PARENTAL KNOWLEDGE OF AND SCREENING FOR PEDIATRIC SLEEP DISORDERED BREATHING IN ORTHODONTIC PRACTICE

By

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DEDICATION

Dedicated to my wife Melisa and my daughters Ewa and Emilia, who are the reason for everything I do. Also dedicated to my Father who made me into the person I am today.

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Review of the Literature

Sleep is a complicated process. Significant metabolic, neurologic and respiratory changes all occur as we move through the stages of sleep. During sleep, most body systems are in an anabolic, or rebuilding state. These restorative processes are necessary to maintain mood, cognitive function and the immune system. There are two main phases of sleep; non-rapid eye movement sleep, or NREM, and rapid eye movement sleep, or REM. NREM sleep can be further broken down to N1, N2, and N3; the latter also called slow wave sleep (SWS). These stages of sleep exhibit unique brain activity and can be differentiated with an EEG during sleep (Carskadon et al 1978).

Respiration During Sleep

Respiration also changes during sleep, and not for the better. In 2001 Marcus stated: "We all breathe better awake than asleep". Lopes et al in 1983 showed that upper airway resistance more than doubles during NREM sleep. Then during REM sleep the drive to breathe is decreased and thus the rate of respiration becomes erratic. To compound these issues in a younger population, adolescents exhibit an increase in lymphoid tissue until around age twelve, which can also increase upper airway resistance and make breathing during sleep difficult. Finally, the ventilatory drive, or the drive to breathe, also decreases throughout childhood and adolescence during sleep

(Marcus 2001). It is by these mechanisms that breathing while sleeping is more difficult even, in the absence of any pathology.

Sleep Disordered Breathing

Sleep disordered breathing, or SDB, is a broad term that includes several disorders; this group of disorders may also be referred to as sleep related breathing disorders. It can include obstructive sleep apnea, central sleep apnea, upper airway resistance syndrome and others. The most common of the sleep related breathing disorders is obstructive sleep apnea, or OSA.

OSA is defined by the obstruction of the airway during sleep resulting in a partial or complete cessation in breathing. In order to restore the airway patency, the subject has an arousal, which interrupts the normal sleep cycle. This prevents many of the restorative processes characteristic of normal sleep from being completed properly. The studies included in this review may be focused on OSA specifically, or on SDB more broadly. Any research pertinent to our topic is included, and the focus of each paper is represented accordingly.

Airway Physiology

In 2004, Arens et al described the dynamics of how and why the airway collapses in individuals suffering from OSA. They called this concept

"critical closing pressure" or P_{crit}. The upper respiratory system has three anatomic parts; a collapsible portion in the middle, the downstream portion that is the hypopharanx, and an upstream portion that is the nasal airway. In the adult, the resistance and diameter of the upstream and downstream portions are fixed. The pressure in the upstream area is similar to atmospheric pressure, and the pressure in the downstream portion is created by the action of the diaphragm. In a normal subject where nasal resistance is low, the downstream pressure will never come near the P_{crit} and the airflow will not be limited. However, if the downstream pressure falls below P_{crit}, the airflow is then determined by the resistance of the upstream portion and by P_{crit} itself. If the pressure upstream falls below the P_{crit}, then the airflow becomes zero and the middle part of the airway collapses.

It is important to understand that P_{crit} is a dynamic measure that can be changed by morphology and neuromuscular activation. Understanding this concept highlights why OSA should be thought of as a neurogenic failure, because if it were completely morphological, people afflicted could not breathe while awake or asleep. That is not to say that it cannot be influenced or exacerbated by structural deficiencies, but we cannot point to these issues as the cause of the disorder.

Complications

Complications of SDB in children cover a broad range. Luckily, many of the more severe outcomes are much less common thanks to early diagnosis and treatment. Conditions such as cor pulmonale and failure to thrive can be attributed to SDB, but are rarely seen today (Marcus 2001). In a systematic review of the literature in 2009, Bonuck et al found that standardized height, weight and growth factor production increased significantly after adenotonsillectomy which is the first line treatment for OSA and will be discussed later. In 13 of the 20 studies reviewed, SDB was an inclusion criterion. This would suggest that SDB may have an effect on normal growth.

Additional evidence shows a correlation between pediatric SDB and cognitive deficiencies. In 2004 O'Brien et al showed that even primary snoring, which is on the more mild end of the SDB spectrum has significant consequences for cognition. The patients identified as snorers in this study were shown to score lower on standardized tests designed to assess language and visuospatial ability. In 1998, Gozal showed that first grade students in the bottom 10% of their class had a much higher prevalence of SDB (18.1%) than the general population. Children in this study who underwent treatment also showed significant improvement in grades while the untreated group had no change.

In 2012, Young et al completed a review of the literature regarding sleep disorders and ADHD. They found that up to 30% of children diagnosed with ADHD had symptoms of sleep disorders. They state that while the relationship between the two is unclear, there are a large number of children who are misdiagnosed with ADHD who have a sleep disorder as their primary ailment. Of note, this study included all sleep disorders and not just sleep related breathing disorders. This finding is corroborated by another study where they state that hyperactivity is the behavioral issue that is most commonly associated with SDB (Marcus et al 2012).

Diagnosis

The gold standard for the diagnosis of OSA in children is polysomnography, or PSG. Specifically, nocturnal, attended laboratory PSG is the gold standard (Poels et al 2003). This test provides objective assessment of disturbances in breathing and sleep patterns. This allows for the severity of the OSA to be more accurately determined as opposed to just a positive or negative output. The severity of the condition is important in determining the treatment and expectations for outcomes. However, PSG is an expensive and cumbersome test, and it is not feasible to test every child, thus more precise screening methods are evolving (Moss et al 2005).

Portable monitoring is the use of a take-home device that is worn while sleeping in the patient's home. These devices are classified based on the

number of leads that gather information during the night (Collop et al 2011). Research has shown that quality sleep data can be gathered using these devices, and they may be used for clinical diagnoses in certain situations (Collop et al 2011). The downside of a portable monitor is that there are no EEG readings so the device cannot reliably distinguish between the stages of sleep. Additionally, since the test is taken at home and not monitored by a technician, a dislodged or misplaced lead may make the data unusable (Tan et al 2015). The positive aspects are that they are much cheaper to administer, and the patient is sleeping in his/her own bed, which should give a more accurate assessment of normal sleep patterns.

There have been attempts to diagnose SDB using completely non-invasive methods. Chervin et al published a sleep questionnaire in 2000 that performed better than analyzing medical history alone. The pediatric sleep questionnaire, or PSQ, is a 22-question survey that was built out of a larger bank of questions dealing with sleep health. They had 54 subjects with a sleep related breathing disorder diagnosed with a PSG, and 108 subjects who were presumed not to have a sleep related breathing disorder, though this was not certain. They had the subjects answer a large number of questions and then analyzed the responses. They removed questions that had low odds ratios or ones that had lower ratios than similar questions in one half of the SDB group.

They ended with 22 questions and dichotomized the questionnaire as positive or negative if more or less than 33% of the questions had been answered positively. In the group they used to create the 22 questions, they found a sensitivity and specificity of 85% and 87%. When they applied these criteria to the other half of the SDB group they found a sensitivity and specificity of 81% and 87%. The same questionnaire was re-validated in a later study (Chervin et al 2007), and it showed a sensitivity of 78% and a specificity of 72% in another group of children who had been diagnosed with SDB on a PSQ. The final outcome of the study was that while the PSQ was not sufficient to diagnose a sleep related breathing disorder in clinical practice, it was a good tool for epidemiological research when a PSG is not feasible.

In 2000, Preutthipan et al used a 13-question sleep survey to determine the presence or severity of OSA in children. They found that parents of children with severe OSA were more likely to observe loud snoring or cyanosis, and were more likely to watch their children sleep or shake them at night to get then to breathe. However, none of these questions alone or combinations of them had sufficient sensitivity or specificity to diagnose OSA. In 2005 Goodwin et al published a similar study using a 10-question survey to determine if certain questions regarding sleep symptoms could predict OSA in children. They found that snoring, daytime sleepiness, and learning issues had a high specificity but low sensitivity for the diagnosis of OSA.

