSAS. Over a 5-month period and a mean weight loss of 58 kg, the mean AHI decreased from 9.2/hour pre-surgery to 0.65/hour post-surgery ($p<0.01$). However, to this date, the outcome of weight loss is unknown due to poor sample studies and loss of follow-ups.(43)

2.4.5 Intra-nasal steroids

Intra-nasal steroids have also been used for treating milder cases of pediatric OSAS. For example, a study done by Kheirandish-Gozal demonstrated that a budesonide protocol treatment in 48 children reduced their mean AHI from 3.7 ± 0.3 to 1.3 ± 0.2 ($p < 0.001$). Furthermore, the beneficial aspect of the medication persisted for at least 2 months post-treatment.(44) Others have shown that intra-nasal steroids such as prednisone or fluticasone could be helpful in pediatric OSA. In a case-control study, the treatment group showed a AHI value decrease from 10.7 ± 2.6/hour to 5.8 ± 2.2/hour ($p = 0.03$). Practically, intra-nasal steroids have been used in various clinical trials in children and are mostly indicated as second-resort treatment in children with mild OSAS.(11)

2.4.6 Body position

The role of body position on OSAS has not been well studied in children. Four conflicting studies have been analyzed based on retrospective sleep laboratories data correlating spontaneous body positioning with OSAS. The first study conducted by Pereira et al. showed an elevated respiratory disturbance index (RDI) in children younger than 3 years old sleeping in the supine position.(45) Another study conducted by the same research team showed no correlation between body position and OSAS in infants aged 8-12 months.(46) On the other hand, Fernandes do Prado et al. concluded that children aged 1-10 years old breathed better in a supine position.(47) Lastly, Dayyat et al. showed that although AHI was greater in children in the supine position, no significant AHI differences were found between body position and sleep. Further prospective studies are needed to evaluate appropriately body position with sleep in children.(48)

2.4.7 Myofunctional therapy

Current literature demonstrates the benefits of myofunctional therapy as an adjunct therapy in pediatric OSAS before and after T&A. It has been estimated to reduce AHI by 62% in children.(49) A study done by Villa et al. reported a 62% reduction in AHI between the experimental and control group in which 14 post-T&A children were randomly assigned to either a 2 month oropharyngeal exercise group or the control group.(50) Another study by Guilleminault et al. showed no recurrence of OSA in children 4 years post-T&A and maxillary expansion in combination with myofunctional therapy in comparison to a control group in which OSA relapsed.(51)

A recent meta-analysis by Huynh et al. reviewed the role of orthodontic treatment on obstructive sleep apnea management in children. Both RME and myofunctional mandibular advancement appliances were studied. Bearing in mind that only eight studies were deemed to fit all inclusion criteria, RME as well as myofunctional therapy may be effective in pediatric sleep apnea management. The authors conclude by stating that although orthodontic treatments may correct craniofacial morphology, a possible risk factor in pediatric SDB, one should be cautious in interpreting those results due to the lack in quantity and quality of studies.(38)

2.5 Morbidity

Although still not fully understood, the pathophysiology of OSA in children is multifactorial. Hypertrophic tonsils and adenoids are the principal cause of this condition.(8) Nevertheless, isolated adenotonsillar hypertrophy cannot be solely blamed as some children

develop OSA after adenotonsillectomy and some with enlarged tonsils and adenoids do not present any symptoms of OSA.(52)

The consequences of pediatric OSA can affect multiple physiological systems. An overview of the different systems will be expanded below. These consequences could have a direct and indirect impact on the quality of life and daily wellbeing of the patient as well as his entourage.

2.5.1 Increased nocturnal respiratory effort

Increased energy expenditure is a common side effect in children with OSA due to an increased respiratory effort. Other potential co-morbidities include dysphagia due to adenotonsillar hypertrophy and reduced tissue and systemic levels of insulin growth factor-1.(53) No evidence of irreversible somatic side effects has been proven once SDB is resolved in children.(8)

2.5.2 Intermittent hypoxemia

Persistent pulmonary hypertension, a major consequence of pediatric OSA, may arise from an increase in pulmonary artery pressure due to hypoxia-induced increases in pulmonary vasomotor contractility.(54) Intermittent hypoxia could also affect left ventricular function.(55) Although cardiovascular morbidity data is still limited in pediatric OSAS, studies have reported an increase of arterial blood pressure due to augmentation of sympathetic activity. This could induce permanent changes in the physical properties of blood vessels and therefore elevate blood pressure.(56)

2.5.3 Sleep fragmentation

Unlike adults, children with OSA experience sleep fragmentation instead of sleep deprivation. Sleep deprivation is defined as the occurrence of many arousals and awakenings at night in correlation with obstructive respiratory events. The consequences of sleep fragmentation are not known to this date.(8) Furthermore, unlike adults, assessment by parental questionnaires or a multiple sleep latency test show that children with OSA do not generally display excessive daytime sleepiness.(57) Therefore, we can generally assume that sleep fragmentation related-morbidity is of a lesser consequence in children since sleep architecture is slightly modified.(58) The main reason for this phenomenon is that children have less electroencephalogram (EEG) arousals than adults, and can therefore maintain and preserve sleep architecture better.(59) However, sleep pressure is increased and consequently leads to decreased arousability in children and therefore an increased frequency of nocturnal enuresis.(60)

2.5.4 Learning & behavior

A clear relationship between school performance and children with OSA has been well established.(61) Poor school performance in those children can translate into restlessness, hyperactivity, aggressive behavior and poor test performance. Poor memory and learning have also been shown to be directly correlated with children suffering of OSAS. However, considerable improvement in school performance and behavior has been documented in children post-treatment. This suggestive data demonstrates that neurocognitive side effects are partially reversible.(62) A cohort study conducted by Gozal et al. found that in the lowest 10^{th} percentile performing 1^{st} graders, a 6-9 fold increase in OSA incidence was shown. Furthermore, in children accepting T&A, significant improvement was demonstrated while

those refusing treatment continued to perform poorly.(25) However, this needs to be taken into consideration since we cannot know for certain the actual learning potential of the child post-treatment and as a result cannot determine at which extent the reversibility of learning performance occurred (i.e. partial or complete).

There also exists a clear relationship between OSA and attention-deficit-hyperactivity disorder (ADHD) in children. Furthermore, up to 30% of children who display signs of frequent and loud snoring can show signs of hyperactivity and inattention.(63) PSG readings also show different sleep architecture in children with ADHD in comparison to healthy subjects.(64)

2.5.5 Alveolar hypoventilation

Alveolar hypoventilation is the result of continued upper airway resistance with diminished compensatory responses during sleep in children with OSA. It is developed by repeated periods of CO_2 elevations and increased upper airway resistance during sleep.(3)

2.6 Untreated OSAS in children

The natural history of OSA in children can be variable mainly due to puberty and airway volume growth. A survey study conducted by Anuntaseree et al. in 1008 7-year-old children followed over a 3-year period concluded that 65% of habitual snorers did not continue snoring as they got older. However, 9% of children had developed OSAS with time. Deferment of treatment could have negative consequences, especially in children who have been diagnosed with mild OSAS.(65) Another study reported that no differences were found between healthy controls and children with primary snoring with a 3-year follow-up PSG. The authors conclude that it could be safe to defer treatment in children with primary snoring.(66,

67) However, it must be noted that these 13 children were diagnosed with primary snoring, an early sign of OSA, and not OSAS.(66) Li et al. identified key factors associated with OSAS development in children if left untreated; boys, especially those presenting with tonsillar hypertrophy and obesity.(68) Lastly, the Childhood Adenotonsillectomy study suggests that children with mild to moderate OSA show a complete resolution with watchful waiting in patients with smaller waist circumferences.(69) When comparing watchful waiting to T&A in school-aged children, surgical treatment did not significantly improve attention in children measured with neuropsychological testing. However, it positively affected quality of life, polysomnographic findings, and reduced signs & symptoms. Polysomnographic data was normalized in 79% of the surgical treatment group, whereas the randomly assigned watchful waiting group showed 46% normalization in polysomnographic findings for reasons such as development of airway or regression of lymphoid tissue, regression to the mean and routine medical visits. Changes were more impressive in the more severe apneic children with surgical treatment. (70) In conclusion, observational waiting may be an acceptable management plan in children with mild OSA under few conditions: older age, normal airway volume and non-obese children.

2.7 Persistent OSAS

Numerous studies have shown a prevalence of up to 50% of children presenting with persistent OSAS after T&A confirmed by PSG.(71-73) Properly identifying persistent OSAS post-T&A is of outmost importance because of its high frequency. Tagaya et al. examined the risk factors in persistent OSAS 1.5 years post-T&A in 49 normal-weight children with up to three PSGs per child. 27% of children were still symptomatic at the 1.5-year follow-up. The principal risk factor in persistent OSAS after T&A was found to be obesity.(30) Symptomatic

children commonly presented with allergic rhinitis and allergies ($p<0.05$). In both the symptomatic and asymptomatic groups, the following factors were both prevalent: atopic dermatitis, bronchial asthma food allergy, family history of OSAS, nocturnal enuresis, otitis media with effusion and sinusitis**.** Adenoid regrowth was found in 12% of children and all of them were still symptomatic post-T&A.(74)

When children present with persistent OSAS post-T&A, drug-induced sleep endoscopy (DISE) is a powerful diagnostic tool in analyzing remaining partial or complete sites of obstruction (nasal cavity, tongue base, velum/palate, oropharyngeal walls, epiglottis) in the airway system. DISE examines different levels of upper airway during spontaneous ventilation with the patient induced pharmacologically into an unconscious sedation simulating sleep. A retrospective study done by Truong et al. showed that DISE was an effective diagnostic tool for evaluating remaining sites of obstruction in children with persistent OSAS after T&A. Lingual tonsillar hypertrophy as well as laryngomalacia are possible remaining obstruction sites in persistent pediatric OSAS identified by DISE.(75)

Additional surgical therapies exist in children with persistent OSAS after T&A. Uvulopalatopharyngoplasty (UPPP) is a common secondary surgical option in children with persistent OSAS (Figure 7).

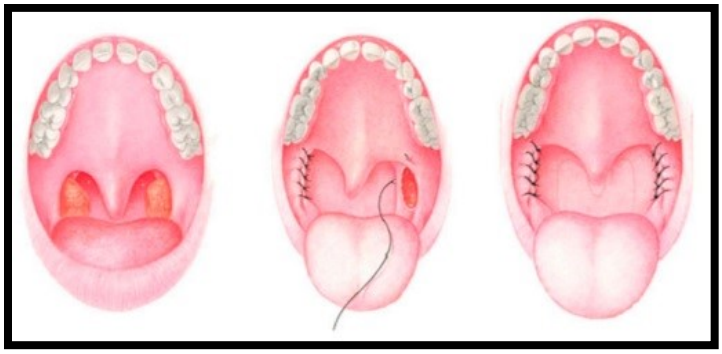


Figure 7: UPPP(31)

(Figure adapted from: Won et al. Surgical treatment of obstructive sleep apnea: upper airway and maxillomandibular surgery. 2008)

Furthermore, tongue base obstruction may be resolved with genioglossal advancement or lingual tonsillectomy, these with different varying degrees of success (Figure 8). (76) RME in pre-pubertal children has also shown great benefits in reducing AHI events. According to a study by Villa et al., 10 out of 14 children greatly benefited from a RME showing an AHI reduction, a decrease of daytime symptoms and snoring. The same results were also conclusive after a 24-month follow-up.(40)

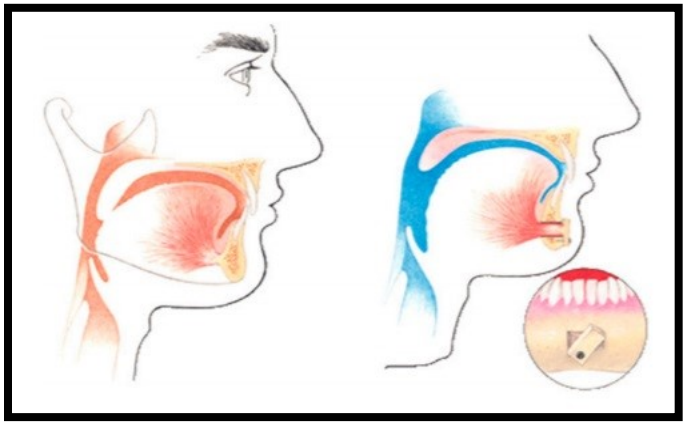


Figure 8: Genioglossus advancement(31)

(Figure adapted from: Won et al. Surgical treatment of obstructive sleep apnea: upper airway and maxillomandibular surgery. 2008)

2.8 Craniofacial anatomy

Craniofacial abnormalities are a frequent assessment in children with OSA due to its impingement on upper airway dimensions. Today, craniofacial morphology analysis is becoming significantly more important in the diagnosis and treatment planning in pediatric OSAS.(77)

The principal risk factor in pediatric OSAS is adenotonsillar hypertrophy. Kang et al. examined the effect of adenotonsillar size and AHI in pediatric sleep apnea in 495 children. Brodsky's scheme was used to evaluate tonsil size (Figure 9). A positive correlation was found between tonsil size and AHI in all different age groups (toddler, preschool, school and adolescents). However, adenoid size and AHI was positively associated in all groups except in the adolescent group. The following is consistent with normal adenoid growth pattern i.e. adenoid size decrease in adolescence.(78) At age 4, the adenoidal-nasopharyngeal space is the narrowest. Between ages 7 to 10, the face grows quickly and the space reaches its maximum volume. The space then continues to progressively decline until the age of 12 and decreases abruptly from 12 to 15 years old.(79) However, tonsil size is still prominent in both children and adolescents and is conclusive with Kang's findings. Furthermore, the additive effects of both adenoid and tonsillar hypertrophy increase pediatric symptoms more than adenoidal hypertrophy or tonsillar hypertrophy only.(78)

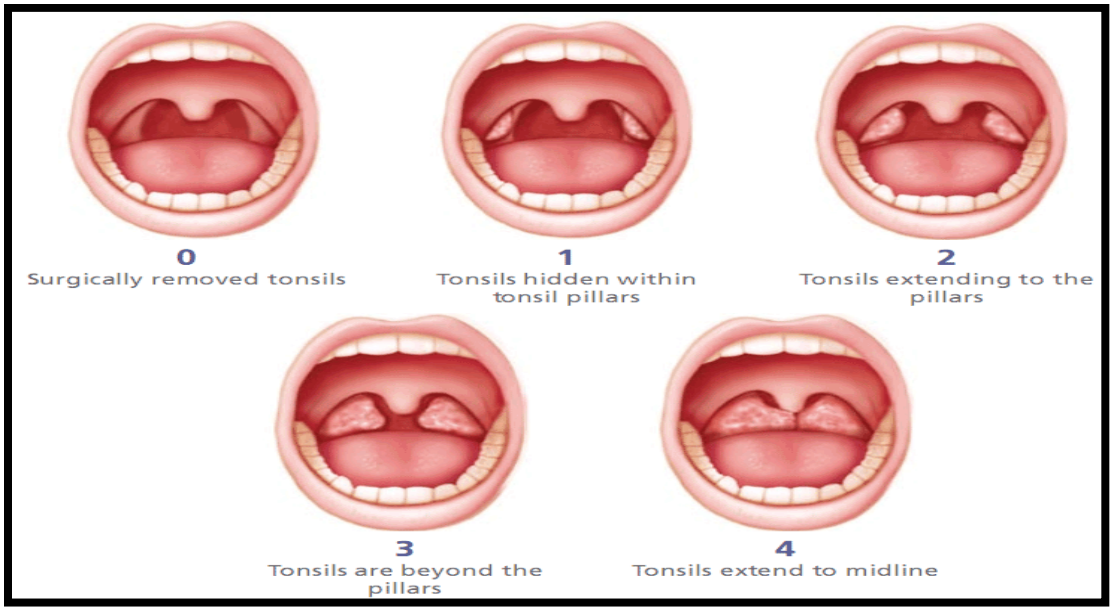


Figure 9: Tonsillar grading(80)

(Figure adapted from: Brodsky et al. A comparison of tonsillar size and oropharyngeal dimensions in children with obstructive adenotonsillar hypertrophy. International journal of pediatric otorhinolaryngology. 1987

Huynh et al. conducted a study on 604 children in a general orthodontic setting to assess associations between SDB with facial and dental morphology. They demonstrated that dolichofacial morphology and increased mandibular plane angle were significantly associated with several SDB symptoms. Also, SDB symptoms such as snoring, mouth breathing and daytime sleepiness were positively correlated with a narrow palate and decreased maxillary width. In the sagittal plane, retrognathia and overjet were not highly associated with SDB symptoms; however, they were statistically correlated with sleep bruxism and morning headaches. (10) Another study by Ameli et al. reported that in a suspected SDB pediatric population, 65% of subjects presented with dental malocclusions.(81) These findings support the association between SDB and craniofacial morphology.

