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Validation of a Screening Tool for Pediatric Obstructive Sleep Apnea: A Pilot Study

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Validation of a Screening Tool for Pediatric Obstructive Sleep Apnea: A Pilot Study

A thesis submitted in partial fulfillment of the requirements for the degree of Master of Science in Dentistry at Virginia Commonwealth University.

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Abstract

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DEPARTMENT OF ORTHODONTICS

Purpose: The purpose of this study was to evaluate the Pediatric-Modified-STOP-Bang (PM-STOP-Bang) as a screening tool to assess sleep related breathing disorders (SRBD) within a pediatric population. The specific aims of this study were: 1) compare the PM-STOP-Bang scores with those from the validated Pediatric Sleep Questionnaire (PSQ), 2) determine the ability of the PM-STOP-Bang and the PSQ screening tools to accurately detect children of high risk for SRBD from a one-night home sleep apnea test (HSAT).

Methods: Orthodontic patients were screened using the PM-STOP-Bang at VCU Graduate Clinic. Patients (n=10) who were recruited and enrolled. Five were determined to be at high risk based on PM-STOP Bang score. After enrollment, a guardian completed the PSQ, the Pediatric Symptom checklist (PSC), and the child completed a one-night HSAT. Five age-matched controls were enrolled.

Results: Scores of PM-STOP-Bang compared to PSQ related to a sensitivity for the PM-STOP-Bang of 67% (95% CI: 29-100%) and a specificity of 75% (95% CI: 33-100%). Comparing the PM-STOP-Bang to the HSAT results, the PM-STOP-Bang had a sensitivity of 100% (95% CI: 100%, 100%) and a specificity of 71% (95% CI: 38%, 100%). The PSQ sensitivity was 67% (95% CI: 13%, 100%) and specificity 43% (95% CI: 6%, 80%).

Conclusions: The PM-STOP-Bang achieved a greater sensitivity and specificity than the PSQ in identifying children at high risk for obstructive sleep apnea when cases were confirmed with the HSAT. A future study with a larger sample size is needed to validate the PM-STOP-Bang.

Introduction

Sleep-related breathing disorders (SRBD) are a diagnostic category that reflect an array of obstructive phenomena and conditions ranging from primary snoring, upper airway resistance syndrome, and obstructive sleep apnea (OSA).[1] OSA is characterized by prolonged, partial or complete upper airway obstruction that disrupts normal ventilation and normal sleep in patients.[2] The constricted airway and resultant reduced airflow cause an increased respiratory effort. Furthermore, a relative decrease in serum oxygen may initial cortical arousal from sleep: The activation of sympathetic nervous system can then lead to increased heart rate, blood pressure and arrhythmia.[3]

OSA affects both adults and children, but there are differences in the symptoms, prevalence, etiology and pathophysiology depending on the age of the patient. According to the American Sleep Apnea Association, 1-4% of children suffer from sleep apnea.[4] The etiology of pediatric obstructive sleep apnea (POSA) is complex and multifactorial; there is evidence that pathogenesis is associated with neuromuscular tone dysfunction and upper airway constrictions, most often due to adenotonsillary hypertrophy.[5]

Symptoms of POSA range from chronic snoring, increased respiratory effort at breathing, daytime fatigue and sleepiness, nocturnal enuresis, irritability, and behavioral and neurocognitive changes.[6] In severe cases on untreated or poorly controlled POSA, clinical sequelae include failure to thrive, cor pulmonale, pulmonary hypertension, and other cardiovascular complications.[5] Furthermore, about 25% of children diagnosed with Attention Deficient

Hyperactivity Disorder (ADHD) have symptoms of POSA. In these children, learning difficulties and behavior problems attributed to ADHD are actually the result of sleep problems.

Unfortunately, eighty percent of adult men and ninety percent of women with OSA remain undiagnosed despite available screening tools. Appropriate diagnosis of POSA or SRBD is even more elusive in children because of less distinct signs and symptoms.[7] Therefore, the diagnosis of POSA and SRBD tends to be frequently delayed or missed.

Diagnostic confirmation of SRBD is carried out by a sleep medicine physician through in-center polysomnography (PSG), or out-of-center tests (OCST).[6] The PSG sleep study involves recording of a variety of body functions including electrical activity of the brain, eye movements, muscle activity, heart rate, breathing patterns, air flow and blood oxygen levels. PSG is the "gold standard" for the diagnosis of OSA. During sleep, the number of apneas and hypopneas is recorded and apnea-hypopnea index (AHI) is calculated by determining the number of events per hour. AHI is then used to determine the severity of sleep apnea. In adults, while an index of less than 5 is considered normal, values higher than 30 indicate severe sleep apnea.[2]

An in-center PSG sleep study is labor-intensive: it causes discomfort and inconvenience to patients and their families. In addition, it has around a 6-month wait time between referral, evaluation, and diagnosis.[8] There are other limitations with the use of PSG including the high cost and lack of access to the study centers, which usually only occur at integrative medical centers.[9]

An home sleep apnea test (HSAT) is a type of OCST which includes 4-7 channels of monitoring but does not include electroencephalogram.[2] The home-based PSG studies have advantages in that they are simplified and performed in the subject's natural sleep environment.