According to Marcus et al in 2012, studies have looked at the potential utility of using radiographic imaging as a diagnostic tool for OSA. There is some evidence that airway constriction on a lateral radiograph increases the probability of predicting OSA diagnosed with a PSG. None of the studies reviewed reported any positive or negative predictive values. As discussed above, as OSA is mainly a neurogenic failure, it follows that static radiographic images would have minimal predictive value.

In the past, positive results using nocturnal oximetry has been used as an effective screening tool for OSA. The use of this technique was based on a single study done by Brouillette et al in 2000. They reported that if a positive trend was noted in the oximetry reading there is a 97% positive predictive value for OSA. A more recent article by Kirk et al found that nocturnal oximetry desaturation index in children over 4 had poor agreement with a PSG established AHI. While there may be utility as a screening tool, in 2001 Marcus et al stated that oximetry alone is insufficient for the diagnosis of OSA in children.

Epidemiology

The prevalence of pediatric OSA has been reported in several studies with a range of 0.8%-24% (Marcus et al 2012). The reason for such a broad range is due to the different ways that OSA is diagnosed, and the different criteria used. In 2003, O'Brien et al completed 700 PSG studies on children

aged 5-12. They determined that moderate SDB (AHI $\geq$ 5) had a prevalence of 1.2%. There are few studies that use a PSG for diagnosis and as stated above, there are no agreed upon criteria for the true diagnosis of OSA in children using alternative techniques. In 2003, Rosen et al used a portable monitor to test for SDB in 850 children aged 8-11. They reported a prevalence rate of 2.2% using AHI $\geq$ 5 in this population. In 2008 Lumeng and Chervin reviewed the available epidemiological data on pediatric OSA and reported a prevalence of 1%-4% when actual diagnostic tests were used. They also report that pediatric SDB is likely under diagnosed and under treated in the population.

Rorhra et al (2016) used the Pediatric Sleep Questionnaire developed by Chervin et al (2007), described above, to estimate the number of orthodontic patients potentially at risk for SDB. They gave the questionnaire to 302 patients coming to the orthodontic department at Case Western Reserve University for treatment in 2015. When at least 33% of the answered questions were positive, that patient was determined to be high risk for SDB. They reported that 7.3% of this population was at risk for SDB.

Treatment

The first line treatment for OSA in children is adenotonsillectomy, or AT (Suen et al 1995). In a study where 26 subjects had pre and post AT PSG, Suen et al showed that all subjects exhibited a reduced RDI, and all except 4

had a post operative RDI below 5. Other proposed treatments for OSA in children include CPAP, which is the gold standard in adults and corticosteroids to reduce the size of the lymphoid tissue in the airway (Marcus et al 2012).

In 2015, Villa et al completed a PSG study on 40 young children (ages 4.3-10.5) who were in need of palatal expansion and had symptoms of OSA. They expanded the patients until the palatal cusps of the upper molars were in contact with the buccal cusps of the lower molars. They then kept the expander in place for 12 months at which point they had the patients undergo another PSG. They showed that rapid maxillary expansion reduced PSG-measured AHI by more than 20% in 34 of the 40 subjects studied, though it should be noted that there was no control group studied.

Parental Knowledge

Another complicating factor regarding the recognition and diagnosis of pediatric SDB is that it requires a parent or caregiver to recognize potential signs and symptoms. Parental knowledge of children's sleep has been found to be quite poor (McDowall et al 2016). In 2003, Blunden et al surveyed parents and physicians in Australian primary care medical offices. They asked the parents of children younger than 16 to complete a survey prior to their visit asking about snoring and other sleep breathing related questions. They then asked the physician about the topic of discussion during the visit. They

found that over 80% of the parents who reported snoring on the questionnaire did not discuss the snoring with the physician at the visit. This is important because it is recommended that children with primary snoring have an objective evaluation for SDB (Brockmann et al 2013). This underreporting and lack of parent knowledge may be contributing to the overall under diagnosis of this disorder (Lumeng and Chervin 2008).

In 2007, Strocker and Shapiro completed a cross sectional study of 584 parents who were over 18 and had a child who was younger than 18. Of the sample, 261 of the parents had a child who underwent AT, and 323 had a child who had not undergone the same procedure. The parents responded to a voluntary online questionnaire regarding different issues related to pediatric sleep and sleep disordered breathing. They found that 95% of parents viewed pediatric OSA as a "serious condition", but only 15% considered themselves knowledgeable about the disorder. 68% of the parents who had a child that underwent AT recognized snoring as a symptom of SDB, while only 58% of the parents with a child who had not undergone AT recognized this. While this difference was not statistically significant, it does demonstrate some improvement in knowledge for parents of patients who had been treated for SDB. Additionally, only 38% of parents recognized that daytime irritability could be a sequelae of SDB.

In 2011, Owens et al carried out a cross sectional study of caregivers having at least one child between the ages of three months and twelve years.

253 parents attending a children's museum completed surveys; 96% of the caregivers reported completing college. The survey contained a 10-question section on parental knowledge of sleep. 35% of respondents answered more than half of these questions incorrectly. It should be noted that none of the questions in this study were related to sleep disordered breathing, though the lack of knowledge about healthy sleep behavior is important to note.

Also in 2011, Owens and Jones surveyed 184 parents in the waiting room of a pediatric primary care facility. They included parents of children from 3 months to 12 years. The survey was the same as in the above study by Owens et al. The results of this study were similar; they found that parents in general had a poor understanding of healthy sleep behavior in children and adolescents. Of note, more than half of the respondents thought being underweight was a consequence of poor sleep, and more than half also though that snoring was a sign of quality sleep. They reported that questions about the consequences of unhealthy sleep were answered incorrectly more often than questions about healthy sleep.

In 2014 Wilson et al carried out a randomized trial of an educational intervention. They randomly split families with pre-school children into control and intervention groups. The intervention families were given a questionnaire to assess knowledge, attitudes, self-efficacy and beliefs (KASB) regarding healthy sleep practices for their preschool aged child. The parents were then shown a 45-minute educational video about healthy sleep and the

questionnaire was given again immediately following the educational intervention. The questionnaire was given a final time a month later. The findings show that there was a significant increase in KASB at the immediate post-intervention time point, but the increase has dissipated by the follow-up one month later.

In 2013, Jones et al also tested the value of educational intervention with regard to parental knowledge of sleep health for their children. They approached parents in the waiting room of a pediatric primary care facility and administered a questionnaire, followed by an informational brochure followed again by the same questionnaire. They also found that there was a significant increase in parental knowledge of healthy sleep practices and as well as an increase in the number of parents who plan to change their child's sleep behaviors.

A recent systematic review (McDowall et al 2016) found only a single study (Strocker and Shapiro 2007) that focused specifically on parent knowledge of signs, symptoms and consequences of pediatric SDB. As described above, the parental understanding of these concepts was considered very poor. The overall findings of the systematic review show that parental knowledge of healthy sleep and of SDB is very low. They also report that educational interventions appear to have at least a transitory increase in parental knowledge of these topics.

Summary

The results of this literature review have shown that pediatric sleep disordered breathing is a significant health concern. The prevalence is likely somewhere between 1% and 5% of the population and a similar prevalence in an orthodontic treatment seeking population. The disorder is likely under diagnosed. The definitive diagnosis of this disorder is difficult and expensive. Parental knowledge of SDB is generally low. Efficient screening methods would be of great value to orthodontic practices.