A skeletal class II, increased overbite, maxillary constriction and a inferior hyoid bone position are all factors that may predispose children to apnea.(82) A dolichocephalic facial pattern and a narrow upper airway are common craniofacial characteristics in children with OSA.(40) A prospective study conducted by Schutz et al. showed a decrease in RERAs and RDIs in 16 children treated with an acrylic splint Herbst appliance combined with a maxillary expander (Figure 10). These children presented with a skeletal Class II pattern and a mild maxillary constriction. Furthermore, post-orthopedic functional treatment magnetic resonance imaging (MRI) showed a statistically significant increase in nasopharynx, oropharynx, hypopharynx total volume. A 6.1mm increase in effective mandible length as well as a 3.2mm maxillary expansion was calculated.(82)

Figure 10: Herbst appliance(83)

(Figure adapted from:http://mdgsleepacademy.com/sleep-appliances/)

In general, a retrognathic mandible is concomitant with class II functional oral appliances which produces an anterior displacement of the mandible and the hyoid bone causing an anterior traction of the tongue.(84) As a result, in Schutz's study, a 3.2mm posterior airway increase and reduced airway resistance was observed in children treated with a Herbst appliance. A proper swallowing pattern was also observed in those children due to anterior repositioning of the tongue. Proper swallowing also reduces tongue hypotonia, therefore helping the tongue to not fall back during the hypotonic stage of rapid-eye movement (REM) sleep.(82)

As discussed above, maxillary constriction may be seen in children with OSA. Rapid maxillary expansion is a potential adjunct treatment in pediatric sleep apnea, especially in post-adenotonsillectomy persistant OSA. Its main goal aside its sleep apnea benefit is to correct an existing maxillary posterior crossbite. A study by Villa et al. in 2007, demonstrated that an orthodontic treatment with RME significantly decreased OSA symptoms in 71.4% and AHI in 78.4% of children.(85) In a 36-month follow up study conducted by Villa et al. on the same 10 children, 80% of subjects showed a stable decrease in clinical and polysomnographic signs and symptoms of OSA.(40) Another study suggested that both RME and adenotonsillectomy may be essential to correct completely OSA and mouth breathing in children.(71) Guilleminault et al. showed that in 14.5% of children post-T&A, symptoms of OSA were still present three months post-surgery. He suggests that adjunct RME is necessary to resolve those signs and symptoms.(86) Villa et al. also demonstrated that mouth breathing was resolved in almost all children. RME therapy widens the buccal cavity and distorts the maxillary bone which enlarges space for adenoids and tonsils. These findings suggests that

adjunct orthodontic therapy should be proposed in children with OSA to help correct oral breathing as well provide benefits to airway obstruction.(40)

In healthy subjects, the nose is accountable for half of respiratory resistance. Therefore, any nasal obstruction due to craniofacial abnormalities could add to OSA factors. One of the principal goals of RME is to reduce nasal resistance. This is done by palatal expansion which increases the volume of the nasal and buccal cavities. Pharyngeal obstruction is therefore reduced by proper tongue repositioning in the buccal cavity. Also, in this study, 78.5% of children had hypertrophic tonsils and were chronic snorers. After RME, daytime and nighttime respiratory symptoms were reduced due to the enlargement of the buccal cavity.(87)

Children with deep/retrusive bites and crossbites had a superior improvement after RME in symptoms and polysomnographic variables with comparable amounts of intermolar distance gain than other children. This can be explained by additional benefits of orthodontic treatment in those cases in allowing proper tongue positioning and swallowing. Therefore, orthodontic treatment in children presenting with OSA and dental malocclusions should be commenced early to avoid developing its associated morbidities.(85) In that same study, the authors found no significant correlations between tonsillar hypertrophy and severity of SDB. AHI values were comparable no matter tonsillar grade. This outcome can perhaps explain how tonsillar hypertrophy is not always the mere risk factor in pediatric OSAS, particularly since RME improved the condition even in severe tonsillar hypertrophy. Relatively speaking, the new enlarged buccal cavity post-RME therapy makes tonsils appear relatively smaller.(85)

According to some authors, another craniofacial characteristic common in pediatric OSA is an ogival palate. During development, a posterior tongue position can contribute to the lateral palatine processes to expand vertically contouring the tongue before fusing at the midline causing that high-arched palate shape.(40) However, Smith et al. showed no difference in palatal height in children with OSA compared to controls using dental casts as measurement.(88)

According to Pirila-Parkkinen, cephalometric analysis is a valid method for measuring the dimension of the nasopharyngeal and retropalatal region.(89) A number of cephalometric studies have been conducted to better understand the craniofacial morphologic features of patients with SDB. Compromised breathing during sleep can arise from a combination of pathophysiological and anatomical features resulting in the narrowing of the upper airway. However, the precise location of the obstruction may differ from one person to another. These results reveal that individuals with SDB display few morphological dissimilarities in skeletal and soft tissue proportions, airway dimensions and hyoid positions. A shortened cranial base with reduced antero-posterior skeletal dimensions, variable outcomes in relation to hyoid bone position and an increase in both soft palate length and thickness are all examples of anatomical features which contribute to SDB.(90) Other factors such as a short mandibular body and mandibular retrusion are also associated with SDB.(91) Increased total and lower anterior face heights and larger craniofacial angles are also reported in SDB patients.(92)

Dental arch morphology is another risk factor in children with SDB. For example, narrower maxillae, deeper palatal height and shorter lower dental arch are associated with

SDB.(93) Increased overjet and reduced overbites are other significant examples of measures related to SDB.(94) Also, compared to controls, children with SDB present to have shorter maxillary arches and reduced intercanine widths.(88)

2.9 Sleep bruxism (SB)

Sleep bruxism is a sleep-related movement disorder also classified as a parafunction in dentistry.(95) Typically, SB teeth grinding is reported during childhood and adolescence with an overall prevalence ranging between 8% and 38% (96-98) and tends to decrease after adulthood from 8% to 3% in older adults.(99-101) SB is characterized by episodes of rhythmic masticatory muscle activity (RMMA) of the masseter and temporalis muscles. This activity can be observed and scored when electromyographic recordings are performed during sleep.(102) SB scoring relies on the recognition of RMMA, a succession of jaw muscle contractions, over the sleep period occurring mainly in light sleep stage N2. Grinding sounds, due to tooth contacts with jaw displacements, is the pathognomonic sign of SB that is usually reported by the patient's sleep partner, siblings and/or parents. However, teeth grinding sounds do not occur during all RMMA/SB episodes. In children, SB can be associated with orofacial pain and headaches, and tooth damage.

Although the etiology of SB remains unknown, the multifactorial physiopathology is partly explained by re-activation of the cerebral cortex and autonomic nervous system during sleep, a process named sleep arousal, that occurs during periods of sleep instability.(103, 104) Increased respiratory amplitude is associated with RMMA,(105) which supports the hypothesis of an association between RMMA and breathing during sleep.

Studies have shown a higher incidence of sleep apnea or SB when they are comorbid. Moreover, SB teeth grinding decreased or disappeared in most children with sleep apnea who underwent adenotonsillectomy.(106, 107) Likewise, Bellerive et al. reported on a 32 patient sample study that although sleep and respiratory variables persisted, 65% of bruxers saw a reduction in RMMA after expansion.(108)

Chapter 3. Objectives and Hypothesis

3.1 Problematic

Our preliminary data suggest that from 604 patients (7-17 years) seen at the orthodontic clinic, up to 18% of respondents have compromised breathing during sleep and in whom comorbidities are also present.(10) Mandibular retrognathia, a narrow maxillary/mandibular ratio, a long and narrow face may be associated with these phenomena.(92, 94, 109, 110) Furthermore, preliminary studies on rapid palatal expansion or surgically assisted expansion, suggest an improvement in SDB.(36) Establishing a prevalence count in a multi-centric study across Canada and early detection of SDB and craniofacial development abnormalities may reduce the risk of developing the associated consequences.

3.2 Type of study

This study consists of a multi-centric prospective study in which the data will be collected in 5 clinical sites. Each clinical site will be responsible of recruiting 100 children completing a sleep study for a total of 500 subjects. One of the sites will recruit 100 children for the control group who do not present any sleep apnea symptoms confirmed by PSG.

3.3 Study purpose

The purpose of this prevalence study is to count the number of patients who would benefit from a dental and orthodontic evaluation among those presenting with respiratory sleep disorders seen at the CHU Sainte-Justine. Sleep questionnaire, polysomnographic, orthodontic and craniofacial data will be further analyzed.

3.4 Hypothesis

1. Research hypothesis:

Our research hypothesis is that the prevalence of malocclusions and dento-skeletal abnormalities would be different between apneic and non-apneic children.

2. Null hypothesis:

The prevalence of malocclusions and dento-skeletal abnormalities would not be different between apneic and non-apneic children.

Chapter 4. Materials and Methods

4.1 Ethics committee

The project received the approval of the ethics committee of the CHU Sainte-Justine on March 31st 2014 and has been renewed yearly. Refer to annex 1 for ethics committee approval.

4.2 Patient selection

Patients referred for a polysomnographic sleep recording for proper diagnosis of SDB at the sleep laboratory of CHU Sainte-Justine were seen in this study. A total of 100 patients were examined. The patients seen at CHU Sainte-Justine take part in a national prospective cross-sectional study. Data is being collected at 4 other clinical sites, as well at a site that will provide control participants with no sleep apnea confirmed by polysomnography. Each of the other 4 clinical sites is recruiting patients with the same profile while the Dalhousie University site will recruit healthy participants. Gozal's questionnaire as well as craniofacial and orthodontic data collection took place during the scheduled appointment on the night of the polysomnographic sleep study. No additional visit or follow-up were required.

Inclusion Criteria

- Children aged 4-17 years willing to complete a polysomnographic sleep study at the sleep laboratory of the CHU Sainte-Justine for evaluation of snoring and apnea.

Exclusion Criteria

- Children with craniofacial anomalies linked to genetic syndromes.
- Children currently under CPAP treatment.

4.3 Data collected

Facial and orthodontic data were collected. Refer to Annex 2 for full clinical exam. An adapted questionnaire from Spruyt & Gozal et al. was handed to one parent for further sleep information on the child presenting for the sleep study. Refer to Annex 3 for full sleep questionnaire.

The following polysomnographic data was collected. Sleep technicians trained in each site used a standardized evaluation method according to the parameters established by the American Academy of Sleep Medicine.

- Height
- Weight
- Total sleep time
- Sleep efficiency
- Respiratory Disturbance Index
- Mean and minimum oxygen saturation
- Oxygen desaturation index
- Minimum and maximum respiratory rate
- Minimum and maximum heart rate
- $TcCO_2$ range
- Central apnea counts and index
- Total apnea counts and index

- Total Hypopneas counts
- Apnea Hypopnea Index (AHI)

4.4 Initial examination

Explanation of the procedure was given to the parent/guardian and the child. A customized consent formed was then given and read aloud before receiving proper consent. Refer to annex 4 for complete consent form. A standard orthodontic evaluation was completed on the night of the child's polysomnographic sleep study at the CHU Sainte-Justine. A standard dental examination kit was used to record data as well as a Boley gauge to record distance. Following complete consent, data, and questionnaire collection, a 20$ gift certificate of their choice was given to the patient for accepting to participate in the study.

Children were then accompanied to their sleep study room. Complete type 1 polysomnographic measures were then set up by the on call registered inhalotherapist nurse. Sleep data was extracted for analysis in this study.

<u>Dependent variables:</u>

- Respiration during sleep
- Questionnaires
- Sleep quality

<u>Independent variables:</u>

- Craniofacial massif

- Dentition

4.5 Statistical analysis

An electronic data capture was used: Redcap ™ (Research Electronic Data Capture). The data was codified with an alphanumeric code to prevent patient identification. The Redcap ™ system uses a secure Web connection requiring authentication. Only members of the research project had access to the data.

Intraclass correlation was performed with an experienced orthodontist, Dr. Andrée Montpetit, for patient orthodontic evaluation. Fischer's Exact Test, Mann-Whitney U-tests and two-sample t-test were used for prevalence calculation of dental malocclusions in children with SDB as well as Spearman correlation between dental malocclusions and type 1 polysomnographic data. A logistic regression was also done. Data was analyzed using SPSS 24 by an experienced statistician.

Chapter 5. Results

5.1 Patient description

In this prevalence study, 100 patients were recruited; 58 of them were male and 42 were female (Figure 11). The age ranged between 3-18 years old, the mean being 9.6 years old ± 4.05 (Figure 12). No patients were excluded from the study and all parents/guardians and patients consented to participate in the study. All patients included in this study responded to the inclusion criteria. No patients were secondarily excluded from the study after polysomnographic data analysis. Orthodontists were blinded to polysomnographic scores when doing clinical evaluation of patients on the night of sleep study. Kappa scores were rated excellent for ICC calculations. Subjects were separated in two different AHI groups (AHI < 2, AHI ≥ 2) for analytic purposes per the AAP guidelines.(111, 112)

The clinical data reported are based on the clinical evaluation done on the day of the polysomnography at the CHU Sainte-Justine. The polysomnographic data originate from the sleep study done the same night as the clinical evaluation compiled by sleep technicians from the site.

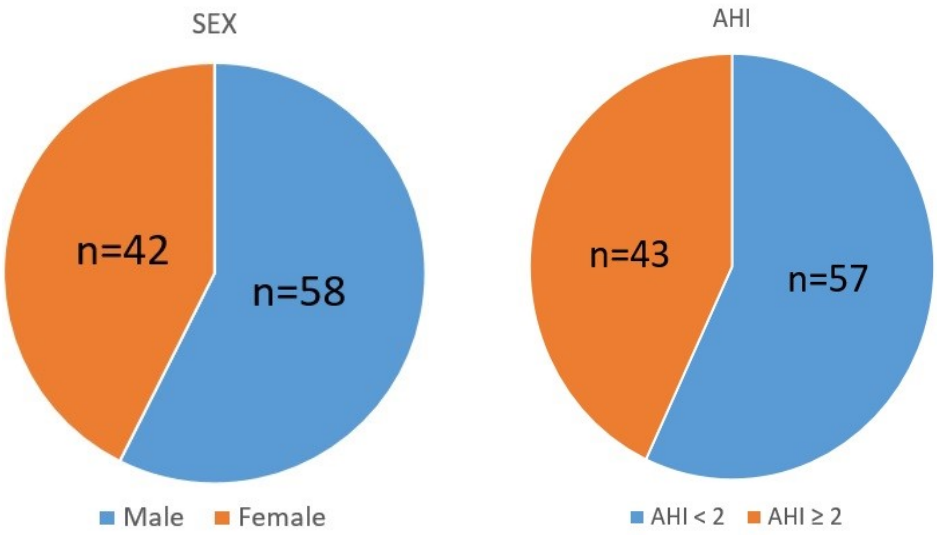


Figure 11: Sex & AHI distribution

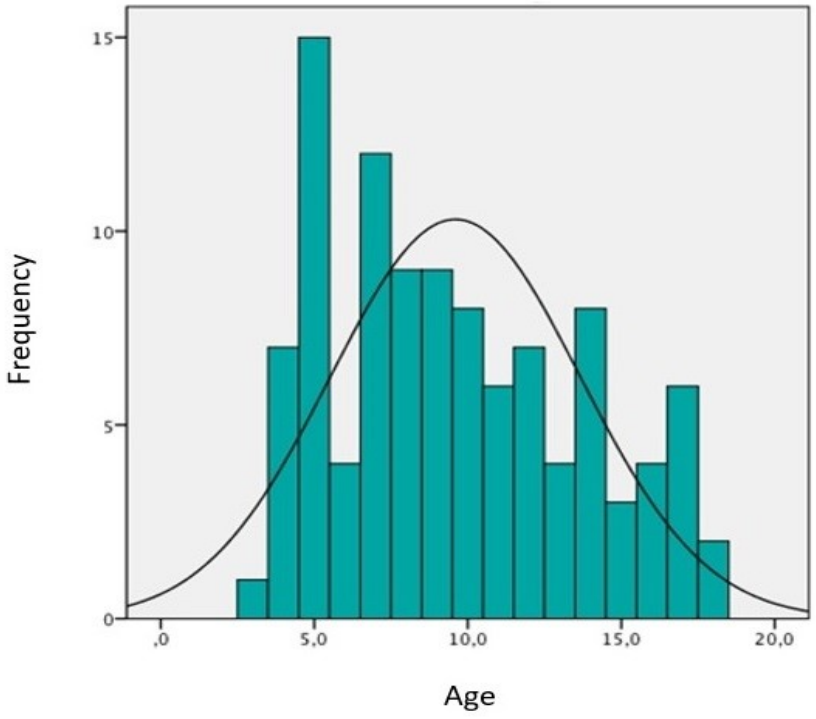


Figure 12: Age distribution

The AHI <2 group had a mean age of 9.98 ± 3.83 and the AHI ≥ 2 group had a mean age of 9.16 ± 4.34 (Figure 13). No difference in age was found between AHI groups using t-test analysis (p=0.320). However, Fisher's exact test showed that boys were more likely to present with sleep apnea (p=0.043).