In addition, there is no wait time, and the equipment is more readily available.[10] However, because a home test only evaluates breathing, results may be inconclusive or false negative. In the literature, sensitivity and specificity of the home-based studies are reported to range from 86-100% and 64-100%, respectively.[11]

As mentioned earlier, a PSG at a sleep center is the standard method for the diagnosis of OSA. However, today HSATs are popular because of their simplicity, cost-effectiveness, and reliability.[12] The NOX-T3 portable device is a type of HSAT monitor that received the Food and Drug Administration's acceptance for an off-site sleep testing service in 2008. The monitor is approved for use on patients two years of age and older.[13]

Previous studies have shown that the HSAT using the NOX-T3 had a good agreement compared to PSG with a high degree of sensitivity for detecting OSA.[12] Moderate to severe OSA was diagnosed with 93% sensitivity and 85% specificity with the use of the NOX-T3. The simultaneous in-laboratory T3 study showed an even higher agreement with PSG (100% sensitivity and 94% specificity) highlighting that the differences found between the home NOX-T3 study and in-lab PSG are mainly due to the night-to-night variability and differences in sleep environment.[12] While all of this information regarding HSATs is hopeful and reduces barriers relative to in-center PSG, patients must still be referred to a sleep physician once they are identified as high-risk for OSA.

As discussed previously, POSA can be serious and life-threatening in children. According to Jennum et al.[14] early diagnosis of POSA in pediatric population is of paramount importance. However, OSA remains heavily undiagnosed despite well-documented symptoms and complications.[15] Nevertheless, prescribing every child over the age of five with an in-center

PSG is not a realistic or valid approach for the diagnosis OSA and SRBD. Using an HSAT would be a more convenient option, but only board-certified physicians may administer and interpret the data. Another alternative and cost-effective option is for health care providers to administer screening questionnaires to identify children of high risk. Non-PSG screening tools obtaining information on the pathophysiology, symptoms, and typical craniofacial presentations offer a good alternative to the traditional invasive in-lab test. However, the major limitation in using non-PSG screening tools is that they are insufficient when used alone to definitively diagnosis OSA. Nevertheless, they can guide the clinician when it is appropriate to make a referral to a sleep specialist who can diagnose sleep apnea.

There are many non-PSG screening tools that have been developed in an effort to screen for SRBD in children, however the Pediatric Sleep Questionnaire (PSQ) is the only instrument that has been validated to use pediatric patients.[8] The PSQ is a 22-item survey administered to explore prominent symptoms associated with SRBD. There are questions regarding behavior, breathing, sleepiness, and other important signs of the disorder. When compared with PSG data, PSQ instrument has been shown to have a sensitivity of 85% and a specificity of 87% for identifying SRBD in children 2-18.[16] While the PSQ has the best diagnostic accuracy, its shortcomings does not allow for replacing the current gold standard, PSG.

Another non-PSG screening tool that can be used as an adjunct in assessment of SRDBs is the Pediatric Symptom Checklist (PSC). The PSC is a psychosocial one-page screening tool aimed to assess signs and behavior to allow for appropriate interventions.[17] This well-established and validated questionnaire can be used as a supplement to specifically address the behavioral components of the disorder, but is not intended to use solely for the diagnosis SRDBs.

There has been some research to show that questionnaires, physical examinations or both could be useful for reliably diagnosing SRDB. In 2008, Chung et al.[18] developed and validated a STOP-Bang questionnaire as a screening tool for OSA in adult patients 18 years and older. This questionnaire consists of four "yes" or "no" questions: "Do you **S**nore loudly?", "Do you feel **T**ired during the daytime?", "Has anyone **O**bserved you stop breathing during sleep?", and "Do you have high blood **P**ressure?" Furthermore, body mass index (**B**), age (**A**), neck size (**N**), and gender (**G**) were found to have a sensitivity of 83.6%, 92.9% and 100% for mild, moderate and severe OSA, respectively.[18]

Due to a need for a non-PSG screening tool that takes into account both physical and behavioral presentations in high-risk children for POSA and SRBD, Chiang et al.[19] modified the STOP-Bang instrument to accommodate pediatric populations. The Pediatric Modified-STOP-Bang (PM-STOP-Bang) was revised after the study to include the presence of snoring (S), tonsillar hypertrophy (T), observed obstruction (O), the BMI percentage for age and gender, the age at diagnosis (A), the presence of neuromuscular disorder (N), and the presence of genetic/ congenital disorder (G). The screening tool proved to be predictive of pediatric OSA, but needed more exploration of ethnicity factors, presence of asthma, and family history of OSA.[19] Although, it has not been validated to use for POSA and SRBD screening, it provides merit in its ability to identify the physical and behavioral manifestations of the disorder.