Literature References

Arens, R., & Marcus, C. L. (2004). Pathophysiology of upper airway obstruction: a developmental perspective. *Sleep*, *27*(5), 997–1019. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/15453561

Blunden, S., Lushington, K., Lorenzen, B., Wong, J., Balendran, R., & Kennedy, D. (2003). Symptoms of Sleep Breathing Disorders in Children Are Underreported by Parents at General Practice Visits. *Sleep and Breathing*, *7*(4), 167–176. http://doi.org/10.1007/s11325-003-0167-8

Bonuck, K. A., Freeman, K., & Henderson, J. (2009). Growth and growth biomarker changes after adenotonsillectomy: systematic review and meta-analysis. *Archives of Disease in Childhood*, *94*(2), 83–91. http://doi.org/10.1136/adc.2008.141192

Brockmann, P. E., Schaefer, C., Poets, A., Poets, C. F., & Urschitz, M. S. (2013). Diagnosis of obstructive sleep apnea in children: a systematic review. *Sleep Medicine Reviews*, *17*(5), 331–40. http://doi.org/10.1016/j.smrv.2012.08.004

Brouillette, R. T., Morielli, A., Leimanis, A., Waters, K. A., Luciano, R., & Ducharme, F. M. (2000). Nocturnal pulse oximetry as an abbreviated testing modality for pediatric obstructive sleep apnea. *Pediatrics*, *105*(2), 405–12. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/10654964

Carskadon MA, K, H., WC, D., C, G., FB, S., & TF, A. (1978). Respiration during sleep in children. *The Western Journal of Medicine*, *128*(6), 477–481.

Chervin, R. D., Weatherly, R. A., Garetz, S. L., Ruzicka, D. L., Giordani, B. J., Hodges, E. K., … Guire, K. E. (2007). Pediatric Sleep Questionnaire Prediction of Sleep Apnea and Outcomes. *Arch Otolaryngol Head Neck Surg*, *133*, 216–222.

Collop, N. A., Tracy, S. L., Kapur, V., Mehra, R., Kuhlmann, D., Fleishman, S. A., & Ojile, J. M. (2011). Obstructive Sleep Apnea Devices for Out-Of-Center (OOC) Testing: Technology Evaluation. *Journal of Clinical Sleep Medicine*. http://doi.org/10.5664/jcsm.1328

Goodwin, J. L., Kaemingk, K. L., Mulvaney, S. A., Morgan, W. J., & Quan, S. F. (2005). Clinical screening of school children for polysomnography to detect sleep-disordered breathing--the Tucson Children's Assessment of Sleep Apnea study (TuCASA). *Journal of Clinical Sleep Medicine : JCSM : Official Publication of the American Academy of Sleep Medicine*, *1*(3), 247–54. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/16429591

Gozal, D. (1998). Sleep-disordered breathing and school performance in children. *Pediatrics*, *102*(3 Pt 1), 616–20. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/9738185

Jones, C. H., Owens, J. A., & Pham, B. (2013). Can a brief educational intervention improve parents' knowledge of healthy children's sleep? A pilot-test. *Health Education Journal*, *72*(5), 601–610. http://doi.org/10.1177/0017896912464606

Kirk, V. G., Bohn, S. G., Flemons, W. W., & Remmers, J. E. (2003). Comparison of Home Oximetry Monitoring With Laboratory Polysomnography in Children*. *Chest*, *124*(5), 1702–1708. http://doi.org/10.1378/chest.124.5.1702

Lopes, J. M., Tabachnik, E., Muller, N. L., Levison, H., & Bryan, A. C. (1983). Total airway resistance and respiratory muscle activity during sleep. *Journal of Applied Physiology: Respiratory, Environmental and Exercise Physiology*, *54*(3), 773–7. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/6841222

Lumeng, J. C., & Chervin, R. D. (2008). Epidemiology of pediatric obstructive sleep apnea. *Proceedings of the American Thoracic Society*, *5*(2), 242–52. http://doi.org/10.1513/pats.200708-135MG

Marcus, C. L., Brooks, L. J., Draper, K. A., Gozal, D., Halbower, A. C., Jones, J., … Kelly, J. (2012). Diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*, *130*(3), e714-55. http://doi.org/10.1542/peds.2012-1672

Marcus, C. L. (2001). Sleep-disordered Breathing in Children. *American Journal of Respiratory and Critical Care Medicine*, *164*(1), 16–30. http://doi.org/10.1164/ajrccm.164.1.2008171

Mcdowall, P. S., Galland, B. C., Campbell, A. J., & Elder, D. E. (n.d.). Parent knowledge of children's sleep: A systematic review. http://doi.org/10.1016/j.smrv.2016.01.002

Moss, D., Urschitz, M. S., Von Bodman, A., Eitner, S., Noehren, A., Urschitz-Duprat, P. M., ... Poets, C. F. (2005). Reference Values for Nocturnal Home Polysomnography in Primary Schoolchildren. *Pediatric Research*, *58*(5), 958–965. http://doi.org/10.1203/01.PDR.0000181372.34213.13

O'Brien, L. M., Mervis, C. B., Holbrook, C. R., Bruner, J. L., Klaus, C. J., Rutherford, J., ... Gozal, D. (2004). Neurobehavioral implications of habitual snoring in children. *Pediatrics*, *114*(1), 44–9. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/15231906

Owens, J. A., & Jones, C. (2011). Parental Knowledge of Healthy Sleep in Young Children: Results of a Primary Care Clinic Survey. *Journal of Developmental & Behavioral Pediatrics*, *32*(6), 447–453. http://doi.org/10.1097/DBP.0b013e31821bd20b

Owens, J.A. Jones, C, & Nash, R. (2011). Caregivers' Knowledge, Behavior, and Attitudes Regarding Healthy Sleep in Young Children. *Journal of Clinical Sleep Medicine*, *7*(4), 345–350. http://doi.org/10.5664/jcsm.1186

Poels, P. J. P., Schilder, A. G. M., van den Berg, S., Hoes, A. W., Joosten, K. F. M., D, G., ... RT, B. (2003). Evaluation of a New Device for Home Cardiorespiratory Recording in Children. *Archives of Otolaryngology–Head & Neck Surgery*, *129*(12), 1281. http://doi.org/10.1001/archotol.129.12.1281

Preutthipan, A., Chantarojanasiri, T., Suwanjutha, S., & Udomsubpayakul, U. (2000). Can parents predict the severity of childhood obstructive sleep apnoea? *Acta Paediatrica (Oslo, Norway : 1992)*, *89*(6), 708–12. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/10914968

Rorha, A., Hans, M. . (2016). *Prevalence of Sleep disordered Breathing in an Orthodontic treatment seeking Population* Unpublished master's thesis, Case Western Reserve University, Cleveland, Ohio.

Rosen, C. L., Larkin, E. K., Kirchner, H. L., Emancipator, J. L., Bivins, S. F., Surovec, S. A., ... Redline, S. (n.d.). PREVALENCE AND RISK FACTORS FOR SLEEP-DISORDERED BREATHING IN 8-TO 11-YEAR-OLD CHILDREN: ASSOCIATION WITH RACE AND PREMATURITY. http://doi.org/10.1067/mpd.2003.28

Ross, K. R., & Rosen, C. L. (2014). Sleep and respiratory physiology in children. *Clinics in Chest Medicine*, *35*(3), 457–67. http://doi.org/10.1016/j.ccm.2014.06.003

Schreck, K. A., & Richdale, A. L. (2011). Knowledge of childhood sleep: a possible variable in under or misdiagnosis of childhood sleep problems. *Journal of Sleep Research*, *20*(4), 589–597. http://doi.org/10.1111/j.1365-2869.2011.00922.x

Stores, R., & Stores, G. (2004). Evaluation of Brief Group-Administered Instruction for Parents to Prevent or Minimize Sleep Problems in Young Children with Down Syndrome. *Journal of Applied Research in Intellectual Disabilities*, *17*(2), 61–70. http://doi.org/10.1111/j.1360-2322.2004.00174.x

Strocker, A. M., & Shapiro, N. L. (2007). Parental understanding and attitudes of pediatric obstructive sleep apnea and adenotonsillectomy. *International Journal of Pediatric Otorhinolaryngology*, *71*(11), 1709–1715. http://doi.org/10.1016/j.ijporl.2007.07.016