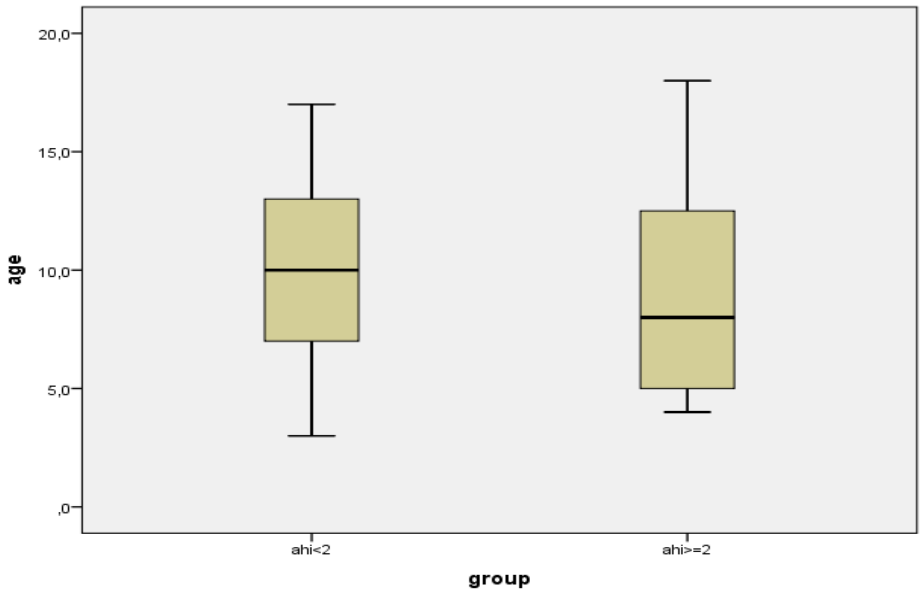


Figure 13: Age per AHI groups

Table 3: Descriptive and polysomnographic data according to AHI
Data shown as Mean +/- SD

	AHI < 2	AHI ≥ 2	p value
AGE (years)	9.98 +/- 3.83	9.16 +/- 4.34	0.320
BMI	21.13 +/- 7.78	23.21+/- 10.61	0.303
Oxygen Desaturation Index	5.12 +/- 19.69	18.62 +/- 30.59	0.000
Apnea-Hypopnea Index	0.79 +/- 0.53	7.79 +/- 8.03	0.000

5.2 Body mass index

BMI per subject was calculated by dividing their weight (kg) by their height squared (m). No significant difference was found between AHI groups and BMI value in children (p=0.303) (Figure 14). The mean BMI value in the AHI < 2 group was 21.13 ± 7.78 and 23.21 ± 10.61 in the AHI ≥ 2 group.

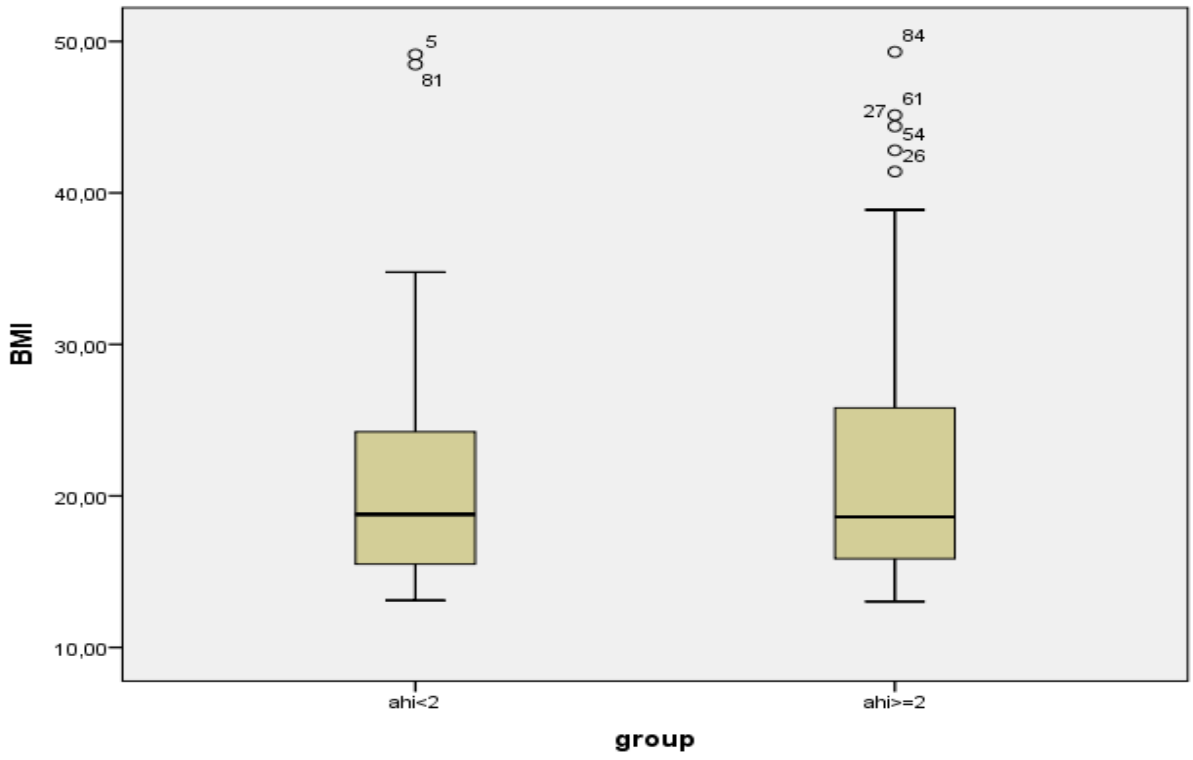


Figure 14: BMI per AHI groups

5.3 Gozal score

The Gozal questionnaire is a helpful tool consisting of a set of six ordered questions along the SDB spectrum (frequency and intensity of snoring, breathing) given to the parent/legal tutor which allows screening of children at risk of SDB. No significant difference was found between Gozal score and AHI groups (p=0.220) (Figure 15). The mean Gozal score in the AHI < 2 group was 1.58 ± 0.92 and 1.82 ± 1.05 in the AHI ≥ 2 group.

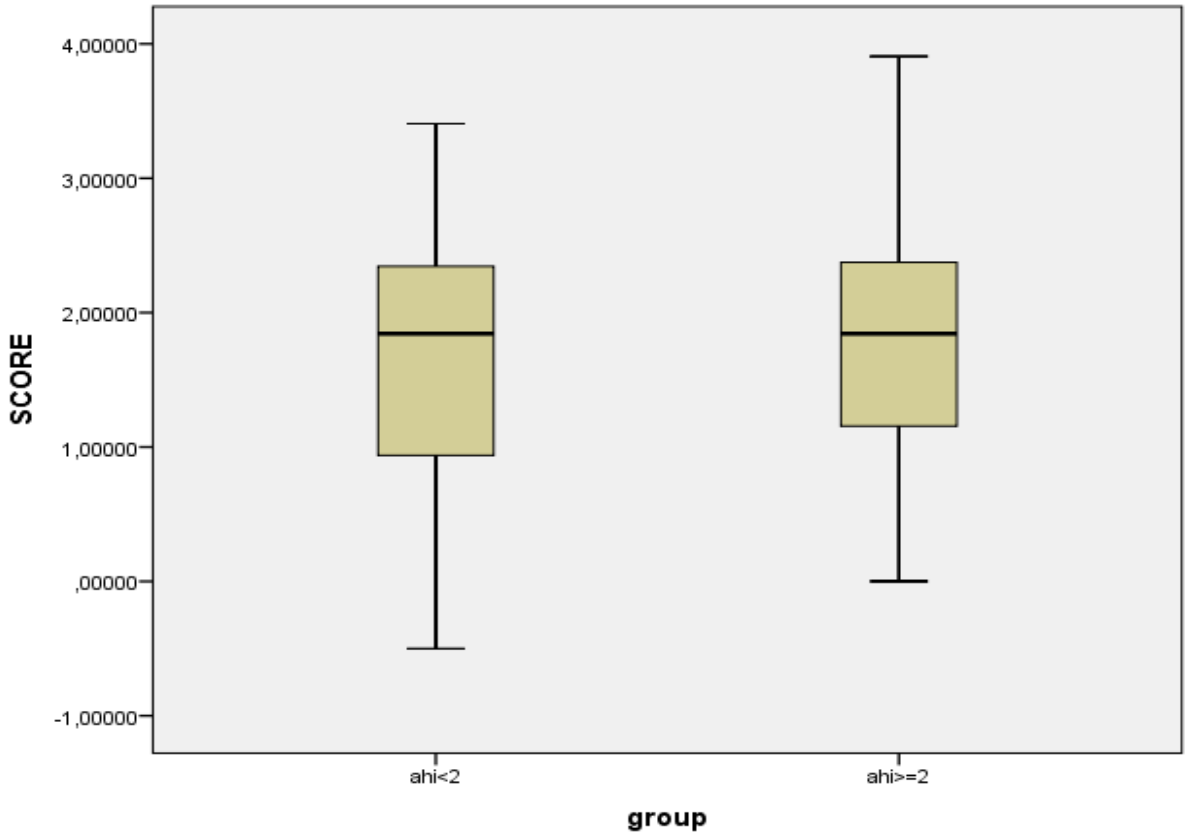


Figure 15: Gozal score per AHI groups

Spruyt and Gozal severity score was then studied with a filter of ≥ 2.72. Spruyt and Gozal showed that pediatric patients with a score ≥ 2.72 have an increased risk of presenting an AHI ≥ 3.(23) Patients with a score < 2.72 had an AHI median of 1.4 (0-40.7) and patients with a score ≥ 2.72 had a median of 5.8 (0.3-23.4) (Figure 17). Mann-Whitney U test showed a statistical difference in that Gozal score was significant in predicting more severe AHI for a score ≥ 2.72 (p=0.011) (Figure 16).

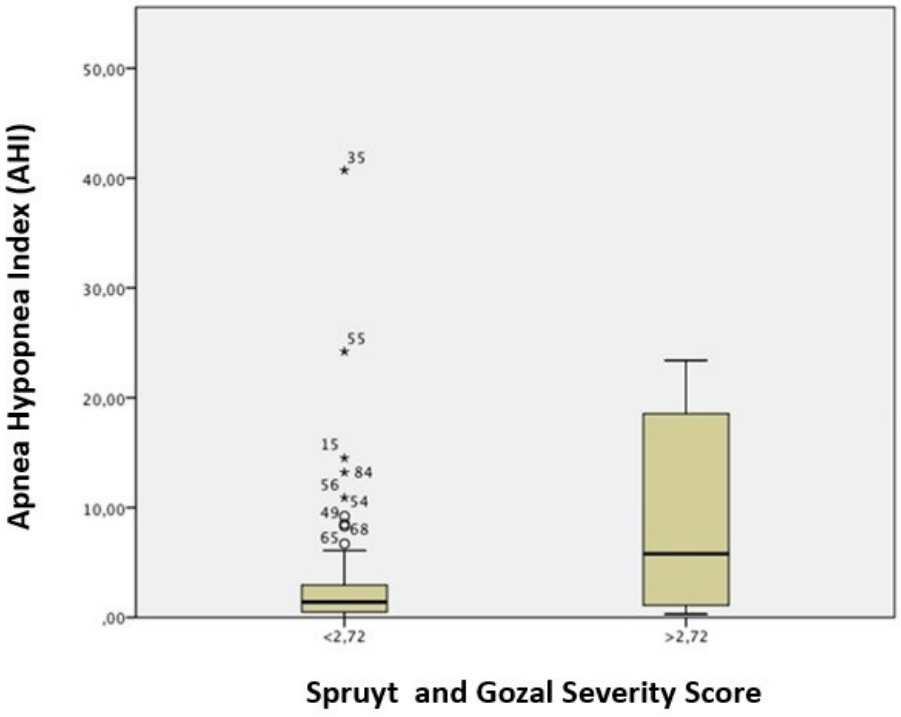


Figure 16: AHI per Spruyt & Gozal severity score

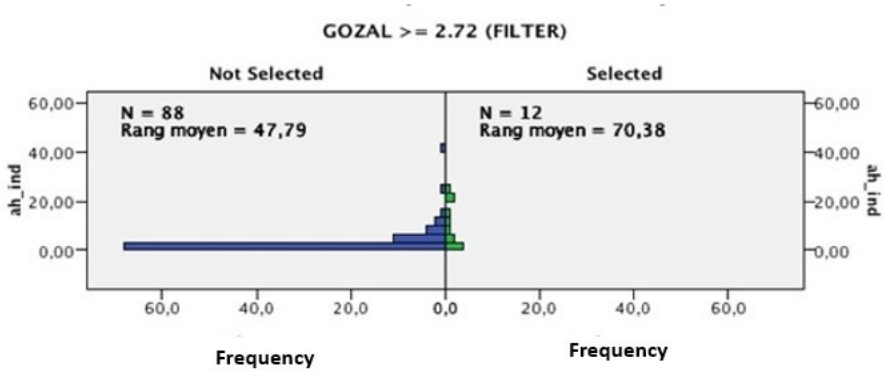


Figure 17: AHI distribution per Gozal score

5.4 Descriptive polysomnographic data

The following data summarizes sleep recorded data for all subjects analysed by sleep technicians at the CHU Sainte-Justine (Table 4).

Table 4: Descriptive polysomnographic data

Group		ox_desat_ind	tcco2_r	resp_dist_ind
ahi<2	N	48	34	45
	Mean	5,1250	41,29	,9556
	Std. Deviation	19,69325	4,407	1,08283
	Median	,9000	41,50	,7000
	Minimum	,00	33	,00
	Maximum	99,00	50	7,00
ahi>=2	N	36	27	34
	Mean	18,6278	42,63	8,7647
	Std. Deviation	30,59316	5,911	8,74164
	Median	5,4000	43,00	5,0000
	Minimum	,30	26	,10
	Maximum	100,00	52	40,70
Total	N	84	61	79
	Mean	10,9119	41,89	4,3165
	Std. Deviation	25,68023	5,125	6,93781
	Median	2,0000	42,00	1,4000
	Minimum	,00	26	,00
	Maximum	100,00	52	40,70

Group		cap_ind	oah_ind	ma_ind	ta_ind	th_ind	ah_ind
ahi<2	N	55	26	32	55	31	57
	Mean	,3581	,2500	,003	,3296	,5581	,7940
	Std. Deviation	,31427	,34205	,0177	,28383	,49178	,53120
	Median	,3000	,2000	,000	,3000	,4000	,7000
	Minimum	,00	,00	,0	,00	,00	,00
	Maximum	1,30	1,50	,1	1,40	1,90	1,90
ahi>=2	N	43	16	24	43	24	43
	Mean	2,4319	3,4563	,150	3,3672	4,3250	7,7930
	Std. Deviation	5,23218	3,98229	,2766	5,03953	4,23559	8,03331
	Median	1,3000	1,9000	,000	1,4000	2,5500	4,6000
	Minimum	,00	,00	,0	,00	,60	2,00
	Maximum	33,50	12,50	1,0	22,90	18,50	40,70
Total	N	98	42	56	98	55	100
	Mean	1,2680	1,4714	,066	1,6624	2,2018	3,8036
	Std. Deviation	3,60253	2,89080	,1938	3,65198	3,36603	6,29805
	Median	,5000	,3500	,000	,5000	,9000	1,6000
	Minimum	,00	,00	,0	,00	,00	,00
	Maximum	33,50	12,50	1,0	22,90	18,50	40,70

5.5 AHI

Patients were divided into subgroups (AHI < 2, AHI ≥ 2) for analytic purposes. 57 patients had an AHI < 2. The mean AHI for this subgroup was 0.79 ± 0.53 with a median of 0.7. The AHI ≥ 2 subgroup was composed of 43 patients. The variability of this subgroup was greater with a mean of 7.79 ± 8.03 with a median of 4.6 (Figure 18).