According to a recent study, the prevalence of SRBD in orthodontic patients is 10.8%, which is much higher than the prevalence seen in a normal healthy pediatric population (1-4%). (Abtahi, 2019) One of the major reasons for high prevalence in orthodontic patients is because of the known craniofacial manifestations of POSA and SRBD that result in malocclusion.

Currently, the PSQ is the only recommended screening tool to be used by dentists and orthodontists to identify pediatric SRBD.[20] However, the PSQ does not take into account physical examination, such as tonsillar hypertrophy.[8]

The specialty of orthodontics provides much more value to patients than simply aligning teeth and this is evident with the current role of orthodontists in the management of sleep apnea. In 2019, The American Association of Orthodontists released a white paper discussing the prevalence of OSA and a significant role the orthodontist can play in early diagnosis.[2] Furthermore, orthodontists can easily administer screening for OSA in their broad patient population (child, adolescents, adults) and can therefore be a valuable player in the multidisciplinary management of OSA.

Also, orthodontists are experts in the anatomy and development of craniofacial complex and are skilled to screen for craniofacial morphologies associated with POSA, such as steep mandibular plane, retrusive chin, inferior hyoid bone, narrow maxilla, long facial height, tendency for anterior open bite, and decreased pharyngeal airway.[5] These craniofacial features are well-established risk factors for SRDB and may be easily assessed from routine orthodontic records. Moreover, orthodontists have a long history of working with other specialists in medicine and dentistry to provide collaborative efforts for their patients with specific needs (such as cleft lip, and pediatric patients).[2] Finally, the frequency of routine orthodontic visits and regular updates to health history forms also strengthen the ability of orthodontists to fill this role as early providers. Therefore, the availability of a diagnostic tools, such as a screening questionnaire, may help orthodontists to identify patients at risk and make early referrals to the appropriate physician.

Since there is not a concise questionnaire that explores both physical and behavioral manifestations for pediatric patients, the PM-STOP-Bang screening tool addressing these components of SRBD may therefore be successfully used for the detection of POSA in children.

The purpose of the study was to evaluate the PM-STOP-Bang instrument as a valid screening tool to assess SRBD in children. Specifically, the aims of this study were: 1) to compare the PM-STOP-Bang scores with the results of the Pediatric Sleep Questionnaire (PSQ), 2) to determine the ability of the PM-STOP-Bang and the PSQ to detect patients at high risk for POSA by confirming the disorder with a one-night home sleep apnea test (HSAT).

Methods

Prior to starting the study, ethical approval was obtained from the institutional review board (IRB) of Virginia Commonwealth University (IRB ID: HM20017418). The study sample consisted of children seeking treatment for malocclusion at VCU Orthodontic Clinic. A PM-STOP-Bang survey, as a component of the initial records appointment, was verbally administered to the patient's parents or guardian by the treating doctor (Figure 1). All doctors were previously calibrated on how to appropriately use the 8-item screening questionnaire where each "yes" answer was scored 1 point. In this study, a score of 3 and higher designated a high risk for sleep apnea. This scale was chosen based on the findings of a previous study by Chiang et al.[19]

Children 7-17 years of age without a history of previous orthodontic treatment and an obstructive sleep apnea diagnosis were included in the study. Individuals with craniofacial syndromes and skeletal asymmetries were excluded. At the initial exam appointment, patients who were identified at high risk for POSA based on their PM-STOP-Bang scores were assigned to the experimental group. Once a case was enrolled, an age-matched, low-risk patient with a PM-STOP-Bang score of 2 and lower was assigned to the control group.

A total of 10 participants (n=5, experimental and n=5, control) were recruited into this pilot study. Each patient was assigned a case number containing using a code such as Case 001 and Control 001. No individual identifying information was kept for the analysis. However, for the direct benefit of the child, a key corresponding to each subject was retained on a secure VCU Google drive so that any OSA diagnosis would allow for follow up and referral process.

Regardless of study enrollment decisions, patients identified by the PM-STOP-Bang questionnaire at high risk for OSA were provided with referral to a primary care provider, following the standard of care procedures for VCU Orthodontic Clinic.

During the recruitment process, it was explained that enrollment into the experimental group would require two additional questionnaires (PSQ and PSC, Figure 2 and Figure 3) to be completed as well as a one-night HSAT. A written and verbal consent/ assent was obtained from all study participants. Parents and guardians were provided information about the at-home sleep apnea test. An in-depth description of how to set up and use the portable sleep apnea device (NOX-T3, version 1.0, Nox Medical USA, Suwanee, Georgia) was provided for the parents and the patient. They were also asked to watch a short video, specifically created by the manufacturer for the patient use of the sleep apnea device, demonstrating the hookup and takedown procedures. This video was available to the public on the internet, and the participants were provided with the link so they could watch it again at a later time if needed. A written transcript of the video, as well as the original Nox-T3 instructions manual was provided as well.