Suen, J. S., Arnold, J. E., & Brooks, L. J. (1995). Adenotonsillectomy for treatment of obstructive sleep apnea in children. *Archives of Otolaryngology--Head & Neck Surgery*, *121*(5), 525–30. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/7727086

Tan, H.-L., Kheirandish-Gozal, L., & Gozal, D. (2015). Pediatric Home Sleep Apnea Testing Slowly Getting Th ere! *CHEST*, *148*, 1382–1395. http://doi.org/10.1378/chest.15-1365

Villa, M. P., Rizzoli, A., Rabasco, J., Vitelli, O., Pietropaoli, N., Cecili, M., … Malagola, C. (2015). Rapid maxillary expansion outcomes in treatment of obstructive sleep apnea in children. *Sleep Medicine*, *16*(6), 709–16. http://doi.org/10.1016/j.sleep.2014.11.019

Wilson, K. E., Miller, A. L., Bonuck, K., Lumeng, J. C., & Chervin, R. D. (2014). Evaluation of a Sleep Education Program for Low-Income Preschool Children and Their Families. *SLEEP*. http://doi.org/10.5665/sleep.3774

Young, S., Yoon, R., Jain, U., & Shapiro, C. (2012). Sleep in attention-deficit/hyperactivity disorder in children and adults: Past, present, and future. *Sleep Medicine Reviews*, *16*, 371–388. http://doi.org/10.1016/j.smrv.2011.07.001

Statement of Thesis

The study consists of two sections; the first is a qualitative section where the objective of this study is to determine the level of knowledge obtained by parents of children who have been identified as high risk for sleep disordered breathing. The second is a quantitative section where the objective is to assess the agreement between answers on similar questions appearing on both the existing health history and the separate sleep-specific PSQ.

Null hypothesis: There is no agreement between answers to similar questions on the health history and the PSQ

Alternative hypothesis: There is agreement between answers to similar questions on the health history and the PSQ

Parental Knowledge of and Screening for Sleep Disordered Breathing in Orthodontic Practice

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ABSTRACT

Objective: To evaluate the level of knowledge of parents whose children have been identified as high risk for sleep disordered breathing using the Pediatric Sleep Questionnaire (PSQ) in an orthodontic care setting; and to assess agreement between routine health history questions (HHQ) concerning sleep and the PSQ screening score. **Materials and Methods:** Twenty-two patients with high risk PSQ scores and complete HHQ were identified. Sixteen parents were interviewed about their experience having a child classified as high risk for SDB. Interviews were audio- recorded and subsequently transcribed to identify themes. Forty- four low-risk patients were matched on age with the 22 high-risk patients. Kappa and percent agreement between the PSQ and the health history questions were calculated. **Results:** Parental knowledge of sleep disordered breathing and understanding of the "high-risk" classification was poor. None of the 22 patients followed up on a referral to a sleep physician. Agreement between similar questions on the PSQ and the health history form were generally low and no combination of HHQ questions alone could predict the outcome on the PSQ. However, two questions from the PSQ combined with one from the HHQ were 100% sensitive for predicting high risk for SDB among these patients. **Conclusions:** Informing parents of "high-risk" PSQ scores did not result in significant follow-up treatment for SDB. Using three questions about sleep on

a routine HH would likely identify all high-risk PSQ patients thus minimizing the use of the full PSQ in predominantly low risk populations.

Keywords: Sleep disordered breathing, Obstructive sleep apnea, Pediatric sleep questionnaire, Parental knowledge

Introduction

Sleep disordered breathing, or SDB, in children is a serious health issue that is under diagnosed in the population (Lumeng and Chervin, 2008). In 2012 Young et al discussed how the symptoms of ADHD and SDB have significant overlap and postulated that many children diagnosed with ADHD may actually suffer from SDB. As awareness of the disorder in children has increased, attempts to screen patients for the disorder are increasing particularly in dental and orthodontic offices. Given that sleep disordered breathing in children is not often a specialty of the dentist or orthodontist, specific tools with guidelines for referral are sought. Research has shown that health history and clinical signs and symptoms of sleep-disordered breathing are insufficient to diagnose the disorder in children (Goodwin, 2005). However, a discussion about the presence of symptoms would be sufficient to justify a referral to a sleep physician for further testing, though few dentists or orthodontists do not regularly have this conversation with patients.

A questionnaire known as the pediatric sleep questionnaire, or PSQ, has been validated against overnight polysomnography (PSG) and found to have sufficient sensitivity and specificity to be used as a tool for epidemiological research (Chervin et al 2007) as well as a screening tool to assist with a decision on when to refer a patient to a sleep medicine specialist. It is relatively simple to administer and score, with easy to follow guidelines for referral to a sleep specialist. The PSQ consists of twenty-two

yes/no questions and if the patient answers 33% or more of the questions with a "yes", they are considered high-risk (Chervin et al, 2007) and a referral to sleep medicine is given.

Research has also shown that parental knowledge of pediatric sleep disordered breathing is quite low, and is only temporarily increased with current educational interventions (McDowall et al 2015). Efficient screening methods delivered with personalized information may enhance parent knowledge about their child's SDB and enable them to make informed decisions about seeking evaluation and treatment for their children.

The Case Western Reserve University department of Orthodontics has started to include the PSQ along with the health history for every new patient. If the PSQ is scored as high risk, the parent is to be informed of this finding and given a written referral to a pediatric sleep physician. Based on 300 responses on the PSQ, 7.3% of our patient population is high risk for sleep-disordered breathing (Rorha et al 2016). Considering the volume of patients that are treated here and at most private orthodontic practices, this number is significant.

Our aim in this study was to assess the level of knowledge obtained by the parents of the children who had been identified as high risk for SDB based on their answers to the PSQ. The assessment of this relatively new care path will give us valuable information on whether we need to take different steps to ensure the parents of these patients are becoming fully

informed of the situation, and what it means for the health of their child. This information may also serve as a guide for the private practitioner who wishes to implement a sleep screening protocol in his/her own office.

Additionally, the normal health history form that has been in place has several questions that deal with signs and symptoms of SDB. This form has not been altered since the PSQ has been implemented. Now, parents are answering almost 30 questions dealing with sleep health on two separate forms. This raises the question of whether the health history form should be altered or if the sleep questions should simply be eliminated. We sought to understand the agreement between the health history questions and the questions on the PSQ, with the goal of understanding this relationship and eliminating redundancy in the screening process. Installing a simple and efficient screening protocol will help to ensure patients who may be suffering from this disorder are more likely to receive the care they need.

Qualitative Materials and Methods

The Case Western Reserve University (CWRU) Institutional Review Board (IRB) approved this study (IRB-2016-1779). For this portion of the study, subjects were recruited based on the outcome of the PSQ. Only patients who had PSQs scored, as high risk and a complete health history were included in the study. After review of all the patients who completed a PSQ, 22 were identified as potential participants in the study. Parents of the

22 subjects were recruited in person or over the phone. Of the 22, 16 agreed to take part in the interview. Consent was obtained for participation and audio recording. The parents were asked a list of scripted questions that were designed to assess understanding of pediatric sleep disordered breathing and how it affects their child. The interview script is shown in Table 1. After completion of the questions, the parents were given information about sleep-disordered breathing and why it is important. The content of this additional information was consistent among all of the interviews, and the parent was given the opportunity to ask questions after the additional information had been delivered.

The entire interview was audio-recorded and transcribed verbatim. Two independent investigators coded the interview transcriptions by attaching labels to short sections of the text; these one-two word labels categorized segments of text into possible themes in the narrative answers. The investigators demonstrated an initial 85% agreement on common coding and selection of text segments in the transcripts. After repeated discussion, a common list of themes was generated and the frequency of each was recorded. Selected quotes that represent the themes are presented in Table 4. In the interview, several closed-ended and scaled responses were collected; these responses were summarized and presented, per the quantitative methods below. The answers to these questions are shown in Tables 3 and 4 respectively.