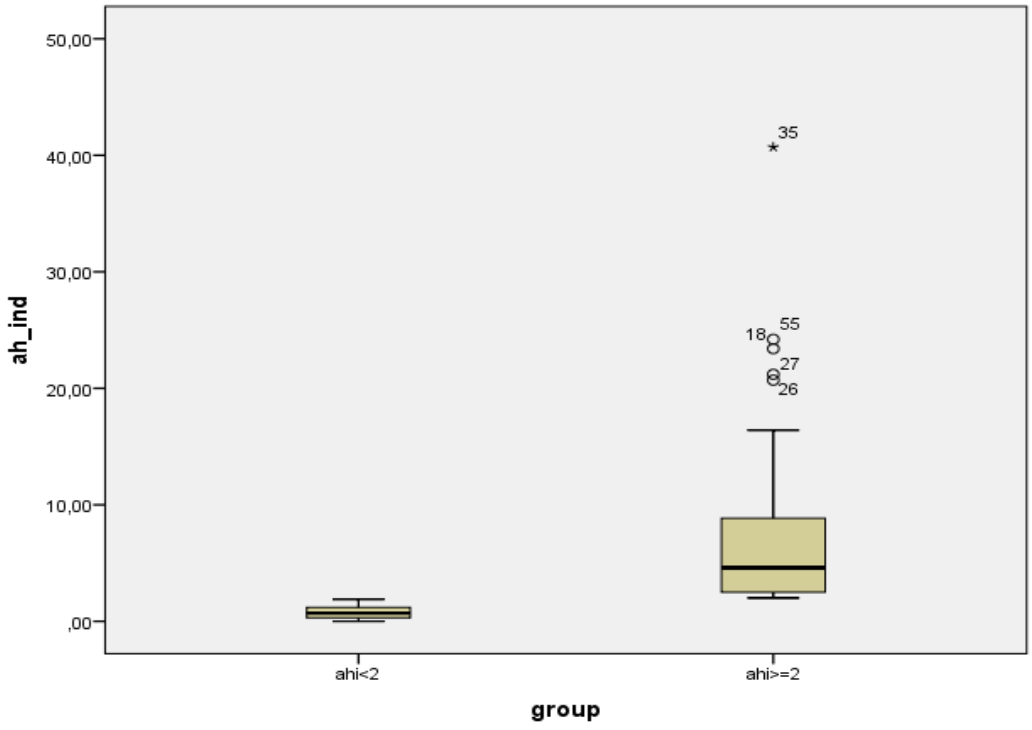


Figure 18: AHI per group

AHI was then correlated with tonsillar hypertrophy grade per Brodsky's tonsillar hypertrophy score. Mann-Whitney U test was used to establish statistical significance. The AHI median in the milder group was of 1.7 (0.1-14.5) and of 1.5 (0-40.7) in the more severe group (Figure 20). No statistical difference was found in AHI in terms of tonsillar hypertrophy grade (p=0.426) (Figure 19).

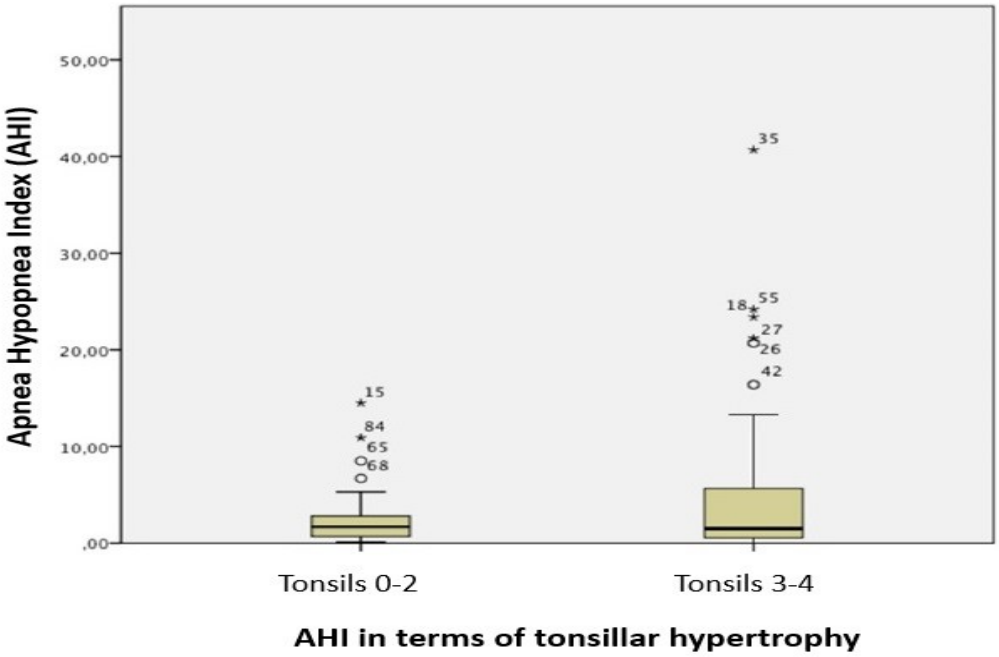


Figure 19 : AHI per tonsillar hypertrophy grade

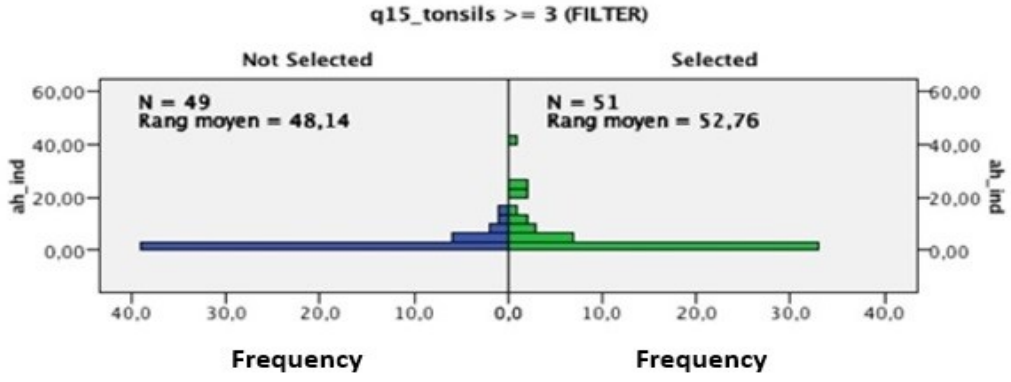


Figure 20 : AHI distribution per tonsillar grade

5.6 Craniofacial morphology

The following histogram summarizes the major craniofacial features in terms of prevalence in both AHI groups (Figure 21). Their statistical significance is listed below in their corresponding tables.

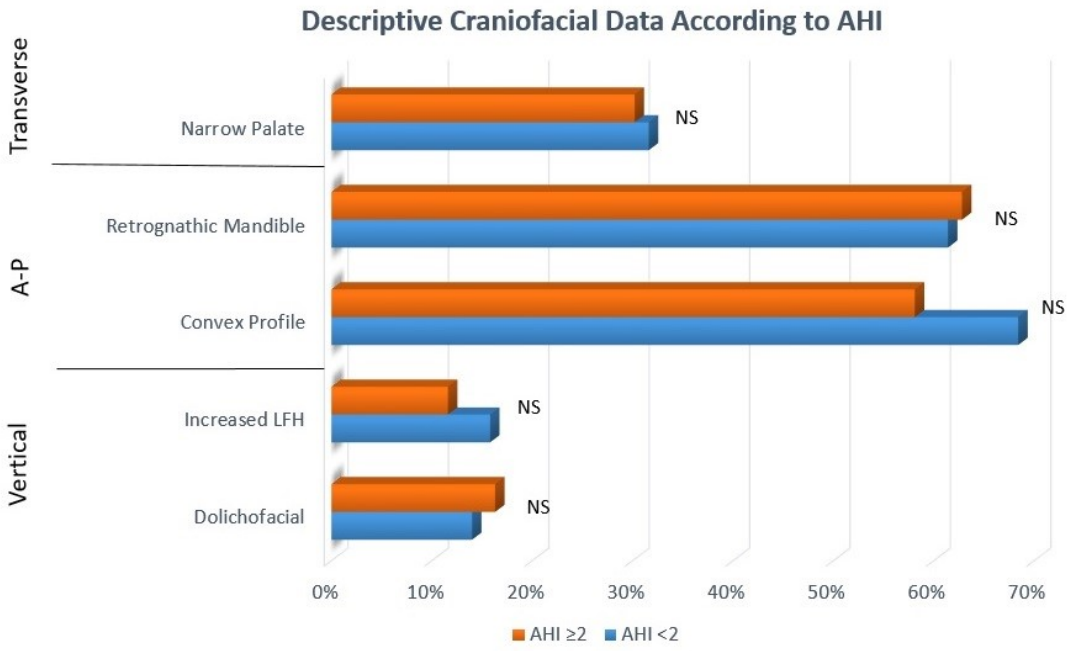


Figure 21: Descriptive Craniofacial Data according to AHI

No significant difference was found between AHI groups for nominal craniofacial morphological characteristics using Fisher's Exact Test (Table 5):

Table 5: Craniofacial data statistical significance

Craniofacial Morphology	p Value
Body type	0.410
Facial type	0.800
Lower face height	0.908
Facial profile	0.572
Maxilla	1.000
Mandible	1.000

5.6.1 Facial soft tissue characteristics

No significant difference was found between AHI groups for nominal facial soft tissue characteristics using Fisher's Exact Test (Table 6):

Table 6: Facial soft tissue data statistical significance

Facial Soft Tissue Characteristics	p Value
Nasolabial Angle	0.509
Upper Lip Position	0.764
Lower Lip Positon	0.940
Lip Strain	0.614

5.6.2 Intra-oral soft tissue characteristics

No significant difference was found between tongue size between AHI subgroups using Fisher's exact test (p=1.0). A total of 5 patients presented with an observable clinical macroglossia in the AHI < 2 group and 4 patients in the AHI ≥ 2 group.

5.6.3 Intra-oral dental characteristics

The following histogram summarizes the major intra-oral dental characteristic in terms of prevalence in both AHI groups (Figure 22). Their statistical significance is listed below in their corresponding tables.

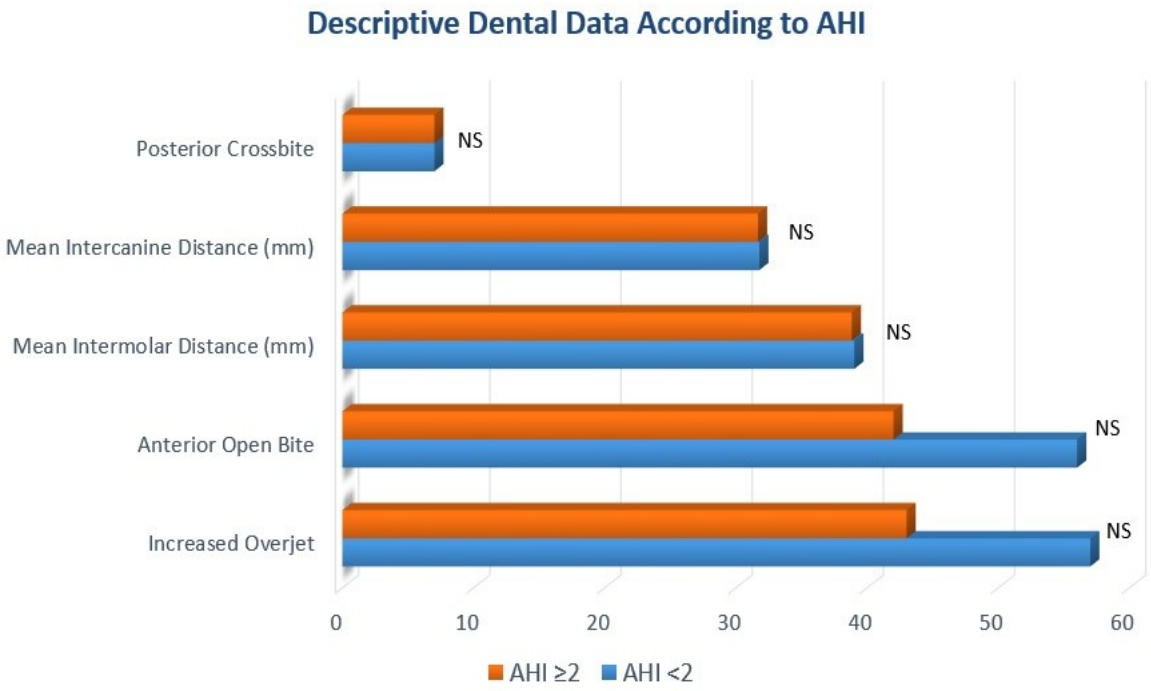


Figure 22: Descriptive Craniofacial Data according to AHI

No significant difference was found between AHI groups for nominal intra-oral characteristics using Fisher's Exact Test (Table 7):

Table 7: Intra-oral data statistical significance

Intra-oral Characteristics	p Value
Anterior Crossbite	1.000
Posterior Crossbite	0.576
Narrow Palate	1.000
CR/CO Shift	1.000
Upper Arch Shape	1.000
Lower Arch Shape	0.649
Dentition Stage	0.327
Crowding	0.261
Spacing	0.833

No significant difference was found between AHI groups for numerical intra-oral characteristics using Mann-Whitney U Test (Table 8):

Table 8: Intra-oral data statistical significance

Intra-oral Characteristics	p Value
Incisor Display at Rest	0.265
Gingival Display at Smile	0.235
Incisor Display at Smile	0.266
Anterior Open-Bite	0.208
Right Posterior Open-Bite	0.258
Left Posterior Open-Bite	0.258
Index of Treatment Need	0.298

No significant difference was found between AHI groups for numerical intra-oral characteristics using two-sample t-test (Table 9):

Table 9: Intra-oral data statistical significance

Intra-oral Characteristics	p Value
Overjet	0.212
Overbite	0.336
Intermolar Distance	0.884
Intercanine Distance	0.906

5.6.4 Functional data

Functional data were also studied and their significance was analysed between AHI groups using Fisher's exact test. Mouth breathers were found to be non-significant between AHI groups (p=0.473). The AHI <2 group had a total of 28 patients with reported mouth breathing and the AHI ≥ 2 had 21 patients with reported mouth breathing. Day or night time mouth breathing was not significant either (p=1.0).

A trend was found between patients with oral habits altogether (nail biting, cheek/lip biting, bruxism, thumb sucking) and an AHI <2 (p=0.064). However, each oral habit alone was found to be non-significant between AHI groups (nail biting p=0.140, cheek/lip biting p=1.0, bruxism p=0.650, thumb sucking p=0.632).

5.6.5 Correlations between malocclusions and polysomnographic data

Correlations between polysomnographic data and clinical variables were calculated by combing all normal values vs abnormal sleep apnea predictors. The clinical values were correlated with the following polysomnographic data: oxygen desaturation index, CO_2 and TCO_2 maximum range, total apnea index and apnea hypopnea index. The following table summarizes the lien between those correlations and their significance using Mann-Whitney U test (Table 10).

Table 10: Correlation between malocclusions and polysomnographic data

	Oxygen Desaturation Index	CO_TCO_2	Total Apnea Index	Apnea Hypopnea Index
Facial Type (N Vs Non-N)	0.738	0.566	0.867	0.964
AHI	0.000	0.013	0.000	0.000
Lower Face Height (N Vs Non-N)	0.598	0.348	0.421	0.990
Facial Convexity (Straight/Concave Vs Convex)	0.219	0.711	0.193	0.231
Maxilla (N Vs Non-N)	0.621	0.877	0.719	0.848
Mandible (N Vs Non-N)	0.677	0.198	0.677	0.667
Mouth Breathing	0.809	0.350	0.630	0.774
Oral Habits	0.152	0.626	0.358	0.174
Bruxism	0.293	0.285	0.877	0.977
Lip Biting	0.171	0.981	0.269	0.824
Nail Biting	0.752	0.770	0.646	0.190
Thumb Sucking	0.851	0.937	0.948	0.983
Other Oral Habits	0.060	0.233	0.827	0.990
Anterior Cross-bite	0.355	0.274	0.535	0.668
Palate (N vs Narrow)	0.994	0.458	0.815	0.993
Posterior Cross-bite	0.439	0.582	0.555	0.651

Spearman correlations test was used to calculate the following variables with the same polysomnographic data (Table 11).