Each HSAT kit included NOX-T3 home sleep testing device with abdomen cable attached, the wrist oximeter and Velcro strap with the oximeter probe attached, two belts and a nasal canula in a sealed plastic bag, bandages and tape to allow for better fastening of the device, and the instructions manual. NOX-T3 portable sleep monitoring device records oro-nasal airflow and oro-nasal sound, a pulse oximetry, heart rate and respiratory effort recorded with a chest belt and a recording unit. These measures were used for diagnosis by a sleep physician who was blinded to the study participants. The total apnea-hypopnea index (AHI), a measure of the

severity of OSA, was then calculated and the case was classified as normal, mild, moderate, or severe.

Pediatric-Modified-STOP-Bang Questionnaire

Child's Name: _____ **Date: __/__/__**

Please circle the correct response.

S Snoring: Does the child snore during naps or at night?	**YES**	**NO**
T Tonsillar hypertrophy: Does the child have large tonsils?	**YES**	**NO**
O Sleep obstruction: Does the child experience sleep obstruction (choking or gasping)?	**YES**	**NO**
P Psychological/ Behavioral symptoms: Does your child have Attention Deficient Hyperactivity Disorder (ADHD), Attention Deficient Disorder (ADD), Oppositional Defiant Disorder (ODD), or daytime irritability?	**YES**	**NO**
B BMI percent: Does the child's BMI percent fall into >85% or <5%? (*See conversion table)	**YES**	**NO**
A Age: Is your child younger than 4 or older than 16?	**YES**	**NO**
N Neuro/Muscular disorder: Does your child have abnormalities of muscle tone that can affect the airway?	**YES**	**NO**
G Genetic/ Congenital disorder: Does your child have a genetic or congenital disorder (for example-Ehlers Danlos Syndrome)?	**YES**	**NO**

***Conversion Table for BMI**

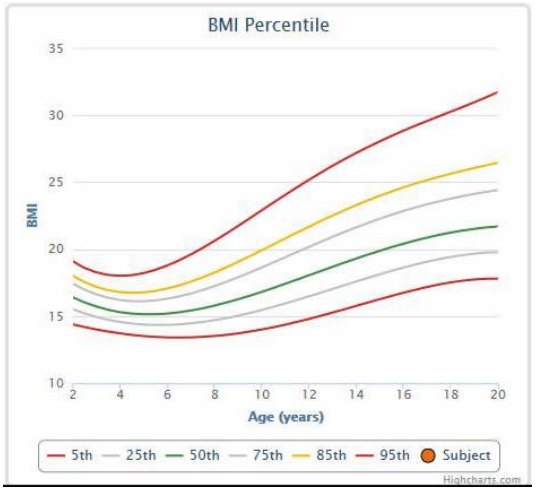

Figure 1: Pediatric-Modified-STOP-Bang

Pediatric Sleep Questionnaire: Sleep-Disordered Breathing Subscale

070129

Child's Name: _____ **Study ID #: _____**
Person completing form: _____ **Date: ____/____/____**

Please answer these questions regarding the behavior of your child during sleep and wakefulness. The questions apply to how your child acts in general during the past month, not necessarily during the past few days since these may not have been typical if your child has not been well. You should circle the correct response or *print* your answers neatly in the space provided. A "Y" means "yes," "N" means "no," and "DK" means "don't know."

1. WHILE SLEEPING, DOES YOUR CHILD:				
Snore more than half the time?……. ……..Y	N	DK	A2
Always snore? ……..Y	N	DK	A3
Snore loudly? ……Y	N	DK	A4
Have "heavy" or loud breathing? ……..Y	N	DK	A5
Have trouble breathing, or struggle to breathe? ……..……Y	N	DK	A6
2. HAVE YOU EVER SEEN YOUR CHILD STOP BREATHING DURING THE NIGHT? ……Y	N	DK	A7
3. DOES YOUR CHILD:				
Tend to breathe through the mouth during the day?……Y	N	DK	A24
Have a dry mouth on waking up in the morning? ……...Y	N	DK	A25
Occasionally wet the bed? ……...Y	N	DK	A32
4. DOES YOUR CHILD:				
Wake up feeling unrefreshed in the morning? ……Y	N	DK	B1
Have a problem with sleepiness during the day? ……..……...Y	N	DK	B2
5. HAS A TEACHER OR OTHER SUPERVISOR COMMENTED THAT YOUR CHILD APPEARS SLEEPY DURING THE DAY? ……..…Y	N	DK	B4
6. IS IT HARD TO WAKE YOUR CHILD UP IN THE MORNING? ……..…Y	N	DK	B6
7. DOES YOUR CHILD WAKE UP WITH HEADACHES IN THE MORNING?…..Y	N	DK	B7
8. DID YOUR CHILD STOP GROWING AT A NORMAL RATE AT ANY TIME SINCE BIRTH? ……..…Y	N	DK	B9
9. IS YOUR CHILD OVERWEIGHT? ……...Y	N	DK	B22
10. THIS CHILD **OFTEN**:				
Does not seem to listen when spoken to directly. ……...Y	N	DK	C3
Has difficulty organizing tasks and activities. ……..……...Y	N	DK	C5
Is easily distracted by extraneous stimuli. ……..……...Y	N	DK	C8
Fidgets with hands or feet or squirms in seat. ……....Y	N	DK	C10
Is "on the go" or often acts as if "driven by a motor". ……Y	N	DK	C14
Interrupts or intrudes on others (eg., butts into conversations or games). ……Y	N	DK	C18