Quantitative Materials and Methods

The same 22 patients who were identified as high risk from the PSQ were included in the quantitative portion of the study. Inclusion criteria for this portion were high-risk PSQ status and having a health history with the seven sleep questions included. The age range of the high-risk patients was representative of all PSQ respondents. A sample of 44 low-risk patients, matched for age, was included in the evaluation of responses to the HH questions and the PSQ. We identified seven questions on the existing health history that related to sleep health and are listed in Table 2. The answers to all of the PSQ questions and the answers to the seven sleep related questions from the existing health history were recorded for all 66 subjects. Two questions on the health history had multiple response categories that were dichotomized for analysis:

Snore at night (even without a cold): Never/Rarely/Sometimes/Often

Do you consider the volume of the snore to be:
Quiet/Moderate/Loud/Very Loud

For the first question, "never" and "rarely" were combined as a 'no' response, while "sometimes" and "often" were combined as a 'yes' response. For the second question, "quiet" and "moderate" were combined as a 'no' response, and "loud" and "very loud" were combined as a 'yes' response.

Statistical Analysis

All statistical techniques were performed in SPSS v24 (IBM, Armonk, NY). Frequencies and percentages of responses on the PSQ and HHQ were calculated for all 66 respondents. To compare the responses between the two questionnaires, percent exact agreement and Cohen's Kappa statistic (McHugh 2012) were calculated for each pair of comparable questions from the PSQ and the HHQ across the 66 subjects. To identify a potential subset of pre-screening questions before administering the PSQ, we used the PSQ High or Low risk categories as the 'gold standard' and evaluated combinations of PSQ and HHQ questions for sensitivity, specificity, as well as positive and negative predictive value.

Qualitative Results: Parental Interviews

Of the responses to the yes/no questions, eleven subjects were unaware that they had filled out the PSQ, and fourteen were not informed or do not remember being informed that their child was high risk for sleep disordered breathing. Similarly, only two subjects reported being given an explanation of the meaning of a positive result, and only two subjects reported being given the written referral to the sleep physician specialist. The frequency of the answers to the yes/no questions is listed in Table 3.

Common interview themes were identified and are listed in Table 4 with their frequencies. When asked to describe their understanding of what

being "high risk" meant, ten of the sixteen subjects responded with nighttime signs or symptoms of sleep disordered breathing. A sample of direct quotes regarding parental understanding is included in Table 5. None of the subjects mentioned snoring as a potential risk factor, and only one respondent mentioned potential daytime symptoms. When the researcher had described the potential signs and symptoms of sleep-disordered breathing, seven of the 16 parents recognized the existence of some of those symptoms in their child but were unaware that they may be related to sleep. This was particularly true of the daytime and behavioral symptoms such as hyperactivity. These seven interviews tended to last longer and the conversation focused on specific sleep behaviors of their child and the effect of poor sleeping on overall health.

To measure parental attitude regarding the discussion of sleep, three questions with a Likert-type scaled response were asked. Of the 16 subjects who were asked how important sleep health was to overall health on a scale of one to five, with five being very important, all 16 responded with a five. The data from the scaled-response questions are shown in Table 6. Following the interview, 13 of the patients requested a new copy of the referral to the sleep physician, and of two of the three who didn't request a referral were already under the care of a sleep physician.

Quantitative Results: Identification of the Pre-Screening Questions

To assess the agreement between comparable questions from the PSQ and HHQ, percent exact agreement and a Kappa statistic was calculated for seven combinations of five PSQ and four HHQ questions. Table 7 shows the combination of questions with the kappa and percent exact agreement. We found a broad range of agreement between similar questions. The snoring question from the health history compared to the snoring halftime questions from the PSQ had substantial agreement with a kappa value of 0.640. Conversely, the questions about the volume of the snore show poor agreement with a value of 0.226. (See Table 7) Questions about mouth or nose breathing were dissimilar enough that low agreement was not surprising. While the questions were similar in intent, the wording differences were sufficient to not be eliciting the same information, thus suggesting to look more closely at which combination of questions could more efficiently identify the high risk patients.

Table 8 shows the answers to each of the individual health history questions and its comparison to the PSQ outcome. These numbers allow us to calculate the sensitivity and specificity for each individual health history question to the outcome of the PSQ. Note that none of the individual health history questions had a very high sensitivity with the positive PSQ results. This implies that the individual health history questions did not capture the same information obtained using the PSQ.

To assess the value of the health history sleep questions as a whole unit, we calculated the number of "yes" answers on the health history questions and compared it to the outcome on the PSQ using responses from the 66 participants. This test is assessing sensitivity and specificity of the HHQ questions taken together to the known outcome of high or low risk as determined by the PSQ. The sensitivity is the rate at which the positive test result can correctly identify the known positive outcome; conversely, the specificity is the rate at which the negative test result correctly identifies the known negative outcome. We did not include the tonsil and adenoid questions on this assessment because 94% of all the patients answered no to these questions. These findings are shown in table 10. Answering at least one of the sleep questions positively on the HHQ gave a sensitivity of 77.3% and a specificity of 61.4%. With two positive answers, the sensitivity drops to 50% and the specificity goes to 86.3%. Even using different cut-offs, the low sensitivity for the outcome of the PSQ demonstrated that as a group the sleep questions on the HHQ were also not providing information that could predict the risk category on the PSQ.

Because the frequency of positive HHQ sleep questions was not providing information that was consistent with the PSQ outcome individually or taken together, we next examined individual PSQ questions. Table 9 shows the sensitivity and specificity of eleven of the individual PSQ questions as compared to the PSQ risk outcome. None of the questions alone gave a

sufficiently high sensitivity to be used on its own. However, a single question did have 73% sensitivity and 90% specificity by itself. Starting with this question and working toward 100% sensitivity for the outcome of the PSQ, three total questions were identified. Figure 1 shows the flow chart for the selection of these questions. These three questions combined provided 100% sensitivity and 75% specificity for the outcome of the PSQ.

Is your child easily distracted by external stimuli Y or N

Does your child snore at night (even without a cold) Never/Rarely or Sometimes/Often

Do you consider the volume of the snore to be loud Y or N

Our sample included 22 high-risk patients, all of whom answered at least one of these questions positively, providing 100% sensitivity (95% CI=81-100%). In our low risk sample of 44 patients of similar ages, 33 patients did not answer any of the questions positively. This resulted in 75% specificity (95% CI=59.3-86.3%).

Another metric we can derive from this group of questions is its positive predictive value for PSQ outcome. Positive predictive value is the proportion of positive test results that are likely to represent true positive disease in an unknown population. In this sample of 66 patients, with a known 33% prevalence of high risk (22/66), the positive predictive value was

quite high at 66.7%. But when applied to a large population, similar to the orthodontic clinic, with a presumed prevalence of 7.3%, the positive predictive value drops to 24% (shown in table 11). While this appears low, it is similar to the positive predictive value of the full PSQ when applied at this prevalence, but the three pre-screen questions would identify most if not all of the actual high-risk patients, while significantly minimizing the administration of the full PSQ. The pre-screen PPV at 24% is effectively the prevalence of the sample to which the PSQ would now be applied. At 24% prevalence, the PSQ now performs far better with a PPV of 67% due to eliminating a substantial number of false positives in this enriched sample (shown in table 12).

Discussion

Screening for sleep-disordered breathing is a new concept, particularly in an orthodontic practice, and adding a screening protocol to any office is a difficult undertaking. The qualitative portion of this study highlighted the importance of implementing a screening protocol, as only three of the high-risk patients reported discussing sleep health with another health practitioner. This finding was consistent with previous evidence that even symptomatic patients do not often discuss sleep health with their physician (Owens and Jones 2011). Our data show that most of the parents were not aware that a sleep questionnaire was part of their child's health assessment nor were they

aware that they had a child who was high-risk for SDB. Most of the parents do not remember receiving any explanation about high-risk status or any information about SDB. Given this information, it is not surprising that only one of the parents understood that daytime symptoms could be associated with sleep disordered breathing in children and none of the parents reported snoring as a potential health issue in children. These findings are consistent with previous studies assessing parental knowledge of SDB (McDowall et al 2015)(Schreck et al 2011). These parents have minimal understanding of something that can have serious health consequences for their child and they may not be getting this information anywhere else.