Table 11: Correlation between malocclusions and polysomnographic data

	Oxygen Desaturation Index	CO_TCO_2	Total Apnea Index	Apnea Hypopnea Index
Tonsils	0.905	0.313	0.517	0.380
Overjet	0.912	0.122	0.536	0.175
Overbite	0.243	0.117	0.187	0.229
Intermolar Distance	0.463	0.094	0.298	0.581
Maxillary Crowding	0.534	0.871	0.921	0.305
Mandibular Crowding	0.213	0.921	0.702	0.939
Maxillary Spacing	0.462	0.056	0.549	0.128
Mandibular Spacing	0.685	0.828	0.745	0.624

5.6.6 Correlation between clinical data and numerical AHI

Correlations between clinical data and numerical AHI were done by combining all normal values vs abnormal known sleep apnea predictors and correlating them to the AHI. The following table summarizes their significance using Mann-Whitney U test (Table 12).

Table 12: Correlation between clinical data and numerical AHI

	AHI
Facial Type (Meso VS Brachy & Dolicho)	0.964
Lower Face Height (N VS Non-N)	0.990
Facial Profile (Straight, Concave VS Convex)	0.231
Maxilla (N VS Non-N)	0.848
Mandible (N VS Non-N)	0.667
Mouth Breathing	0.774
Oral Habits	0.174
Anterior Crossbite	0.668
Posterior Crossbite	0.656
Palate (N VS Narrow)	0.993
Macroglossia	0.487

The following table summarizes other clinical data correlated to AHI using Spearman correlations (Table 13).

Table 13: Correlation between clinical data and numerical AHI

	AHI
Tonsils	0.380
Overjet	0.175
Overbite	0.229
Intermolar Distance	0.581
Upper Crowding	0.305
Lower Crowding	0.939

5.7 Prediction model

A logistic regression was calculated using age, sex, and all morphologic clinical variables with a $p<0.20$ in a univariate analysis. Polysomnographic variables were not included in the regression calculation since the sleep study would reveal if the patient has sleep apnea or not and therefore a predictive model would not be needed. Relevant clinical

significant variables such as sex (p=0.043), oral habits (p=0.064), overjet (p=0.212), and anterior open bite (p=0.208) were included. Furthermore, BMI (p=0.303), age (p=0.320), and Gozal score (p=0.220) values were also incorporated since they are known sleep apnea predictors.

Table 14: Tests of model coefficients

		Chi-square	df	Sig.
Step 1	Step	18,069	7	,012
	Block	18,069	7	,012
	Model	18,069	7	,012

The p for Model (p = 0.012) indicates that the model is significant and predicts better than a model with only one constant (Table 14).

Table 15 : Model summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	104,511	,182	,245

The R of 0.245 indicates that about 25% of the yes/no variance of apnea is predicted by the combination of model variables (Table 15).

Table 16 : Classification table

			Predicted		
			group		
	Observed		ahi<2	ahi>=2	Percentage Correct
Step 1	group	ahi<2	42	10	80,8
		ahi>=2	17	21	55,3
	Overall Percentage				70,0

The table classification shows that the status of 70% of subjects is predicted correctly (63/90) (Table 16).

Table 17: Variables in the logistic regression equation

							95% C.I.for EXP(B)	
		B	S.E.	Wald	df	Sig.	Exp(B)	Lower	Upper
Step 1	age	-,110	,076	2,092	1	,148	,896	,771	1,040
	gender(1)	1,260	,520	5,861	1	,015	3,524	1,271	9,772
	SCORE	,262	,269	,952	1	,329	1,300	,767	2,202
	q17_oral(1)	-1,099	,502	4,797	1	,029	,333	,125	,891
	q18_overjet	-,210	,150	1,956	1	,162	,811	,604	1,088
	q20_ant_ob	-,345	,478	,521	1	,470	,708	,277	1,808
	BMI	,082	,036	5,156	1	,023	1,085	1,011	1,165
	Constant	-,803	,932	,743	1	,389	,448		

Significant results were found with the logistic regression calculation. In general, boys were found to be more at risk of sleep apnea (OR = 3.52, 95% CI 1.27-9.77). Also, subjects with oral habits are less at risk (OR = 0.33, 95% CI 0.13-0.89). Finally, the odds of having apnea increases by 1.09 for each unit of BMI increase (Table 17).

Chapter 6. Discussion

6.1 Subject description

In this study, 100 patients with SDB were recruited at the CHU Sainte-Justine. Out of the total subject population studied, 58 of them were male. Boys were more likely to present with sleep apnea. Furthermore, logistic regression showed that boys were found to be more at risk of sleep apnea. Our results are in accordance with the few recent papers studying gender prevalence in pediatric sleep apnea. The results are much more evident in adults. Literature has shown over the years that the prevalence of sleep apnea in male-to-female ratio is 2:1.(113, 114) Gender differences theories include hormonal effects and anatomical differences in upper airway structures as well as body fat distribution differences between genders.(114) However, gender-based differences in pre-pubertal children studies are quite recent and the results aren't convincing.(115) Certain studies show a higher prevalence in boys.(68, 116) A review conducted by Lumeng et al. indicated a reasonable male majority in pediatric SDB. The results are more conclusive in children 13 years and older. Post-pubertal hormonal change is the probable main factor. After doing an extensive review of 35 gender-based studies in pediatric sleep apnea, the authors conclude that pediatric SDB is 1.5-2X more prevalent in boys than girls.(4)

A possible limit in our study was population based. Our population sample was hospital-based and were referred for polysomnography by ENT doctors for suspicion of SDB. Furthermore, those cases were unclear SDB cases or they would have been operated for T&A with simple clinical history and take-home oximetry. Also, a control group was not included in

this study for comparison. As mentioned above, this population pool takes part in a multi-center trans-Canadian study where a control group is currently being recruited at Dalhousie University when a final more extensive analysis will be done.

6.2 BMI

In our population sample, no significant difference was found between AHI groups with BMI value in children and the difference between groups is minor. The mean BMI value in the AHI < 2 group is 21.13 ± 7.78 and 23.21 ± 10.61 in the AHI ≥ 2 group. Contrary to the adult population, obesity is known to be a secondary factor in pediatric sleep apnea. Several studies show no significant difference between BMI and pediatric sleep apnea.(117, 118)

6.3 Spruyt & Gozal questionnaire

In 2012, Spruyt and Gozal showed that pediatric patients with a severity score ≥ 2.72 on their questionnaire have an increased risk of presenting with an AHI ≥ 3 with 79% sensitivity and 59% specificity. Parent-reported snoring was found to be the relevant discriminant symptom factor in sleep questionnaires in screening apneic vs non-apneic snorers.(23) In our study, our sleep questionnaire analysis based on the severity scale demonstrated similar results. We demonstrated that a statistical difference in the Spruyt & Gozal severity scale was found to be significant in predicting more severe AHI for a score ≥ 2.72.

6.4 Oral habits

Our study showed through logistic regression calculations that oral habits is a protective factor in children for sleep apnea. Moreover, although each oral habit alone was found to be non-significant with AHI groups, a trend was found with patients presenting oral habits altogether and an AHI < 2. Oral habits in children would therefore be a possible sort of reactive protective mechanism in sleep apnea. For example, although a direct causal relationship cannot be concluded between SB and sleep apnea, OSA is a risk factor for SB in children.(119) The two seem to be associated to the patient's effort to induce a patent airway during apneic episodes. Indeed, most SB episodes happen in the supine position, a position known to cause a decrease in airway passage. Co-activation of both opening and closing masticatory muscles following resumption of ventilation causes an airway volume increase consequently reducing upper airway resistance.(120) In fact, according to a study by Lavigne et al., 99% of all RMMA's were related to variations in respiration. Specifically, RMMA-SB muscle activity was related to a respiration increase within arousal.(121) Also, in children, bruxism prevalence is higher in 5-6 year old's. Interestingly, adenotonsillar hypertrophy is also at its greatest then explaining both the peak in SDB and bruxism.(122) Actually, Sjöholm reported a higher prevalence in bruxers with mild OSA than with moderate OSA(123) possibly indicating the protective mechanism bruxism provides in OSA. Another example of a SDB protective mechanism may be nail/cheek biting. Presently, the possible role OSA plays in the pathogenesis of onychophagia is unidentified.(124) Actions such as nail biting forces the mandible to protrude, temporarily acting like a mandibular appliance device, and opens up upper airway therefore reducing SDB. Further studies are however needed to better explain the potential role oral habits play in SDB.

6.5 Tonsils

Our prevalence study showed no significant difference between tonsils and AHI groups. Additionally, subjective tonsillar hypertrophy grade per Brodsky's tonsillar grade score showed no significant statistical difference with AHI. Though, literature has shown that hypertrophic tonsils is the primary causal factor in pediatric sleep apnea.(29, 125) However, according to a meta-analysis conducted by Friedman et al., T&A successfully diminishes AHI score in children but does not offer a curable solution. Therefore, to this date, T&A is the first-line treatment.(29) Besides, in a systematic review, Nolan et al. compared clinical and subjective tonsil size (Brodsky's 0-4+ scale) to objective polysomnographic data in pediatric SDB similarly to our research protocol. The inclusion and exclusion factors were comparable to our subject population. Using the same tonsillar grade scale, they concluded that the association between subjective tonsil size and objective OSAS severity is weak at best. Out of 20 studies reviewed, the 9 highest-quality studies express no association between AHI and tonsillar size in non-obese children. Therefore, subjective tonsil size may not be the perfect diagnostic tool and may somewhat negatively influence clinical decision making. Habitually, parent-reported history of abnormal snoring in children indicates high likelihood of OSAS irrespective of tonsil size. The question of correlation between tonsil size and OSAS severity remains. Logically, the larger the tonsils and adenoids, the smaller the upper airway and therefore the higher chance of OSAS severity.(126) Numerous features can however mask tonsillar role in OSAS severity. Firstly, Brodsky's clinical tonsillar scale may be prone to bias due to its subjectivity. In a study by Ng et al. Brodsky's scale has shown acceptable intraobserver and interobserver ICC scores.(127) However, limited to the pediatric population, obtaining a reproducible score may not be that evident. Many children are not cooperative

with a thorough throat exam. Besides, attainment of a clear visual tonsillar evaluation is problematic, especially when it requires a pronounced gag reflex. Consequently, a swift tonsillar approximation is usually obtained with questionable significance.(126)

Additionally, complexity of upper airway anatomy may influence clinical tonsillar assessment. Brodsky's grading scale evaluates tonsil position in relation to the tonsillar pillars in the oropharynx. True anatomical tonsil size is thus not adequately evaluated. Commonly, hypertrophic tonsils do not protrude much from the tonsillar pillars and are better viewed with tonsillectomy or nasofibroscopia. Three-dimensional oropharyngeal exams may therefore be better assessment tools in objectively evaluating true tonsillar role in OSAS.(126) Clinical conditions were also not ideal in our study. Patients were examined sitting in the waiting room before entering for their polysomnography. Sitting positions may have also influenced our clinical values.

Moreover, it is well known that with natural history, OSA severity decreases in growing children. Some clinicians prefer to not treat and tend to observational watching in children with milder OSAS. The mean age in our study was 9.6 ± 4.06 years old. Yet, tonsillar airway volume is at its minimum at 5-6 years old. The face quickly grows between 7-10 years old and tonsillar space reaches its maximum.(78) Hence, our population study corresponds to the maximum increase in tonsillar space and therefore tonsillar hypertrophy may transiently be relatively small in its anatomical space. Also, Tagaya et al. demonstrated little tonsil size influence on AHI in 58 normal weight schoolchildren (age ≥ 6) and preschool children (age < 6) with an AHI ≥ 2 score. Adenoid hypertrophy was only found to be a key factor in preschool

children. The authors conclude that upper airway anatomy in preschool children differs than in schoolchildren and that tonsil size had neither a significant influence on both groups.(112) Comparatively in our study, adenoid size was not taken into account and our population study corresponds to Tagaya`s study. Valera et al.'s study also demonstrated similar findings. They conclude that adenotonsillar hypertrophy is significantly correlated with AHI severity in preschool children. Degree of soft tissue obstruction and AHI severity are not conclusive in schoolchildren.(128) Lastly, Dayyat et al. studied a sample of 206 children with similar AHI and BMI values to our population study. In his study, the authors found a modest association between adenotonsillar size and AHI in non-obese children.(129)

In addition, upper airway anatomy varies between different age groups in children.(130) Adenoid size was not evaluated in our patient population. However, it is well known, that tonsillar and adenoid hypertrophy are independent risk factors for OSAS.(131) Tagaya's study showed that while adenoid hypertrophy was a key risk factor in OSAS in children, tonsil hypertrophy did not provide the same significant results.(112)

Lastly, our population study presented with non-evident SDB. Patients enrolled in our study were referred for complete polysomnography to clarify SDB. If these cases were more evident, patients would've been operated with T&A with a simple take-home oximetry. While type 1 polysomnography is the gold standard diagnostic tool for OSA, it is known that it is not regularly prescribed in the pediatric population since it is costly and time consuming.(126) Frequently, parent-reported history of snoring is sufficient for initial treatment with curative

T&A and most clinicians do not require a pre-operative polysomnography. The reliability of past medical history is adequate for clinical treatment plans.(24) Yet, regardless of its common use, the clinical value of tonsil size as a predictor of OSA severity is not evidently established.

6.6 Craniofacial characteristics

No significant difference was found between craniofacial characteristics and AHI groups. Furthermore, when combining all known craniofacial characteristic predictors associated to polysomnographic data, no significant difference was found. Indeed, a couple of meta-analyses conclude that there exist no direct causal relationship between craniofacial characteristics and pediatric sleep apnea.(132, 133) A meta-analysis led by Katyal et al. with similar patient inclusion criteria than in our study showed little evidence to support craniofacial features' role in pediatric sleep apnea. A significant outcome in their study was that ANB angle was increased by 1.64° in children with sleep apnea compared to controls with lateral cephalogram study. Although the value may be statistically significant, it is of little clinical significance. It is well known in the orthodontic community that ANB angle is affected by many variables such as maxillary and mandibular incisor position as well as vertical and horizontal position of nasion and therefore is not the appropriate sagittal discrepancy measure.(134, 135) Mandibular plane angle to cranial base was shown to be non-significant in this meta-analysis. Kawashima et al. also showed no relation between SDB patients and controls for hyoid bone position as well as facial type.(136) In another case-control study, Schiffman et al. showed using three-dimensional imaging that a smaller mandible is not characteristic in children with OSAS.(137) In another meta-analysis studying

craniofacial morphological characteristics in children with OSAS, Flores-Mir et al. suggest that although MP-SN, SNB, and ANB values were found to be significant, caution should be applied when interpreting those values. The authors conclude that no cephalometric value should be considered pathognomonic and that a direct causal relationship between OSAS and craniofacial characteristics cannot be confirmed.(133) Besides, for reasons mentioned above, SNB and ANB do not measure proper retrognathia. Zucconi et al. showed that children with OSAS have shorter SN measurement than their controls normalizing SNA, SNB, and ANB values.(138)

Finally, most high-quality studies studying craniofacial morphologic features and pediatric sleep apnea use lateral cephalograms to assess their relationship.(109, 139, 140) A limit in our study could be that since lateral cephalograms were not used to examine craniofacial characteristics, facial soft tissue may have influenced our results. It is well known in the orthodontic community that facial soft tissue thickness may mask skeletal malocclusions. Nevertheless, some authors question the relevance of lateral cephalograms when studying pediatric sleep apnea. Besides its inaccuracy when calculating upper airway dimensions, it lacks sensitivity and specificity to be considered a sole diagnostic tool for pediatric sleep apnea.(133) Future higher quality studies are therefore needed to better assess the role of craniofacial morphology and pediatric SDB.

Chapter 7. Conclusion

This prevalence study has permitted us to draw a couple significant conclusions. Spruyt & Gozal's newly adapted sleep questionnaire has been shown to be an effective screening tool in our sample populations especially in children with a severity score of ≥ 2.72. Our predictive model has shown that sleep apnea prevalence is higher in boys and that BMI increases the odds of having sleep apnea.

Prevalence of dental malocclusions in children were found to be non-significant between AHI groups. No significant correlations were found between craniofacial morphology and sleep data. Nevertheless, this is a preliminary analysis. The following study does not permit to draw conclusions in the role of craniofacial morphology and SDB. The goal of this multi-centric study is to recruit up to 400 children when a more extensive analysis will be done. A larger sample study as well as a control group will allow for better conclusions. Therefore, further studies are recommended to draw better conclusions and improve statistical power in the role of craniofacial and dental morphology in OSA children.

Yet, because of the known impact of SDB in children, collaboration between all medical professionals is necessary in assessing children at risk. We recommend that orthodontists and dentists screen children based on reported patient history, sleep questionnaires, and detailed clinical exams. Only then, prevention and better management of SDB will be made more accessible.