Figure 2: Pediatric Sleep Questionnaire (PSQ)

Pediatric Symptom Checklist

Emotional and physical health go together in children. Because parents are often the first to notice a problem with their child's behavior, emotions or learning, you may help your child get the best care possible by answering these questions. Please mark under the heading that best fits your child.

			Never (0)	Sometimes (1)	Often (2)
1.	Complains of aches/pains	1			
2.	Spends more time alone	2			
3.	Tires easily, has little energy	3			
4.	Fidgety, unable to sit still	4			
5.	Has trouble with a teacher	5			
6.	Less interested in school	6			
7.	Acts as if driven by a motor	7			
8.	Daydreams too much	8			
9.	Distracted easily	9			
10.	Is afraid of new situations	10			
11.	Feels sad, unhappy	11			
12.	Is irritable, angry	12			
13.	Feels hopeless	13			
14.	Has trouble concentrating	14			
15.	Less interest in friends	15			
16.	Fights with others	16			
17.	Absent from school	17			
18.	School grades dropping	18			
19.	Is down on him or herself	19			
20.	Visits doctor with doctor finding nothing wrong	20			
21.	Has trouble sleeping	21			
22.	Worries a lot	22			
23.	Wants to be with you more than before	23			
24.	Feels he or she is bad	24			
25.	Takes unnecessary risks	25			
26.	Gets hurt frequently	26			
27.	Seems to be having less fun	27			
28.	Acts younger than children his or her age	28			
29.	Does not listen to rules	29			
30.	Does not show feelings	30			
31.	Does not understand other people's feelings	31			
32.	Teases others	32			
33.	Blames others for his or her troubles	33			
34.	Takes things that do not belong to him or her	34			
35.	Refuses to share	35			

Total score _____

Does your child have any emotional or behavioral problems for which she/he needs help? ( ) N ( ) Y
Are there any services that you would like your child to receive for these problems? ( ) N ( ) Y

If yes, what services?_____

www.massgeneral.org/psychiatry/assets/PSC-35.pdf 16th May, 2013

Figure 3: Pediatric Symptom Checklist (PSC)

Statistical Methods

All data from the three surveys were entered into a database on Research Electronic Data Capture (REDCap) tools hosted at Virginia Commonwealth University. REDCap is a secure, web-based software platform designed to support data capture for research studies.[21] All data from HSAT was analyzed by a sleep physician to determine the POSA status.

To determine the ability of the PM-STOP-Bang to detect high risk cases of sleep apnea, the PM-STOP-Bang scores were compared to the results from the Pediatric Sleep Questionnaire which is a validated instrument for identifying individuals at high risk for sleep apnea. To determine the ability of the PM-STOP-Bang and the PSQ to identify high risk cases for sleep apnea, the results from these two instruments were compared to the results of the HSAT. Patients with an AHI >1.5 were considered to have sleep apnea. For all comparisons, sensitivity, specificity, positive and negative predictive values and their 95% confidence intervals were reported. The demographic and clinical characteristics were compared between the experimental cases and HSAT results using Fisher's Exact test. T-test was used to compare age between the groups. Significance level was set at 0.05. SAS EG v.8.2 (SAS Institute, Cary, NC) for all analyses.

Results

A total of 10 patients were enrolled in the study. The patient demographics and clinical characteristics are provided in Table 1. Five subjects were considered at high risk for POSA based on the PM-STOP-Bang score and the other 5 individuals served as controls. The average PM-STOP-Bang score for the at high-risk group was 3.4 and ranged from 3 to 4 (out of a total possible 8 points) whereas the control group had an average score of 0.4 and ranged from 0 to 2. Interestingly, there was a significant difference in the distribution of adequate and inadequate transverse occlusion, with a higher percentage of inadequate transverse occlusion among those individuals who were classified as at high risk (80% vs 0%; p-value = 0.0467).