Another reason to screen for SDB and provide information to parents about health consequences is to reinforce the importance of sleep hygiene to overall health. This is supported by the finding that seven of the 16 parents recognized signs and symptoms of sleep disordered breathing in their children but had never made the connection that it could be due to a sleep issue.

Our results show that SDB screening is most valuable if it is followed by a conversation between the provider and the parent about the meaning of the high-risk score. Without this follow-up, parents don't know what the PSQ is or what the outcome means. The provider must have a conversation with the parents, and our data shows that making a personal connection is important when having this conversation.

Individual questions on the health history had poor agreement with similar questions asked on the PSQ. This shows that we are not getting similar information from individual questions on the health history. The poor sensitivity of all of the health history questions using different cut off points shows that we are not getting enough information from the health history alone to replace the PSQ. Our results are in agreement with previous studies where questionnaires using only nighttime signs and symptoms were not useful for diagnosis of SDB (Goodwin et al 2005).

While no single question on the PSQ or HHQ was predictive of high risk, we found that the behavioral questions had higher sensitivity and specificity than did other groups of questions. This finding is consistent with previous studies by Marcus et al (2012). However, we sought to identify a small group of questions with high sensitivity for the outcome of the PSQ, for the purposes of a pre-screen to decrease the number of patients who had to complete the full PSQ. We were not looking for a diagnostic tool, but rather sought to make a more efficient screening tool. We obtained 100% sensitivity to the PSQ in this small group of patients, suggesting we would not miss any patients who would be potentially high-risk.

We propose to alter the health history form so that only the three selected questions are included in the section about sleep health. If the patient answers at least one of those questions positively, they are then given the PSQ to fill out. This simplifies the screening process. Fewer parents will

be asked to complete the full PSQ and those that are asked will know why they are being asked to complete a more comprehensive questionnaire.

When every patient is given the PSQ, the provider has to score all of the PSQs even though 93% of them were for patients with low risk. That is a lot of questions for the patient, and a lot of negative screening results to sift through for the provider. Using the PSQ in low risk populations results in a higher number of false positives being referred to the sleep physician. Table 13 shows what might happen in a hypothetical population of 1000 orthodontic patients. High-risk PSQ scores will be found in about 7.3% of an orthodontic population (Rorha et al 2016). However, we estimate that the positive predictive value of the PSQ is only 34%. This means that of the 183 patients referred to sleep medicine doctors only 62 would actually have SDB based on overnight polysomnography. Tables 11 and 12 show the same 1000 patient sample that has been enriched by the pre-screen;. With pre-screening only 305 PSQ forms are handed out, the PPV becomes 67% and 92 referrals are made to sleep medicine. It is important to note that this pre-screening protocol results in the same number of individuals (i.e. 62) being subsequently identified as having SDB using overnight PSG. The pre-screen reduces the burden on the low-risk patients and the provider and increases the efficiency of the screening process.

When using the proposed pre-screen, it is simple to determine if the PSQ should be given, and the act of giving the PSQ with an explanation

opens the dialogue between the provider and the patient. The patient will want to know what the PSQ is, and why they are being asked to fill it out. This means that at least some information will have to be given even before the PSQ is filled out. Then the patient will want to know the outcome of the PSQ and what it means, resulting in a more meaningful conversation and a higher likelihood of follow-up with the referral.

Limitations

This study has several limitations, the first and foremost being the small sample size. We were able to show that these three questions would have successfully screened for the 22 high risk patients in this group, but there is no way to tell whether every single high-risk patient from a larger group would be identified. Additionally, in several of the subjects, the health history and the PSQ were filled out at different times. It is difficult to determine how this would have affected the answers given on either form.

Conclusions

Screening for sleep-disordered breathing is important in an orthodontic population. Unfortunately, implementing a screening protocol can be difficult to manage and ensure that meaningful information is disseminated to the high-risk patients. Using our three-question pre-screen combined with the PSQ, high-risk patients can be identified, educated and referred efficiently

and effectively. We believe this protocol will be useful for screening sub adult orthodontic patients in a residency or private practice situation.

REFRENCES

Chervin, R. D., Weatherly, R. A., Garetz, S. L., Ruzicka, D. L., Giordani, B. J., Hodges, E. K., … Guire, K. E. (2007). Pediatric Sleep Questionnaire Prediction of Sleep Apnea and Outcomes. *Arch Otolaryngol Head Neck Surg*, *133*, 216–222

Goodwin, J. L., Kaemingk, K. L., Mulvaney, S. A., Morgan, W. J., & Quan, S. F. (2005). Clinical screening of school children for polysomnography to detect sleep-disordered breathing--the Tucson Children's Assessment of Sleep Apnea study (TuCASA). *Journal of Clinical Sleep Medicine : JCSM : Official Publication of the American Academy of Sleep Medicine*, *1*(3), 247–54. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/16429591

Lumeng, J. C., & Chervin, R. D. (2008). Epidemiology of pediatric obstructive sleep apnea. *Proceedings of the American Thoracic Society*, *5*(2), 242–52. http://doi.org/10.1513/pats.200708-135MG

Marcus, C. L., Brooks, L. J., Draper, K. A., Gozal, D., Halbower, A. C., Jones, J., … Kelly, J. (2012). Diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*, *130*(3), e714-55. http://doi.org/10.1542/peds.2012-1672

Mcdowall, P. S., Galland, B. C., Campbell, A. J., & Elder, D. E. (n.d.). Parent knowledge of children's sleep: A systematic review. http://doi.org/10.1016/j.smrv.2016.01.002

McHugh, M. L. (2012). Interrater reliability: the kappa statistic. *Biochemia Medica*, *22*(3), 276–82. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/23092060

Rorha, A., Hans, M. . (2016). *Prevalence of Sleep disordered Breathing in an Orthodontic treatment seeking Population* Unpublished master's thesis, Case Western Reserve University, Cleveland, Ohio.

Young, S., Yoon, R., Jain, U., & Shapiro, C. (2012). Sleep in attention-deficit/hyperactivity disorder in children and adults: Past, present, and future. *Sleep Medicine Reviews*, *16*, 371–388. http://doi.org/10.1016/j.smrv.2011.07.001

APPENDIX 1: TABLES

Table 1: Interview Questions

1. Were you aware that a sleep health questionnaire was included in the health history you completed at your initial appointment with the orthodontic department?

2. If yes, were you aware that your child was deemed as high risk for something called "sleep disordered breathing" based on the answers you gave on the questionnaire?

3. Has your child's physician or any other healthcare provider ever advised you about pediatric sleep disordered breathing or sleep apnea?

4. Please describe your understanding of what it means for your child to be classified as high risk for sleep disordered breathing?

5. Did an orthodontic resident or faculty member give you an explanation on the meaning of a positive result? Yes/No/Don't Remember

6. If yes, on a scale of 1-5 how satisfied were you with the explanation with 1 being the least satisfied and 5 being the most?

7. Were you given a written referral to a sleep doctor specialist? Yes/No/Don't remember

8. If yes, did you contact the sleep doctor for an appointment?

9. If you decided not to follow up with the sleep doctor why did you choose not to?

10. If this sleep health information and referral had come from a different source, would you be more likely to follow through with the referral?

11. On a scale of 1-5 how important is it for your orthodontist to consider the overall health of your child with 1 being the least important and 5 being the most important?

12. On a scale of 1-5 how comfortable are you with the orthodontist screening your child for other health issues with 1 being the least comfortable and 5 being the most comfortable?

13. On a scale of 1-5 how important do you think sleep quality is to your child's overall health with 1 being the least important and 5 being the most important?