References

1. Lavigne GJ, Cistulli P.A., Smith M. T. Odontologie et Medecine du Sommeil: Quintessence International; 2012.
2. Kryger MH. Fat, sleep, and Charles Dickens: literary and medical contributions to the understanding of sleep apnea. Clinics in chest medicine. 1985;6(4):555-62.
3. Alexander NS, Schroeder JW, Jr. Pediatric obstructive sleep apnea syndrome. Pediatr Clin North Am. 2013;60(4):827-40.
4. Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. Proceedings of the American Thoracic Society. 2008;5(2):242-52.
5. Brunetti L, Rana S, Lospalluti ML, Pietrafesa A, Francavilla R, Fanelli M, et al. Prevalence of obstructive sleep apnea syndrome in a cohort of 1,207 children of southern Italy. Chest. 2001;120(6):1930-5.
6. Bixler EO, Vgontzas AN, Lin HM, Liao D, Calhoun S, Vela-Bueno A, et al. Sleep disordered breathing in children in a general population sample: prevalence and risk factors. Sleep. 2009;32(6):731-6.
7. Tang JP, Rosen CL, Larkin EK, DiFiore JM, Arnold JL, Surovec SA, et al. Identification of sleep-disordered breathing in children: variation with event definition. Sleep. 2002;25(1):72-9.
8. Gozal D, O'Brien LM. Snoring and obstructive sleep apnoea in children: why should we treat? Paediatr Respir Rev. 2004;5 Suppl A:S371-6.
9. Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, et al. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics. 2012;130(3):576-84.
10. Huynh NT, Morton PD, Rompre PH, Papadakis A, Remise C. Associations between sleep-disordered breathing symptoms and facial and dental morphometry, assessed with screening examinations. American journal of orthodontics and dentofacial orthopedics : official publication of the American Association of Orthodontists, its constituent societies, and the American Board of Orthodontics. 2011;140(6):762-70.
11. Tapia IE, Marcus CL. Newer treatment modalities for pediatric obstructive sleep apnea. Paediatr Respir Rev. 2013;14(3):199-203.
12. Brouilette R, Hanson D, David R, Klemka L, Szatkowski A, Fernbach S, et al. A diagnostic approach to suspected obstructive sleep apnea in children. The Journal of pediatrics. 1984;105(1):10-4.
13. Zhang G, Spickett J, Rumchev K, Lee AH, Stick S. Snoring in primary school children and domestic environment: a Perth school based study. Respiratory research. 2004;5:19.
14. Corbo GM, Forastiere F, Agabiti N, Pistelli R, Dell'Orco V, Perucci CA, et al. Snoring in 9- to 15-year-old children: risk factors and clinical relevance. Pediatrics. 2001;108(5):1149-54.
15. Ersu R, Arman AR, Save D, Karadag B, Karakoc F, Berkem M, et al. Prevalence of snoring and symptoms of sleep-disordered breathing in primary school children in istanbul. Chest. 2004;126(1):19-24.
16. Johnson EO, Roth T. An epidemiologic study of sleep-disordered breathing symptoms among adolescents. Sleep. 2006;29(9):1135-42.

17. Rosen CL, Larkin EK, Kirchner HL, Emancipator JL, Bivins SF, Surovec SA, et al. Prevalence and risk factors for sleep-disordered breathing in 8- to 11-year-old children: association with race and prematurity. The Journal of pediatrics. 2003;142(4):383-9.
18. Goodwin JL, Babar SI, Kaemingk KL, Rosen GM, Morgan WJ, Sherrill DL, et al. Symptoms related to sleep-disordered breathing in white and Hispanic children: the Tucson Children's Assessment of Sleep Apnea Study. Chest. 2003;124(1):196-203.
19. Polysomnography 2017 [Available from: https://www.thoracic.org/professionals/clinical-resources/sleep/sleep-fragments/images/slide3.jpg.
20. Chan J, Edman JC, Koltai PJ. Obstructive sleep apnea in children. Am Fam Physician. 2004;69(5):1147-54.
21. Spruyt K, Gozal D. Development of pediatric sleep questionnaires as diagnostic or epidemiological tools: a brief review of dos and don'ts. Sleep medicine reviews. 2011;15(1):7-17.
22. Spruyt K, Gozal D. Pediatric sleep questionnaires as diagnostic or epidemiological tools: a review of currently available instruments. Sleep medicine reviews. 2011;15(1):19-32.
23. Spruyt K, Gozal D. Screening of pediatric sleep-disordered breathing: a proposed unbiased discriminative set of questions using clinical severity scales. Chest. 2012;142(6):1508-15.
24. Montgomery-Downs HE, O'Brien LM, Holbrook CR, Gozal D. Snoring and sleep-disordered breathing in young children: subjective and objective correlates. Sleep. 2004;27(1):87-94.
25. Gozal D. Sleep-disordered breathing and school performance in children. Pediatrics. 1998;102(3 Pt 1):616-20.
26. Spruyt K, Verleye G, Gozal D. Unbiased categorical classification of pediatric sleep disordered breathing. Sleep. 2010;33(10):1341-7.
27. Baugh RF, Archer SM, Mitchell RB, Rosenfeld RM, Amin R, Burns JJ, et al. Clinical practice guideline: tonsillectomy in children. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2011;144(1 Suppl):S1-30.
28. Brietzke SE, Gallagher D. The effectiveness of tonsillectomy and adenoidectomy in the treatment of pediatric obstructive sleep apnea/hypopnea syndrome: a meta-analysis. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2006;134(6):979-84.
29. Friedman M, Wilson M, Lin HC, Chang HW. Updated systematic review of tonsillectomy and adenoidectomy for treatment of pediatric obstructive sleep apnea/hypopnea syndrome. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2009;140(6):800-8.
30. Costa DJ, Mitchell R. Adenotonsillectomy for obstructive sleep apnea in obese children: a meta-analysis. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2009;140(4):455-60.
31. Won CH, Li KK, Guilleminault C. Surgical treatment of obstructive sleep apnea: upper airway and maxillomandibular surgery. Proceedings of the American Thoracic Society. 2008;5(2):193-9.
32. Guilleminault C, Lee JH, Chan A. Pediatric obstructive sleep apnea syndrome. Arch Pediatr Adolesc Med. 2005;159(8):775-85.

33. Kureshi SA, Gallagher PR, McDonough JM, Cornaglia MA, Maggs J, Samuel J, et al. Pilot study of nasal expiratory positive airway pressure devices for the treatment of childhood obstructive sleep apnea syndrome. Journal of clinical sleep medicine : JCSM : official publication of the American Academy of Sleep Medicine. 2014;10(6):663-9.
34. Sterni LM, Tunkel DE. Obstructive sleep apnea in children: an update. Pediatr Clin North Am. 2003;50(2):427-43.
35. CPAP.
36. Guilleminault C, Monteyrol PJ, Huynh NT, Pirelli P, Quo S, Li K. Adeno-tonsillectomy and rapid maxillary distraction in pre-pubertal children, a pilot study. Sleep & breathing = Schlaf & Atmung. 2011;15(2):173-7.
37. Guilleminault C, Quo S, Huynh NT, Li K. Orthodontic expansion treatment and adenotonsillectomy in the treatment of obstructive sleep apnea in prepubertal children. Sleep. 2008;31(7):953-7.
38. Huynh NT, Desplats E, Almeida FR. Orthodontics treatments for managing obstructive sleep apnea syndrome in children: A systematic review and meta-analysis. Sleep medicine reviews. 2016;25:84-94.
39. Pirelli P, Saponara M, Guilleminault C. Rapid maxillary expansion in children with obstructive sleep apnea syndrome. Sleep. 2004;27(4):761-6.
40. Villa MP, Rizzoli A, Miano S, Malagola C. Efficacy of rapid maxillary expansion in children with obstructive sleep apnea syndrome: 36 months of follow-up. Sleep & breathing = Schlaf & Atmung. 2011;15(2):179-84.
41. RME.
42. Verhulst SL, Franckx H, Van Gaal L, De Backer W, Desager K. The effect of weight loss on sleep-disordered breathing in obese teenagers. Obesity (Silver Spring). 2009;17(6):1178-83.
43. Kalra M, Inge T, Garcia V, Daniels S, Lawson L, Curti R, et al. Obstructive sleep apnea in extremely overweight adolescents undergoing bariatric surgery. Obes Res. 2005;13(7):1175-9.
44. Kheirandish-Gozal L, Gozal D. Intranasal budesonide treatment for children with mild obstructive sleep apnea syndrome. Pediatrics. 2008;122(1):e149-55.
45. Pereira KD, Roebuck JC, Howell L. The effect of body position on sleep apnea in children younger than 3 years. Archives of otolaryngology--head & neck surgery. 2005;131(11):1014-6.
46. Pereira KD, Rathi NK, Fatakia A, Haque SA, Castriotta RJ. Body position and obstructive sleep apnea in 8-12-month-old infants. International journal of pediatric otorhinolaryngology. 2008;72(6):897-900.
47. Fernandes do Prado LB, Li X, Thompson R, Marcus CL. Body position and obstructive sleep apnea in children. Sleep. 2002;25(1):66-71.
48. Dayyat E, Maarafeya MM, Capdevila OS, Kheirandish-Gozal L, Montgomery-Downs HE, Gozal D. Nocturnal body position in sleeping children with and without obstructive sleep apnea. Pediatr Pulmonol. 2007;42(4):374-9.
49. Camacho M, Certal V, Abdullatif J, Zaghi S, Ruoff CM, Capasso R, et al. Myofunctional Therapy to Treat Obstructive Sleep Apnea: A Systematic Review and Meta-analysis. Sleep. 2015;38(5):669-75.

50. Villa MP, Brasili L, Ferretti A, Vitelli O, Rabasco J, Mazzotta AR, et al. Oropharyngeal exercises to reduce symptoms of OSA after AT. Sleep & breathing = Schlaf & Atmung. 2015;19(1):281-9.
51. Guilleminault C, Huang YS, Monteyrol PJ, Sato R, Quo S, Lin CH. Critical role of myofascial reeducation in pediatric sleep-disordered breathing. Sleep Med. 2013;14(6):518-25.
52. Suen JS, Arnold JE, Brooks LJ. Adenotonsillectomy for treatment of obstructive sleep apnea in children. Archives of otolaryngology--head & neck surgery. 1995;121(5):525-30.
53. Bar A, Tarasiuk A, Segev Y, Phillip M, Tal A. The effect of adenotonsillectomy on serum insulin-like growth factor-I and growth in children with obstructive sleep apnea syndrome. The Journal of pediatrics. 1999;135(1):76-80.
54. Shiomi T, Guilleminault C, Stoohs R, Schnittger I. Obstructed breathing in children during sleep monitored by echocardiography. Acta Paediatr. 1993;82(10):863-71.
55. Amin RS, Kimball TR, Bean JA, Jeffries JL, Willging JP, Cotton RT, et al. Left ventricular hypertrophy and abnormal ventricular geometry in children and adolescents with obstructive sleep apnea. American journal of respiratory and critical care medicine. 2002;165(10):1395-9.
56. Marcus CL, Greene MG, Carroll JL. Blood pressure in children with obstructive sleep apnea. American journal of respiratory and critical care medicine. 1998;157(4 Pt 1):1098-103.
57. Gozal D, Wang M, Pope DW, Jr. Objective sleepiness measures in pediatric obstructive sleep apnea. Pediatrics. 2001;108(3):693-7.
58. Goh DY, Galster P, Marcus CL. Sleep architecture and respiratory disturbances in children with obstructive sleep apnea. American journal of respiratory and critical care medicine. 2000;162(2 Pt 1):682-6.
59. Frank Y, Kravath RE, Pollak CP, Weitzman ED. Obstructive sleep apnea and its therapy: clinical and polysomnographic manifestations. Pediatrics. 1983;71(5):737-42.
60. Brooks LJ, Topol HI. Enuresis in children with sleep apnea. The Journal of pediatrics. 2003;142(5):515-8.
61. Chervin RD, Dillon JE, Bassetti C, Ganoczy DA, Pituch KJ. Symptoms of sleep disorders, inattention, and hyperactivity in children. Sleep. 1997;20(12):1185-92.
62. Ali NJ, Pitson D, Stradling JR. Sleep disordered breathing: effects of adenotonsillectomy on behaviour and psychological functioning. Eur J Pediatr. 1996;155(1):56-62.
63. Ali NJ, Pitson DJ, Stradling JR. Snoring, sleep disturbance, and behaviour in 4-5 year olds. Arch Dis Child. 1993;68(3):360-6.
64. Greenhill L, Puig-Antich J, Goetz R, Hanlon C, Davies M. Sleep architecture and REM sleep measures in prepubertal children with attention deficit disorder with hyperactivity. Sleep. 1983;6(2):91-101.
65. Anuntaseree W, Kuasirikul S, Suntornlohanakul S. Natural history of snoring and obstructive sleep apnea in Thai school-age children. Pediatr Pulmonol. 2005;39(5):415-20.
66. Topol HI, Brooks LJ. Follow-up of primary snoring in children. The Journal of pediatrics. 2001;138(2):291-3.
67. Marcus CL, Hamer A, Loughlin GM. Natural history of primary snoring in children. Pediatr Pulmonol. 1998;26(1):6-11.

68. Li AM, Au CT, Ng SK, Abdullah VJ, Ho C, Fok TF, et al. Natural history and predictors for progression of mild childhood obstructive sleep apnoea. Thorax. 2010;65(1):27-31.
69. Chervin RD, Ellenberg SS, Hou X, Marcus CL, Garetz SL, Katz ES, et al. Prognosis for Spontaneous Resolution of OSA in Children. Chest. 2015;148(5):1204-13.
70. Marcus CL, Moore RH, Rosen CL, Giordani B, Garetz SL, Taylor HG, et al. A randomized trial of adenotonsillectomy for childhood sleep apnea. N Engl J Med. 2013;368(25):2366-76.
71. Guilleminault C, Huang YS, Glamann C, Li K, Chan A. Adenotonsillectomy and obstructive sleep apnea in children: a prospective survey. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2007;136(2):169-75.
72. Tauman R, Gulliver TE, Krishna J, Montgomery-Downs HE, O'Brien LM, Ivanenko A, et al. Persistence of obstructive sleep apnea syndrome in children after adenotonsillectomy. The Journal of pediatrics. 2006;149(6):803-8.
73. Bhattacharjee R, Kheirandish-Gozal L, Spruyt K, Mitchell RB, Promchiarak J, Simakajornboon N, et al. Adenotonsillectomy outcomes in treatment of obstructive sleep apnea in children: a multicenter retrospective study. American journal of respiratory and critical care medicine. 2010;182(5):676-83.
74. Tagaya M, Nakata S, Yasuma F, Mitchell RB, Sasaki F, Miyazaki S, et al. Children with severe or moderate obstructive sleep apnoea syndrome show a high incidence of persistence after adenotonsillectomy. Acta oto-laryngologica. 2012;132(11):1208-14.
75. Truong MT, Woo VG, Koltai PJ. Sleep endoscopy as a diagnostic tool in pediatric obstructive sleep apnea. International journal of pediatric otorhinolaryngology. 2012;76(5):722-7.
76. Lin AC, Koltai PJ. Persistent pediatric obstructive sleep apnea and lingual tonsillectomy. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2009;141(1):81-5.
77. Sato K, Shirakawa T, Sakata H, Asanuma S. Effectiveness of the analysis of craniofacial morphology and pharyngeal airway morphology in the treatment of children with obstructive sleep apnoea syndrome. Dento maxillo facial radiology. 2012;41(5):411-6.
78. Kang KT, Chou CH, Weng WC, Lee PL, Hsu WC. Associations between adenotonsillar hypertrophy, age, and obesity in children with obstructive sleep apnea. PLoS One. 2013;8(10):e78666.
79. Fujioka M, Young LW, Girdany BR. Radiographic evaluation of adenoidal size in children: adenoidal-nasopharyngeal ratio. AJR Am J Roentgenol. 1979;133(3):401-4.
80. Brodsky L, Moore L, Stanievich JF. A comparison of tonsillar size and oropharyngeal dimensions in children with obstructive adenotonsillar hypertrophy. International journal of pediatric otorhinolaryngology. 1987;13(2):149-56.
81. Ameli F, Brocchetti F, Semino L, Fibbi A. Adenotonsillectomy in obstructive sleep apnea syndrome. Proposal of a surgical decision-taking algorithm. International journal of pediatric otorhinolaryngology. 2007;71(5):729-34.
82. Schutz TC, Dominguez GC, Hallinan MP, Cunha TC, Tufik S. Class II correction improves nocturnal breathing in adolescents. Angle Orthod. 2011;81(2):222-8.
83. Herbst Appliance.