Table 1: Patient Demographics and Clinical Characteristics

		Case (n, %)	Control (n, %)	P-value
Age (mean, SD)		14.4, 2.4	13.8, 2.3	0.6964
Gender				
	Male	0, 0	3, 60	0.1667
	Female	5, 100	2, 40	
Classification				0.2857
	Class I	1, 20	2, 40	
	Class II	1, 20	3, 60	
	Class III	3, 60	0, 0	
Transverse				0.0476
	Adequate	1, 20	5, 100	
	Inadequate*	4, 80	0, 0	
Plane Angle				0.5238
	High**	4, 80	2, 40	
	Normal***	1, 20	2, 40	
	Flat****	0, 0	1, 20	

*Inadequate transverse defined as presence of a posterior cross bite of more than 2 teeth, **High plane angle FMA≥30° ***Normal plane angle defined as FMA between 21-29°, ****Flat plane angle FMA ≤20°

To determine the ability of the PM-STOP-Bang to identify individuals who are at high risk for SRBD, the results of the PM-STOP-Bang questionnaire were compared to those of the Pediatric Sleep Questionnaire (PSQ), which is the gold standard for determining individuals at high risk for SRBD (Table 2). Of the 5 subjects previously determined at high risk from the PM-STOP-Bang questionnaire, only four were also considered high risk from PSQ. Of the individuals who were deemed not at risk based on the PM-STOP-Bang scores (n=5, control), 2 were identified at high risk from PSQ. The PM-STOP-Bang had a sensitivity of 67% (95% CI: 29%, 100%) and a specificity of 75% (95% CI: 33%, 100%) for identifying high risk individuals. Furthermore, the positive and the negative predictive values were of 80% (95% CI: 45%, 100%) and 60% (95% CI: 17%, 100%), respectively.

Table 2: Accuracy of PM-STOP-Bang at Identifying Cases High Risk for Sleep Apnea Identified by Pediatric Sleep Questionnaire

		PSQ: High Risk for Sleep Apnea	
		Yes	No
PM-STOP-Bang	Yes	4	1
	No	2	3

Estimate % (95% CI)			
Sensitivity	67 (29,100)	**PPV**	80 (45, 100)
Specificity	75 (33,100)	**NPV**	60 (17, 100)

*HSAT: Home Sleep Apnea Test; PSQ: Pediatric Sleep Questionnaire, PPV: Positive Predictive Value; NPV: Negative Predictive Value

The results of the home sleep apnea test are provided in Table 3 and 4. A board-certified sleep specialist completed the analysis of the data captured from the one-night home sleep study for all 10 participants. Based on their Apnea-Hypopnea Index (AHI), three of the 10 subjects (Cases 1, 2, and 4) were recorded with sleep apnea diagnosis by the sleep physician. An AHI of greater than 1.5 was used for the diagnosis of sleep apnea. Case 3 was presumptively normal. However, this case was included in the table due to the significant increase in supine sleeping time.

All of these 3 subjects were also classified as individuals at high risk for OSA based on their PM-STOP-Bang scores. Comparing the PM-STOP-Bang to the HSAT results, the PM-STOP-Bang had a sensitivity of 100% (95% CI: 100%, 100%) and a specificity of 71% (95% CI: 38%, 100%). The positive predictive value was 60% (95% CI: 17%, 100%) and negative predictive value was 100% (95% CI: 100%, 100%). Surprisingly, the previously validated PSQ identified only 2 of these three individuals at high risk for sleep apnea when in fact all three were diagnosed with POSA with the HSAT. The PSQ also detected an additional 4 subjects at high risk who were not confirmed by HSAT as sleep apnea cases (False positive). The PSQ sensitivity was 67% (95% CI: 13%, 100%) and specificity 43% (95% CI: 6%, 80%). The PSQ positive predictive value was 33% (95% CI: 0%, 74%) and negative predictive value 75% (95% CI: 33%, 100%).

Table 3: Accuracy of PM-STOP-Bang and Pediatric Sleep Questionnaire (PSQ) for Detecting Sleep Apnea Cases Identified by Home Sleep Apnea Test

		HSAT (AHI>1.5) Yes	HSAT (AHI>1.5) No
PM-STOP-Bang	Yes	3	2
	No	0	5

Estimate % (95% CI)			
Sensitivity	100 (100, 100)	**PPV**	60 (17, 100)
Specificity	71 (38,100)	**NPV**	100 (100, 100)

		HSAT (AHI>1.5) Yes	HSAT (AHI>1.5) No
PSQ	Yes	2	4
	No	1	3

Estimate % (95% CI)			
Sensitivity	67 (13, 100)	**PPV**	33 (0, 71)
Specificity	43 (6, 80)	**NPV**	75 (33, 100)

*HSAT: Home Sleep Apnea Test; AHI: Apnea-Hypopnea index; PSQ: Pediatric Sleep Questionnaire; PPV: Positive Predictive Value; NPV: Negative Predictive Value

Table 4: Analysis from Sleep Professional

Case 1	Mildly abnormal, suggests mild OSA, hypopnea dominant + Increase in supine time (limited saturation data - may underestimate)
Case 2:	Mildly abnormal, suggests mild OSA, increase in supine time (limited saturation data - may underestimate)
Case 3:	Presumptively normal, increase supine time, (limited saturation data - may underestimate)
Case 4:	Mildly abnormal, suggests mild OSA, (limited saturation data - may underestimate).