14. Is there anything else you would like to add?

At this point the interviewer will provide the patient with additional information about pediatric sleep disordered breathing. Another written referral to sleep medicine will also be given if required.

15. Do you have any further questions for me or would you like anything clarified?

Table 2: Sleep health questions on the current health history

Does the patient:

Difficulty in breathing through the nose Y/N
Have asthma or allergies Y/N
Snore at night (even without a cold) Never/Rarely/Sometimes/Often
Do you consider the volume of the snore to be Quiet/Moderate/Loud/V.Loud
Had tonsils removed Y/N
Had adenoids removed Y/N
Breathe mainly through the mouth Y/N

Table 3: Yes/No interview questions

Interview Question	Yes	No	Don't Know/Don't Remember
Were you aware that a sleep health questionnaire was included in the health history you completed at your initial appointment with the orthodontic department?	5	11	0
Were you aware that your child was deemed as high risk for something called "sleep disordered breathing" based on the answers you gave on the questionnaire?	2	12	2
Has your child's physician or any other healthcare provider ever advised you about pediatric sleep disordered breathing or sleep apnea?	3	12	1
Did an orthodontic resident or faculty member give you an explanation on the meaning of a positive result?	2	12	2
Were you given a written referral to a sleep doctor specialist?	2	14	0

Table 4: Themes from transcribed parent interviews

Theme	Frequency	Representative Quote
Personal Concern	12	"...are you saying my child in particular?"
Understands nighttime signs of sleep disordered breathing	10	"...they stop breathing periodically through their sleep..."
Recognized potential for daytime symptoms to be caused by sleep issue	1	"...they are not as well rested as they should be and throughout the course of the day may not be functioning at optimal levels."
Connected symptoms of SDB with symptoms in their own child (frequency after delivery of information)	7	"...it wouldn't surprise me if he had something like that...I think he has sleep issues...he struggled with grades..."

Table 5: Responses to the following interview question: Please describe your understanding of what it means for a child to be high risk for sleep disordered breathing.

"...that you could go into distress maybe while you are sleeping"

"...they stop breathing periodically through their sleep or they struggle to catch thier breath so to speak..."

"...she's more likely than others to stop breathing in the middle of the night for whatever reason..."

"...the physiology around the throat will close temporarily while the person is sleeping and then stops them or prevents them from breathing..."

"I understand that it would be that they find it difficult to breathe at night efficiently..."

"Probably reporting that they are not sleeping well, having problems with their breathing, nose or mouth obstructions..."

"I mean I can guess but I'll say no"

"I really don't know"

"I usually think of people who might be overweight, older..."

"That she probably has it and that she needs the, what is it? CPAP machine..."

"They probably have a preexisting; you know maybe a lung condition, asthma, something like that that could precipitate or go into a sleep problem"

Table 6: Scaled interview questions

Scaled Interview Question	Average
On a scale of 1-5 how important is it for your orthodontist to consider the overall health of your child with 1 being the least important and 5 being the most important?	4.65
On a scale of 1-5 how comfortable are you with the orthodontist screening your child for other health issues with 1 being the least comfortable and 5 being the most comfortable?	4.18
On a scale of 1-5 how important do you think sleep quality is to your child's overall health with 1 being the least important and 5 being the most important?	5.00

Table 7: Agreement between PSQ and HHQ questions

PSQ question	HHQ Question	Kappa	Percent exact agreement
While sleeping does your child snore more than half the time?	Does the patient snore at night (even without a cold) N/R/S/O?	0.640	86%
While sleeping does your child always snore?	Does the patient snore at night (even without a cold) N/R/S/O?	0.426	83%
While sleeping does your child snore loudly?	Do you consider the volume of the snore to be Q/M/L/VL?	0.226	74%
While sleeping does your child have trouble breathing or struggle to breathe?	Does the patient have difficulty in breathing through the nose?	0.279	86.7%
While sleeping does your child have trouble breathing or struggle to breathe?	Does the patient breathe mainly through the mouth?	0.345	89%
Does your child tend to breathe through the mouth during the day?	Does the patient have difficulty in breathing through the nose?	0.421	86.2%
Does your child tend to breathe through the mouth during the day?	Does the patient breathe mainly through the mouth?	0.509	89%

Table 8: Percent of High and Low-Risk PSQ patients responding Yes to individual HHQ questions

Question	High-Risk	Low-Risk
Does the patient have difficulty in breathing through the nose?	33.3%	7.1%
Does the patient have asthma or allergies?	45.5%	21.4%
Does the patient snore at night (even without a cold) N/R/S/O?	52.4%	14.6%
Do you consider the volume of the snore to be Q/M/L/VL?	11.8%	4.0%
Has the patient had tonsils removed?	4.8%	4.8%
Has the patient had adenoids removed?	4.8%	7.3%
Does the patient breathe mainly through the mouth?	33.3%	2.4%

Table 9: Percent of High and Low-Risk PSQ patients responding Yes to individual PSQ questions.

Question	High-Risk	Low-Risk
While sleeping does your child snore more than half the time?	57.9%	7.1%
While sleeping does your child always snore?	29.4%	0.0%
While sleeping does your child snore loudly?	30.0%	2.4%
While sleeping does your child have trouble breathing or struggle to breathe?	15.8%	2.3%
Does your child tend to breathe through the mouth during the day?	40.0%	0.0%
My child often does not seem to listen when spoken to directly	47.6%	6.8%
My child often has difficulty organizing tasks and activities	50.0%	2.3%
My child often is easily distracted by extraneous stimuli	72.7%	9.5%
My child often fidgets with hands or feet or squirms in seat	59.1%	4.5%
My child often is 'on the go' or acts as if 'driven by a motor'	47.4%	2.3%
My child often interrupts or intrudes (e.g. butts into conversations or games)	47.6%	2.3%

Table 10: HHQ data taken as a whole using two cutoffs

HHQ questions	High-Risk	Low-Risk
0 Yes	22.7%	61.4%
≥1 Yes	77.3%	38.6%
≥2 Yes	50.0%	13.7%

Table 11: Hypothetical 1000 patient sample with 7.3% prevalence of high risk PSQ status

	PSQ HR	PSQ LR
Pre-screen HR	73	232
Pre-Screen LR	0	695

Table #12: Enriched sample of 305 patients, presumed 24% prevalence of SDB

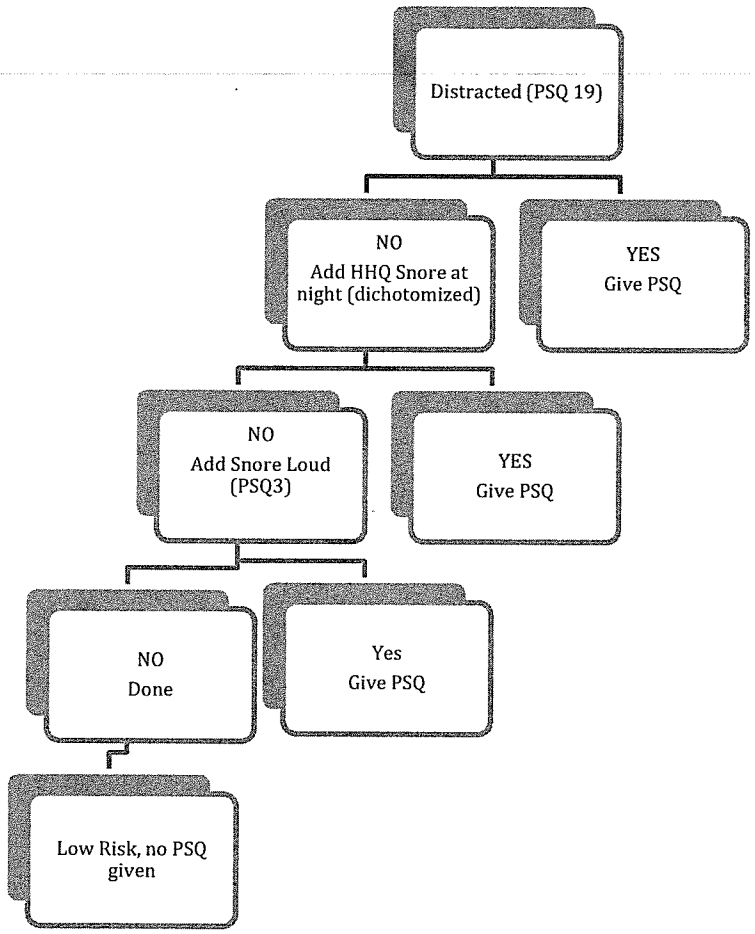
	SDB	Non-SDB
PSQ HR	62	30
PSQ LR	11	202

Table #13: Hypothetical population using only the PSQ with assumed prevalence of 7.3% SDB