84. Hochban W, Brandenburg U. Morphology of the viscerocranium in obstructive sleep apnoea syndrome--cephalometric evaluation of 400 patients. J Craniomaxillofac Surg. 1994;22(4):205-13.
85. Villa MP, Malagola C, Pagani J, Montesano M, Rizzoli A, Guilleminault C, et al. Rapid maxillary expansion in children with obstructive sleep apnea syndrome: 12-month follow-up. Sleep Med. 2007;8(2):128-34.
86. Guilleminault C, Li KK, Khramtsov A, Pelayo R, Martinez S. Sleep disordered breathing: surgical outcomes in prepubertal children. Laryngoscope. 2004;114(1):132-7.
87. Güray E KA. Efffects of Adenoidectomy Dentofacial Structures: A 6-Year Longitudinal Study
World J Orthod. 2002;3:73-81.
88. Smith DF, Dalesio NM, Benke JR, Petrone JA, Vigilar V, Cohen AP, et al. Anthropometric and Dental Measurements in Children with Obstructive Sleep Apnea. Journal of clinical sleep medicine : JCSM : official publication of the American Academy of Sleep Medicine. 2016;12(9):1279-84.
89. Pirila-Parkkinen K, Lopponen H, Nieminen P, Tolonen U, Paakko E, Pirttiniemi P. Validity of upper airway assessment in children: a clinical, cephalometric, and MRI study. The Angle orthodontist. 2011;81(3):433-9.
90. Johal A, Patel SI, Battagel JM. The relationship between craniofacial anatomy and obstructive sleep apnoea: a case-controlled study. J Sleep Res. 2007;16(3):319-26.
91. Ishiguro K, Kobayashi T, Kitamura N, Saito C. Relationship between severity of sleep-disordered breathing and craniofacial morphology in Japanese male patients. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2009;107(3):343-9.
92. Pirila-Parkkinen K, Lopponen H, Nieminen P, Tolonen U, Pirttiniemi P. Cephalometric evaluation of children with nocturnal sleep-disordered breathing. European journal of orthodontics. 2010;32(6):662-71.
93. Johal A, Conaghan C. Maxillary morphology in obstructive sleep apnea: a cephalometric and model study. The Angle orthodontist. 2004;74(5):648-56.
94. Pirila-Parkkinen K, Pirttiniemi P, Nieminen P, Tolonen U, Pelttari U, Lopponen H. Dental arch morphology in children with sleep-disordered breathing. European journal of orthodontics. 2009;31(2):160-7.
95. Aasm. Sleep Related Bruxism. In: Medicine AAoS, editor. ICSD-2 International classification of sleep disorders, 2nd ed: Diagnosis and coding manual. 2nd ed. Westchester, Illinois: American Academy of Sleep Medicine; 2005. p. 189-92.
96. Cheifetz AT, Osganian SK, Allred EN, Needleman HL. Prevalence of bruxism and associated correlates in children as reported by parents. J Dent Child (Chic). 2005;72(2):67-73.
97. Petit D, Touchette E, Tremblay RE, Boivin M, Montplaisir J. Dyssomnias and parasomnias in early childhood. Pediatrics. 2007;119(5):e1016-25.
98. Simola P, Niskakangas M, Liukkonen K, Virkkula P, Pitkaranta A, Kirjavainen T, et al. Sleep problems and daytime tiredness in Finnish preschool-aged children-a community survey. Child: care, health and development. 2010;36(6):805-11.
99. Lavigne GJ, Montplaisir JY. Restless legs syndrome and sleep bruxism: prevalence and association among Canadians. Sleep. 1994;17(8):739-43.
100. Laberge L, Tremblay RE, Vitaro F, Montplaisir J. Development of parasomnias from childhood to early adolescence. Pediat. 2000;106(1):67-74.

101. Ohayon MM, Li KK, Guilleminault C. Risk factors for sleep bruxism in the general population. Chest. 2001;119(1):53-61.
102. Lavigne GJ, Rompre PH, Poirier G, Huard H, Kato T, Montplaisir JY. Rhythmic masticatory muscle activity during sleep in humans. J Dent Res. 2001;80(2):443-8.
103. Lavigne GJ, Huynh N, Kato T, Okura K, Adachi K, Yao D, et al. Genesis of sleep bruxism: motor and autonomic-cardiac interactions. Arch Oral Biol. 2007;52(4):381-4.
104. Carra MC, Huynh N, Morton P, Rompre PH, Papadakis A, Remise C, et al. Prevalence and risk factors of sleep bruxism and wake-time tooth clenching in a 7- to 17-yr-old population. Eur J Oral Sci. 2011;119(5):386-94.
105. Khoury S, Rouleau GA, Rompré PH, Mayer P, Montplaisir J, Lavigne GJ. A significant increase in breathing amplitude precedes sleep bruxism. Chest. 2008;134:332-7.
106. DiFrancesco RC, Junqueira PA, Trezza PM, de Faria ME, Frizzarini R, Zerati FE. Improvement of bruxism after T & A surgery. Int J Pediatr Otorhinolaryngol. 2004;68(4):441-5.
107. Eftekharian A, Raad N, Gholami-Ghasri N. Bruxism and adenotonsillectomy. Int J Pediatr Otorhinolaryngol. 2008;72(4):509-11.
108. Bellerive A, Montpetit A, El-Khatib H, Carra MC, Remise C, Desplats E, et al. The effect of rapid palatal expansion on sleep bruxism in children. Sleep & breathing = Schlaf & Atmung. 2015;19(4):1265-71.
109. Marino A, Malagnino I, Ranieri R, Villa MP, Malagola C. Craniofacial morphology in preschool children with obstructive sleep apnoea syndrome. Eur J Paediatr Dent. 2009;10(4):181-4.
110. Tsuda H, Fastlicht S, Almeida FR, Lowe AA. The correlation between craniofacial morphology and sleep-disordered breathing in children in an undergraduate orthodontic clinic. Sleep & breathing = Schlaf & Atmung. 2011;15(2):163-71.
111. Katz ES, Greene MG, Carson KA, Galster P, Loughlin GM, Carroll J, et al. Night-to-night variability of polysomnography in children with suspected obstructive sleep apnea. The Journal of pediatrics. 2002;140(5):589-94.
112. Tagaya M, Nakata S, Yasuma F, Miyazaki S, Sasaki F, Morinaga M, et al. Relationship between adenoid size and severity of obstructive sleep apnea in preschool children. International journal of pediatric otorhinolaryngology. 2012;76(12):1827-30.
113. Basoglu OK, Tasbakan MS. Gender differences in clinical and polysomnographic features of obstructive sleep apnea: a clinical study of 2827 patients. Sleep & breathing = Schlaf & Atmung. 2017.
114. Franklin KA, Lindberg E. Obstructive sleep apnea is a common disorder in the population-a review on the epidemiology of sleep apnea. Journal of thoracic disease. 2015;7(8):1311-22.
115. Brockmann PE, Koren D, Kheirandish-Gozal L, Gozal D. Gender dimorphism in pediatric OSA: Is it for real? Respiratory physiology & neurobiology. 2016.
116. Goodwin JL, Vasquez MM, Silva GE, Quan SF. Incidence and remission of sleep-disordered breathing and related symptoms in 6- to 17-year old children--the Tucson Children's Assessment of Sleep Apnea Study. The Journal of pediatrics. 2010;157(1):57-61.
117. Sardon O, Perez-Yarza EG, Aldasoro A, Bordoy A, Mintegui J, Emparanza JI. [Obstructive sleep apnea-hypopnea syndrome in children is not associated with obesity]. Archivos de bronconeumologia. 2006;42(11):583-7.

118. Mallory GB, Jr., Fiser DH, Jackson R. Sleep-associated breathing disorders in morbidly obese children and adolescents. The Journal of pediatrics. 1989;115(6):892-7.
119. Kobayashi Y., H. S. The relationship between sleep apnoea, bruxism, and the time it took the patient to seek for treatment in TMD patients. Journal of Oral Rehabilitation. 2002;29(9):885.
120. Yoshida K. A polysomnographic study on masticatory and tongue muscle activity during obstructive and central sleep apnea. Journal of oral rehabilitation. 1998;25(8):603-9.
121. Khoury S, Rouleau GA, Rompre PH, Mayer P, Montplaisir JY, Lavigne GJ. A significant increase in breathing amplitude precedes sleep bruxism. Chest. 2008;134(2):332-7.
122. Lavigne GJ MC. Bruxism. 4th ed. Philadelphia,
Pa: W.B. Saunders Company; 2005.
123. Sjoholm TT, Lowe AA, Miyamoto K, Fleetham JA, Ryan CF. Sleep bruxism in patients with sleep-disordered breathing. Archives of oral biology. 2000;45(10):889-96.
124. Nino G, Singareddy R. Severe onychophagia and finger mutilation associated with obstructive sleep apnea. Journal of clinical sleep medicine : JCSM : official publication of the American Academy of Sleep Medicine. 2013;9(4):379-81.
125. Shine NP, Coates HL, Lannigan FJ. Obstructive sleep apnea, morbid obesity, and adenotonsillar surgery: a review of the literature. International journal of pediatric otorhinolaryngology. 2005;69(11):1475-82.
126. Nolan J, Brietzke SE. Systematic review of pediatric tonsil size and polysomnogram-measured obstructive sleep apnea severity. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2011;144(6):844-50.
127. Ng SK, Lee DL, Li AM, Wing YK, Tong MC. Reproducibility of clinical grading of tonsillar size. Archives of otolaryngology--head & neck surgery. 2010;136(2):159-62.
128. Valera FC, Avelino MA, Pettermann MB, Fujita R, Pignatari SS, Moreira GA, et al. OSAS in children: correlation between endoscopic and polysomnographic findings. Otolaryngology--head and neck surgery : official journal of American Academy of Otolaryngology-Head and Neck Surgery. 2005;132(2):268-72.
129. Dayyat E, Kheirandish-Gozal L, Sans Capdevila O, Maarafeya MM, Gozal D. Obstructive sleep apnea in children: relative contributions of body mass index and adenotonsillar hypertrophy. Chest. 2009;136(1):137-44.
130. Arens R, Marcus CL. Pathophysiology of upper airway obstruction: a developmental perspective. Sleep. 2004;27(5):997-1019.
131. Xu Z, Jiaqing A, Yuchuan L, Shen K. A case-control study of obstructive sleep apnea-hypopnea syndrome in obese and nonobese chinese children. Chest. 2008;133(3):684-9.
132. Katyal V, Kennedy D, Martin J, Dreyer C, Sampson W. Paediatric sleep-disordered breathing due to upper airway obstruction in the orthodontic setting: a review. Aust Orthod J. 2013;29(2):184-92.
133. Flores-Mir C, Korayem M, Heo G, Witmans M, Major MP, Major PW. Craniofacial morphological characteristics in children with obstructive sleep apnea syndrome: a systematic review and meta-analysis. J Am Dent Assoc. 2013;144(3):269-77.
134. Jacobson A. The "Wits" appraisal of jaw disharmony. 1975. American journal of orthodontics and dentofacial orthopedics : official publication of the American Association of Orthodontists, its constituent societies, and the American Board of Orthodontics. 2003;124(5):470-9.

135. Nanda RS, Merrill RM. Cephalometric assessment of sagittal relationship between maxilla and mandible. American journal of orthodontics and dentofacial orthopedics : official publication of the American Association of Orthodontists, its constituent societies, and the American Board of Orthodontics. 1994;105(4):328-44.
136. Kawashima S, Peltomaki T, Sakata H, Mori K, Happonen RP, Ronning O. Absence of facial type differences among preschool children with sleep-related breathing disorder. Acta odontologica Scandinavica. 2003;61(2):65-71.
137. Schiffman PH, Rubin NK, Dominguez T, Mahboubi S, Udupa JK, O'Donnell AR, et al. Mandibular dimensions in children with obstructive sleep apnea syndrome. Sleep. 2004;27(5):959-65.
138. Zucconi M, Caprioglio A, Calori G, Ferini-Strambi L, Oldani A, Castronovo C, et al. Craniofacial modifications in children with habitual snoring and obstructive sleep apnoea: a case-control study. The European respiratory journal. 1999;13(2):411-7.
139. Agren K, Nordlander B, Linder-Aronsson S, Zettergren-Wijk L, Svanborg E. Children with nocturnal upper airway obstruction: postoperative orthodontic and respiratory improvement. Acta oto-laryngologica. 1998;118(4):581-7.
140. Zettergren-Wijk L, Forsberg CM, Linder-Aronson S. Changes in dentofacial morphology after adeno-/tonsillectomy in young children with obstructive sleep apnoea--a 5-year follow-up study. European journal of orthodontics. 2006;28(4):319-26.

Annex 1 : Ethics committee

Le 06 mai 2015

Madame Nelly Huynh
Clinique dentaire
Étage Bloc

OBJET: Titre du projet: NH-12XP-Prévalence: Étude de prévalence de malocclusions dentaires chez les enfants atteints de troubles du sommeil - évaluation de préfaisabilité

No. de dossier: 3639

Responsables du projet: Nelly Huynh Ph. D., chercheure principale. Co-chercheures : Sheila Jacob, MD et Sophie Laberge, MD - Collaborateurs : Andrée Montpetit, DMD, MSc, Jérémie Abikhzer, DMD.

Madame,

L'approbation éthique de votre projet cité en rubrique a été renouvelée par le comité d'éthique de la recherche en date du 06 mai 2015. Vous trouverez ci-joint la liste des documents approuvés ainsi que vos formulaires d'information et de consentement estampillés dont nous vous prions de vous servir d'une copie pour distribution.

Tous les projets de recherche impliquant des sujets humains doivent être réexaminés annuellement et la durée de l'approbation de votre projet sera effective jusqu'au **06 mai 2016**. Notez qu'il est de votre responsabilité de soumettre une demande au Comité pour le renouvellement de votre projet avant la date d'expiration mentionnée. Il est également de votre responsabilité d'aviser le Comité de toute modification à votre projet et/ou tout événement pouvant toucher à la sécurité des participants.

Nous vous souhaitons bonne chance dans la continuité de votre projet et vous prions de recevoir nos meilleures salutations.

Carolina Martin , éthicienne
Responsable du suivi annuel du Comité d'éthique de la recherche

CM/sa

CHU Sainte-Justine
Le centre hospitalier universitaire mère-enfant

Pour l'amour des enfants

Université de Montréal

Comité d'éthique de la recherche
ethique@recherche-ste-justine.qc.ca
Tél. : 514-345-4931 poste 3819
Téléc. : 514- 345-4698

Présidente :
Geneviève Cardinal, avocate
514-345-4931 poste 5102
Genevieve.cardinal@recherche-ste-justine.qc.ca

Vice-président
Patrick Gogognon, éthicien
514-345-4931 poste 5102
Patrick.gogognon@recherche-ste-justine.qc.ca

Responsable des renouvellements :
Carolina Martin, éthicienne
514-345-4931 poste 3012
Carolina.martin@recherche-ste-justine.qc.ca

Agentes de gestion

Nicole Dontigny
Responsable de la coordination
514-345-4931 poste 3819
nicole.dontigny@recherche-ste-justine.qc.ca

Samira Akrab
514-345-4931 poste 3030
Samira.akrab@recherche-ste-justine.qc.ca

Marie-Hélène La France
514-345-4730
marie-helene.la.france@recherche-ste-justine.qc.ca

3175, Côte-Sainte-Catherine
Montréal (Québec)
H3T 1C5

CHU Sainte-Justine
Le centre hospitalier universitaire mère-enfant

Pour l'amour des enfants

Université de Montréal

Comité d'éthique de la recherche
ethique@recherche-ste-justine.qc.ca
Tél. : 514-345-4931 poste 3819
Téléc. : 514- 345-4698

Présidente :
Geneviève Cardinal, avocate
514-345-4931 poste 4342
Genevieve.cardinal@recherche-ste-justine.qc.ca

Vice-président
Patrick Gogognon, éthicien
514-345-4931 poste 3102
Patrick.gogognon@recherche-ste-justine.qc.ca

Responsable des renouvellements :
Carolina Martin, éthicienne
514-345-4931 poste 3912
Carolina.martin@recherche-ste-justine.qc.ca

Agentes de gestion

Nicole Dontigny
Responsable de la coordination
514-345-4931 poste 3819
nicole.dontigny@recherche-ste-justine.qc.ca

Samira Akrah
514-345-4931 poste 3040
Samira.akrah@recherche-ste-justine.qc.ca

Marie-Hélène La France
514-345-4750
marie-helene.la.france@recherche-ste-justine.qc.ca

Liste des documents approuvés par le CÉR

Titre du projet:
NH-12XP-Prévalence: Étude de prévalence de malocclusions dentaires chez les enfants atteints de troubles du sommeil - évaluation de préfaisabilité

No. de dossier: 3639

Date de réapprobation : mercredi 06 mai 2015

Responsables du projet: HUYNH NELLY Ph. D., chercheure principale. Co-chercheures : Sheila Jacob, MD et Sophie Laberge, MD - Collaborateurs : Andrée Montpetit, DMD, MSc, Jérémie Abikhzer, DMD.