Each study subject also completed the Pediatric Symptoms Checklist (PSC) and 2 of the 10 subjects met criteria for high risk for emotional or behavioral problems (PSC score≥28). Both of these subjects were also identified at high risk for SDBDs based on their PSQ score. However, these subjects were not confirmed as cases with sleep apnea by the physician based on their home sleep apnea test. Interestingly, the PM-STOP-Bang test did not identify these patients at high risk either indicating the ability of the new questionnaire in correctly identifying the true negative cases.

The associations between the patient characteristics and the results of the home sleep apnea test are provided in Table 5. There was no significant association between POSA diagnosis with HSAT for any demographic or orthodontic characteristics evaluated. Specifically, there was no significant difference associated with age, gender, molar classification, presence of transverse discrepancy, or plane angle and the diagnosis of POSA. However, when comparing the cases and controls enrolled in the study, the individuals at high risk (cases based on a PM-STOP-Bang score of 3 and higher) had a statistically significant tendency to have inadequate transverse dimension.

Table 5: The Associations between Patient Characteristics and Results of Home Sleep Apnea Test (HSAT)

		Yes (AHI>1.5)	No (AHI<1.5)	P-value
Age (mean, SD)		15.7, 2.3	13.4, 2.0	0.1563
Gender				0.4750
	Male	0, 0%	3, 43%	
	Female	3, 100%	4, 57%	
Classification				0.2000
	Class I	1, 33%	2, 29%	
	Class II	0, 0%	4, 57%	
	Class III	2, 67%	1, 14%	
Transverse				0.5000
	Adequate	1, 33%	5, 71%	
	Inadequate*	2, 67%	2, 29%	
Plane Angle				>0.999
	High**	2, 67%	4, 57%	
	Normal***	1, 33%	2, 29%	
	Flat****	0, 0%	1, 14%	

*Inadequate transverse defined as presence of a posterior cross bite of more than 2 teeth, **High plane angle FMA≥30° ***Normal plane angle defined as FMA between 21-29°, ****Flat plane angle FMA ≤20°

Discussion

The purpose of screening tests is to detect the presence of a disease in a subject. Based on the results of these tests, clinicians make decisions for further testing or treatment options. The four important measures for the reliability of these tests are sensitivity (true positive), specificity (true negative), positive predictive value (the percent of positive tests that are truly positive), and negative predictive value (the percent of negative tests that are truly negative). The current research aimed to validate the PM-STOP-Bang questionnaire for its use as a sleep apnea screening tool in a dental clinic setting, in orthodontics or pediatric dentistry. Specifically, its sensitivity, specificity, positive predictive value, and negative predictive value were investigated.

There are several tools that can be used to screen for SRBD and POSA. One reliable tool is the previously validated Sleep Related Breathing Disorder scale (SRBD scale), a subscale of the PSQ. This is a 22-item questionnaire that explores behavior, breathing, sleepiness, and other important signs of the disorder. It is designed to take 5-7 minutes to complete and a positive response to more than 7 questions indicates possibility of OSA. When compared with the PSG data, the PSQ instrument was previously reported to achieve a sensitivity of 85% and a specificity of 87% for identifying SRBD in children ages 2-18.[16]

Similarly, the Children's Sleep Habits Questionnaire is a 35-item parent-reported screening tool designed to assess behavioral and medically based sleep problems in school children.[22] In this 8 sleep-domain survey, each answer is scored on a 1 to 3 point scale. According to Owens et al., [22] this screening test has a sensitivity and a specificity of 80% and 72%, respectively, for the diagnosis of SRBD in children 4-10 years old.

Finally, another tool is the Sleep Disordered Inventory for Students which has 35 to 40 questions designed to determine if a child or adolescent has a high probability of a major sleep disorder requiring treatment.[23] This screening tool was reported to have a good sensitivity for children and adolescents (81% and 82%, respectively) and a very high specificity (91% and 95%, respectively) for identifying children and adolescents who would need a referral to a sleep specialist.[23]