	SDB	Non-SDB
PSQ HR	62	121
PSQ LR	11	806

APPENDIX B: FIGURES

Figure #1: Selection of questions for the pre-screen



APPENDIX C: IRB APPORVAL/AMENDMENTS

CASE WESTERN RESERVE UNIVERSITY EST. 1826

NOTICE OF APPROVAL

CWRU IRB Protocol Number:	**IRB-2016-1779**
Protocol Title:	***Educational Outcomes of Parents of Patients Identified as High Risk for Sleep Disordered Breathing in an Orthodontic Office***
Responsible Investigator (RI):	**Mark Haas**
Co-Investigator (CI):	**Brian Potocki, Catherine Denko, Juan Martin Palomo**
RI Department:	**Case Western Reserve University IBC and SBER IRB - DEN - Orthodontics**
Type of Review:	**Expedited**
Risk Level:	**Minimal**
Vulnerable Population(s):	**Children**
Approval Date:	**01/30/2017**
CONTINUING REVIEW DEADLINE:	**01/15/2018**
EXPIRATION DATE:	**01/29/2018**

The CWRU Institutional Review Board (IRB) has approved the above new protocol through the **EXPEDITED** review process.

When conducting Human Subjects' Research, your responsibilities include the following:

1. Report all adverse events and unanticipated problems involving human subjects to the IRB Office, located in the Office of Research Administration (ORA), within three (3) business days of your knowledge of the occurrence.
2. Provide the IRB with a complete Continuing Review form (available in our electronic application system at http://sberirb-app.case.edu or from the ORA) by the continuing review deadline noted above, and when the study is to be terminated.
3. Submit all proposed changes to the protocol to the IRB and wait for IRB approval before implementing any protocol change or modification.
4. Keep all research data and original consent documents in your possession for at least three (3) years after the study is terminated.
5. Note that completely de-identified data can be kept and used for research indefinitely. The IRB is primarily concerned about identifiers/identifiable data. If you want to terminate your study, identifiers/identifiable data must be destroyed. This includes paper or electronic master lists, contact lists, codes/codebooks, transcripts containing identifiers and video and audio recordings.
6. If applicable, please use the most current IRB-approved consent forms. Feel free to use copies of these forms as long as they are identical to what was originally IRB approved. If you wish to change the forms or any other part of the study, you must submit an addendum request/protocol modification with revised copy(ies) of the relevant document(s) and wait for IRB approval before a modification can be implemented.
7. ***Discontinue all work pertaining to this protocol if a continuing review approval is NOT finalized by the expiration date noted above.*** No further work on this protocol is allowable until the proper continuing review materials or required revisions are approved by the IRB.

    a. Please note that, if the continuing review materials or required revisions are not received by the expiration date, the RI and this study would be automatically placed on Administrative Hold for 30 days. This means that the RI loses their IRB privileges and research from this study must cease.
    b. If after 30 days, the CWRU IRB still does not have the protocol for processing, this protocol will be administratively terminated and your IRB privileges will be revoked for all your protocols.

Thank you for your attention to this matter. Please contact the IRB office at 216-368-6993 if we can be of further assistance.

CASE WESTERN RESERVE UNIVERSITY EST. 1826

NOTICE OF ADDENDUM APPROVAL

CWRU IRB Protocol Number:	**IRB-2016-1779**
Protocol Title:	***Educational Outcomes of Parents of Patients Identified as High Risk for Sleep Disordered Breathing in an Orthodontic Office***
Responsible Investigator (RI):	**Mark Hans**
Co-Investigator (CI):	**Brian Potocki, Catherine Demko, Juan Martin Palomo**
RI Department:	**Case Western Reserve University IBC and SBER IRB - DEN - Orthodontics**
Protocol Expiration Date:	**01/29/2018**
Approved Change	**Modified the timeframe for the destruction of the master list of identifiers; addition of Catherine Demko to data access; altered questions to survey and interview script**
Addendum Approval Date:	**03/08/2017**

The CWRU Institutional Review Board (IRB) has **APPROVED** the submitted addendum for the above protocol.
As an investigator of human subjects, your responsibilities include the following:

1. Report all adverse events involving human subjects to the IRB within three (3) business days of the occurrence.
2. Submit any further changes to an approved protocol or consent form to the IRB, and receive approval from the IRB, before implementation of the change.
3. Keep all research data and original consent documents in your possession for at least three (3) years after the study is terminated.

Please use the most current IRIS IRB Application version of all study documents; i.e., informed consent & recruitment documents.

CASE WESTERN RESERVE UNIVERSITY EST. 1826

NOTICE OF ADDENDUM APPROVAL

CWRU IRB Protocol Number:	**IRB-2016-1779**
Protocol Title:	***Educational Outcomes of Parents of Patients Identified as High Risk for Sleep Disordered Breathing in an Orthodontic Office***
Responsible Investigator (RI):	**Mark Haas**
Co-Investigator (CI):	**Brian Potocki, Catherine Demko, Juan Martin Palomo**
RI Department:	**Case Western Reserve University IRC and SBER IRB - DEN - Orthodontics**
Protocol Expiration Date:	**01/29/2018**
Approved Change	**Interview to be completed over the phone**
Addendum Approval Date:	**03/28/2017**

The CWRU Institutional Review Board (IRB) has **APPROVED** the submitted addendum for the above protocol.
As an investigator of human subjects, your responsibilities include the following:

1. Report all adverse events involving human subjects to the IRB within three (3) business days of the occurrence.
2. Submit any further changes to an approved protocol or consent form to the IRB, and receive approval from the IRB, before implementation of the change.
3. Keep all research data and original consent documents in your possession for at least three (3) years after the study is terminated.

Please use the most current IRBIS IRB Application version of all study documents; i.e., informed consent & recruitment documents.

CASE WESTERN RESERVE UNIVERSITY EST. 1826

NOTICE OF ADDENDUM APPROVAL

CWRU IRB Protocol Number:	**IRB-2016-1779**
Protocol Title:	***Educational Outcomes of Parents of Patients Identified as High Risk for Sleep Disordered Breathing in an Orthodontic Office***
Responsible Investigator (RI):	**Mark Haas**
Co-Investigator (CI):	**Brian Potocki, Catherine Demko, Juan Martin Palomo**
RI Department:	**Case Western Reserve University IBC and SBER IRB - DEN - Orthodontics**
Protocol Expiration Date:	**01/29/2018**
Approved Change	**access to the health history of the existing patients at the orthodontic clinic; comparing the answers to the 22 question sleep questionnaire with the answers the sleep questions on the existing health history**
Addendum Approval Date:	**05/11/2017**

The CWRU Institutional Review Board (IRB) has **APPROVED** the submitted addendum for the above protocol.
As an investigator of human subjects, your responsibilities include the following:

1. Report all adverse events involving human subjects to the IRB within three (3) business days of the occurrence.
2. Submit any further changes to an approved protocol or consent form to the IRB, and receive approval from the IRB, before implementation of the change.
3. Keep all research data and original consent documents in your possession for at least three (3) years after the study is terminated.

Please use the most current IRIS IRB Application version of all study documents; i.e., informed consent & recruitment documents.