Liste:

- Protocole de recherche amendé daté du 14 février 2014

- Protocole de banque non daté, réapprouvé le 06 mai 2015

- Formulaire d'information et de consentement daté du 24 mars 2015 (Version française)

- Formulaire d'information et de consentement daté du 14 novembre 2014 (Version anglaise)

3175, Côte-Sainte-Catherine
Montréal (Québec)
H3T 1C5

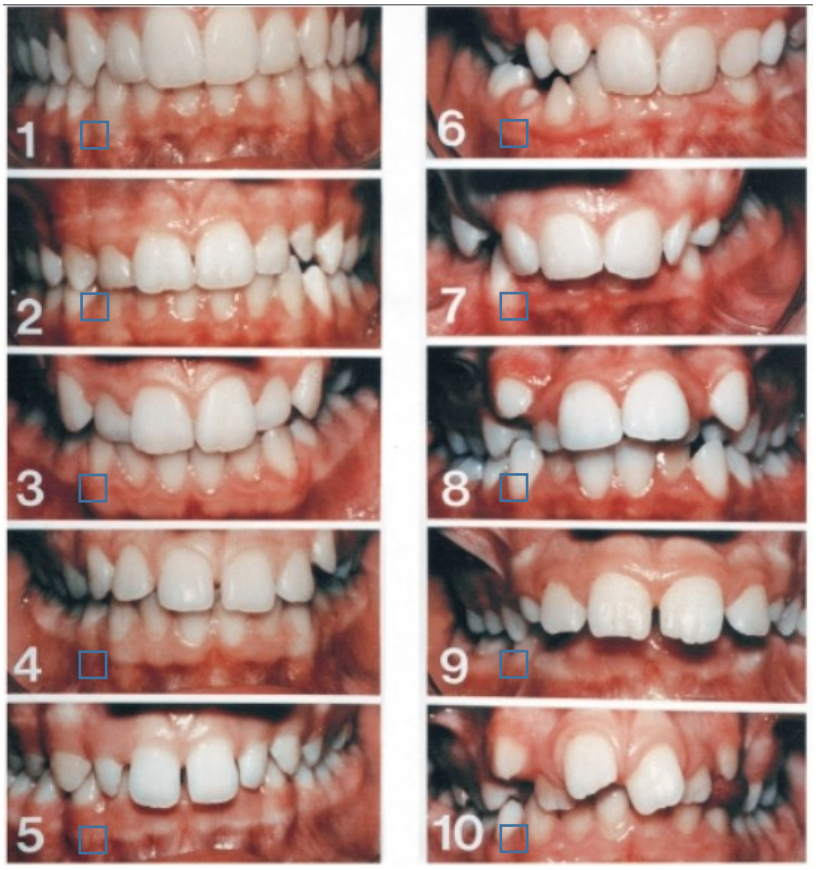
Annex 2 : Clinical exam

Type de cas – Plainte principale			
Origines	Pays d'origine de la mère: _____	Pays d'origine du père_____	
Morphologie (Si limite, choisir mésomorphe)	☐ Ectomorphe	☐ Mésomorphe	☐ Endomorphe
Vue de face			
1. Type facial (Si limite, choisir mésofacial)	☐ Mésofacial ☐ brachyfacial		☐ Dolichofacial
2. Hauteur faciale inférieure	☐ Normale	☐ Augmentée	☐ Diminuée
3. Symétrie (si très légère, choisir symétrique)	☐ Symétrique ☐ Déplacement de la mandibule vers la droite ☐ Déplacement de la mandibule vers la gauche		
4. Médianes (ligne médiane - utiliser aube de la lèvre supérieure)	Supérieure : ☐ sur la ligne médiane du visage ☐ déplacement vers la droite ; ____mm ☐ déplacement vers la gauche ; ____mm Inférieure : ☐ sur la ligne médiane du visage ☐ déplacement vers la droite ; ____mm ☐ déplacement vers la gauche ; ____mm		
5. Incisives visibles au repos :	____mm		
6. Exposition des gencives au sourire :	____mm		
7. Incisives visibles au sourire	____mm		
Vue de profil			
8. Profil facial	☐ Droit	☐ Concave	☐ Convexe

9. Profil maxillaire	☐ Normal ☐ Rétrusif ☐ Protrusif
10. Profil mandibulaire	☐ Normal ☐ Rétrusif ☐ Protrusif
11. Angle naso-labial	☐ Normal 90º-100 º ☐ Aigü (moins que 90 º) ☐ Obtus (plus que 100 º)
Position des lèvres (utiliser définition du plan-E, même si menton retrusif) 12. Par rapport à la ligne esthétique : lèvre supérieure 13. Par rapport à la ligne esthétique : lèvre inférieure	 ☐ Normal ☐ Rétrusive ☐ Protrusive ☐ Normal ☐ Rétrusive ☐ Protrusive
14. Au repos, tension des lèvres (Si légère ouverture sans effort, mettre Non)	☐ Oui ☐ Non
Fonctionnel	
15. Amygdales	☐ Enlevées ☐ 1+ ☐ 2+ ☐ 3+ ☐ 4+ ("*kissing tonsils* ")
16. Historique de la respiration buccale	☐ Oui : **Si oui,** spécifiez : ☐ Durant la journée ☐ Durant la nuit ☐ Non
Intra orale	
17. Habitudes orales	☐ Oui ☐ Non Depuis quand :_____ans
Lesquelles ?	☐ Onychophagie (rongement des ongles) ☐ Succion du pouce/doigt ☐ Bruxisme du sommeil ☐ Mordillement lèvre/joue ☐ Autre : _____
18. Surplomb horizontal (moyenne des deux incisives	*Overjet* : ☐☐☐mm

centrales, labial à labial)	
19. Surplomb vertical (moyenne des deux incisives centrales, labial à labial)	*Overbite* : ☐☐☐ %
20. Béance antérieure (mesurée aux incisives centrales - moyenne)	*Open bite*: ☐☐☐mm
21. Béance postérieure droite (mesurée à la « pire » prémolaire)	☐☐☐mm
22. Béance postérieure gauche (mesurée à la « pire » prémolaire)	☐☐☐mm
23. Odontogramme	<table><tr><td>8</td><td>7</td><td>6</td><td>5</td><td>4</td><td>3</td><td>2</td><td>1</td><td>1</td><td>2</td><td>3</td><td>4</td><td>5</td><td>6</td><td>7</td><td>8</td></tr><tr><td></td><td></td><td></td><td>E</td><td>D</td><td>C</td><td>B</td><td>A</td><td>A</td><td>B</td><td>C</td><td>D</td><td>E</td><td></td><td></td><td></td></tr><tr><td></td><td></td><td></td><td>E</td><td>D</td><td>C</td><td>B</td><td>A</td><td>A</td><td>B</td><td>C</td><td>D</td><td>E</td><td></td><td></td><td></td></tr><tr><td>8</td><td>7</td><td>6</td><td>5</td><td>4</td><td>3</td><td>2</td><td>1</td><td>1</td><td>2</td><td>3</td><td>4</td><td>5</td><td>6</td><td>7</td><td>8</td></tr></table>
24. Occlusion croisée (incluant “edge-to-edge bite”)	Occlusion antérieure : ☐ Oui ; spécifiez le nombre de dents maxillaires impliquées:_____ ☐ Non Occlusion postérieure : ☐ Oui, spécifiez ☐ Unilatéral; spécifiez le nombre de dents maxillaires impliquées:_____ ☐Bilatéral ☐ Non
25. Palais étroit	☐ Oui☐ Non
26. Glissement RC/OC	☐ Oui, spécifiez: ☐Postéro-antérieurement ☐Verticalement ☐Vers la droite ☐Vers la gauche

	☐ Non	
27. Distance inter-molaire (mesurée de "mid-palatal groove" à la marge gingivale)	☐☐☐mm	
28. Distance inter-canine (mesurée de "cusp tip")	☐☐☐mm	
29. Taille de la langue	☐ Normale ☐ Microglossie ☐ Macroglossie	
30. Forme d'arcade (forme de l'os alvéolaire – ne pas considerer les dents)	Haut : ☐ Forme en U ☐ Forme en V Bas : ☐ Forme en U ☐ Forme en V	
31. Profondeur du palais	☐☐☐mm	
32. Stade de dentition	☐ Primaire ☐ Mixte ☐ Permanente (aucune dent primaire)	
33. Classification des molaires (<1/2 cuspide = cl.1)	Permanente : Droit : ☐ I ☐ II ☐ III Gauche : ☐ I ☐ II ☐ III	Mixte/Primaire: Droit : ☐ Mesial ☐ Au même niveau ☐ Distal Gauche: ☐ Mesial ☐ Au même niveau ☐ Distal
34. Classification des canines (<1/2 cuspide = cl.1)	Droit : ☐ I ☐ II ☐ III Gauche : ☐ I ☐ II ☐ III	
35. Analyse de l'espace	☐ Chevauchement: Haut : ☐ <3 mm ☐ 4-9 mm ☐ >10mm Bas : ☐ <3 mm ☐ 4-9 mm ☐ >10mm ☐ Espacement: Haut : ☐ <3 mm ☐ 4-9 mm ☐ >10mm Bas : ☐ <3 mm ☐ 4-9 mm ☐ >10mm	

36. Échelle esthétique IOTN (choisir selon l'attrait du sourire)	

Annex 3 : Adapted Gozal questionnaire

Au cours des 6 derniers mois:

Merci de cocher pour tous les items suivants.

	jamais	**rare** (1 nuit par semaine)	**occasionnelle** (2 nuits par semaine)	**fréquente** (3 à 4 nuits par semaine)	**quasi toujours** (plus de 4 nuits par semaine)
1 – Avez-vous déjà été obligé de secouer votre enfant dans son sommeil pour qu'il se remette à respirer ?	☐	☐	☐	☐	☐
2 – Est-ce que votre enfant s'arrête de respirer pendant son sommeil ?	☐	☐	☐	☐	☐
3 – Est-ce que votre enfant a des difficultés pour respirer pendant son sommeil ?	☐	☐	☐	☐	☐
4 – Est-ce que la respiration de votre enfant pendant son sommeil a déjà été un motif d'inquiétude pour vous ?	☐	☐	☐	☐	☐

	légèrement perceptible ou faible	**modérément fort**	**fort**	**très fort**	**extrêmement fort**
5 – Quelle est l'intensité du bruit de son ronflement ?	☐	☐	☐	☐	☐

	jamais	**rare** (1 nuit par semaine)	**occasionnelle** (2 nuits par semaine)	**fréquente** (3 à 4 nuits par semaine)	**quasi toujours** (plus de 4 nuits par semaine)
6 – À quelle fréquence votre enfant ronfle-t-il ?	☐	☐	☐	☐	☐

Annex 4 : Consent form

CHU Sainte-Justine
Le centre hospitalier universitaire mère-enfant

Pour l'amour des enfants

Université de Montréal

FORMULAIRE D'INFORMATION ET DE CONSENTEMENT

Numéro interne du protocole :	**NH-12XP-Prevalence**
Titre du protocole :	**Étude de prévalence de malocclusions dentaires chez les enfants atteints de troubles du sommeil (évaluation de la préfaisabilité)**

Chercheure responsable :	Nelly Huynh, PhD, (Faculté de médecine dentaire, Université de Montréal)
Co-chercheure :	Dre Sheila Jacob, MD, (CHU Sainte-Justine)
Collaboratrice :	Andrée Montpetit, DMD, MSc (Faculté de médecine dentaire, Université de Montréal)

Ce projet de recherche est financé par le fond de démarrage du Dre Huynh.

La clinique interdisciplinaire des troubles du sommeil (CPITS), en collaboration avec la faculté de médecine dentaire de l'Université de Montréal, participe à des recherches dans le but d'améliorer les traitements chez les enfants souffrant de trouble du sommeil. Nous sollicitons aujourd'hui la participation de votre enfant. Nous vous invitons à lire ce formulaire d'information afin de décider si vous êtes intéressé à ce que votre enfant participe à ce projet de recherche. Il est important de bien comprendre ce formulaire. N'hésitez pas à poser des questions. Prenez le temps nécessaire pour prendre votre décision.

Quelle est la nature de cette recherche ?

Si vous recevez ce formulaire c'est que votre enfant souffre potentiellement de troubles du sommeil et effectuera un enregistrement de sommeil à la clinique interdisciplinaire des troubles du sommeil (CPITS) ou un enregistrement des gaz sanguins à la maison. Outre l'esthétique, les traitements d'orthodontie, guident la croissance faciale pour corriger les déséquilibres crâniofaciaux, améliorent la déglutition, repositionnent la langue et rétablissent la respiration nasale. Nous désirons obtenir plus d'informations sur ces interventions, notamment leur efficacité dans le traitement des troubles respiratoires du sommeil. Nous allons calculer le nombre de patients qui bénéficieraient

potentiellement d'une évaluation dentaire et orthodontique parmi ceux qui ont des troubles respiratoires de sommeil. Cette information sera utilisée pour évaluer la préfaisabilité d'une étude clinique de traitements orthodontiques chez cette population visée. Nous invitons donc votre enfant à recevoir une évaluation dentaire avant son enregistrement du sommeil ou des gaz sanguins pour calculer le pourcentage d'enfant se présentant à la CPITS avec une malocclusion dentaire (mauvais positionnement entre les dents du haut et celles du bas). Pour ce faire, nous désirons recruter 40 enfants ayant des troubles respiratoires de sommeil.

Comment se déroulera le projet ?

Lors de la visite de votre enfant pour son enregistrement de sommeil ou des gaz sanguins, une évaluation dentaire sera faite à la CPITS par un membre de l'équipe de recherche. Cette évaluation consiste à une observation dentaire visuelle, durera environ 15 minutes et sera faite durant votre temps d'attente pour voir l'inhalothérapeute. Un membre de l'équipe de recherche notera différentes mesures sur le positionnement des dents de votre enfant à partir de son observation visuelle. Aucune visite supplémentaire n'est nécessaire puisque l'évaluation dentaire s'effectuera durant votre rendez-vous à la CPITS.

De plus, l'équipe de recherche consultera le dossier médical de votre enfant pour obtenir les informations pertinentes à cette recherche dont les résultats de l'enregistrement du sommeil ou des gaz sanguins effectué dans le cadre usuel de son traitement à la CPITS du CHU Sainte-Justine.

Quels sont les avantages et bénéfices ?

Dans le cadre de votre participation à ce projet, vous recevrez de l'information sur la dentition de votre enfant. Tout patient présentant une malocclusion recevra la recommandation d'aller consulter un orthodontiste de leur choix.

La participation de votre enfant à cette étude sera bénéfique à l'avancement général des connaissances et pour évaluer des possibilités de traitements orthodontiques pour les enfants souffrants de troubles du sommeil.

Quels sont les inconvénients et les risques ?

Aucun risque n'est associé à cette étude.

Comment la confidentialité est-elle assurée ?

Tous les renseignements obtenus sur votre enfant pour ce projet de recherche seront confidentiels, à moins d'une autorisation de votre part ou d'une exception de la loi. Pour ce faire, ces renseignements seront codifiés par un code alphanumérique. La clé du code, reliant son nom au dossier de recherche, sera conservée par la chercheure responsable. Les dossiers de recherche seront conservés pendant 7 années après la fin

Consentement et assentiment

On m'a expliqué la nature et le déroulement du projet de recherche. J'ai pris connaissance du formulaire de consentement et on m'en a remis un exemplaire. J'ai eu l'occasion de poser des questions auxquelles on a répondu à ma satisfaction. Après réflexion, j'accepte que mon enfant participe à ce projet de recherche. J'autorise l'équipe de recherche à consulter le dossier médical de mon enfant pour obtenir les informations pertinentes à ce projet.

Prénom et nom du participant (Lettres moulées)	Signature du participant	Date

Assentiment verbal de l'enfant incapable de signer mais capable de comprendre la nature de ce projet: oui___ non___

Nom du parent ou tuteur (Lettres moulées)	Consentement (signature)	Date

J'ai expliqué au participant et/ou à son parent/tuteur tous les aspects pertinents de la recherche et j'ai répondu aux questions qu'ils m'ont posées. Je leur ai indiqué que la participation au projet de recherche est libre et volontaire et que la participation peut être cessée en tout temps.

Nom de la personne qui a obtenu le consentement (Lettres moulées)	Signature	Date