In children, a common manifestation of pediatric obstructive sleep apnea is hyperactivity. Unfortunately, about 25% of children diagnosed with ADHD actually have undiagnosed POSA. In 2007, despite the PSQ being deemed as a reliable screening tool, to further validate this questionnaire, Chervin et al.[24] also utilized behavioral and cognitive testing with the Child Symptoms Inventory-4: Parent Checklist. This was done in an effort to screen for hyperactivity or emotional changes to behavior that may be due to an undiagnosed SRBD. This previously validated Child Symptom Inventory contains 108 items that screen children aged 5 to 12 years for a variety of emotional and behavioral disorders based on the Diagnostic and Statistical Manual of Mental Disorders, 4 Edition, (*DSM-IV).* Similarly, in the current study, to assess various emotional and various behavioral disorders related to POSA, the Pediatric Symptom Checklist (PSC) was administered. The PSC with only 35-item screening questionnaire can be easily completed by parents as opposed to 108 question survey. Furthermore, it is designed to help pediatricians in an outpatient setting identify school-age children with difficulties in psychosocial functioning. The PSC is a previously validated tool with a specificity of 68% and a sensitivity of 95% when compared to pediatrician ratings.[25] As stated earlier, unlike adults, some children with obstructive sleep apnea may exhibit hyperactivity in behavior. Therefore, it was

thought that the PSC may possibly be used as a shorter adjunct tool for dental clinicians to use when screening for POSA if there is a suspected behavior manifestation of the disorder. It was considered that a positive score may allow for increased ability to capture high risk children for the disease and make appropriate referrals to a sleep physician. In a similar thought process, screening with the PSC may also help identify children early enough to prevent a misdiagnosis of ADHD. In an effort to evaluate the merits of using the PSC as an adjunct screening tool that is sensitive enough to notice these behavior manifestations of POSA, PSC was completed for all study participants for statistical evaluation.

While all of these surveys are designed to be simple, the large number of items and various scoring protocols make them difficult and impractical to complete in a busy clinical setting. Additionally, none of these questionnaires take into account the physical or craniofacial characteristics such as a retrognathia or enlarged tonsils. As mentioned previously, approximately 4% of children have sleep apnea.[4] However, the majority of children remain undiagnosed because screening for POSA is challenging due to the complexity of in-center polysomnography tests. Therefore, the availability of a simple yet reliable screening questionnaire in an orthodontic clinic may help orthodontists to identify patients at risk and make early referrals to the appropriate physician.

The STOP-Bang has only 8 questions that can easily be answered and scored. Therefore, it may serve as a simple yet effective screening tool for OSA in a busy dental setting. Previous studies have reported high sensitivity (83.6%, 92.9% and 100% for mild, moderate and severe OSA, respectively) and moderate specificity for STOP-Bang to reliably detect sleep apnea in adults.[18] However, in a population younger than 18 years of age, the instrument was shown to

exhibit low sensitivity, and therefore, the STOP-Bang was not recommended when screening children for POSA.[26] Instead, the PM-STOP-Bang was modeled after the STOP-Bang screening questionnaire and the physical manifestations of pediatric high-risk markers of the disorder were taken into consideration.[19] For example, enlarged tonsils, considered to be the most common site of obstruction in children, was included in this modified version of STOP-Bang.[5,19]

The purpose of the current study was to evaluate the ability of the PM-STOP-Bang to identify high-risk children for POSA by comparing the results of the new questionnaire to those of the previously validated gold standard PSQ and a one-night home sleep test. When compared to the PSQ findings, the PM-STOP-Bang had a moderate sensitivity of 67% and moderate specificity of 75% for identifying high risk individuals. However, when the PM-STOP-Bang results were compared to those of the HSAT, the new screening tool was shown to be highly sensitive (100%) and moderately specific (76%) in the identification of patients at high risk for OSA.

Surprisingly the gold standard screening tool PSQ accurately identified only 2 of the 10 patients at high risk who were later confirmed to have POSA with the sleep test. However, PSQ missed 1 patient who actually had the disease. Additionally, the PSQ incorrectly identified 4 participants as high-risk for POSA while the PM-STOP-Bang incorrectly identified only 2 participants. Overall, the PSQ yielded a sensitivity of 71% and a specificity of 43%. On the other hand, the PM-STOP-Bang accurately identified all 3 subjects out of 10 at risk who were later shown to have POSA (a sensitivity of 100%). Therefore, the most remarkable result to emerge from the data was the higher sensitivity and specificity of the PM-STOP-Bang than that of PSQ.

A secondary aim of the study was to evaluate the ability of the PSC to be used as an adjunct screening tool for SRBD. In children, a common manifestation of POSA is hyperactivity. Unfortunately, about 25% of children diagnosed with ADHD actually have undiagnosed POSA. In 2007, despite the PSQ being deemed as a reliable screening tool, to further validate this questionnaire, Chervin et al.[24] also utilized behavioral and cognitive testing with the Child Symptoms Inventory-4: Parent Checklist. This was done in an effort to screen for hyperactivity or emotional changes to behavior that may be due to an undiagnosed SRBD. This previously validated Child Symptom Inventory contains 108 items that screen children aged 5 to 12 years for a variety of emotional and behavioral disorders based on the Diagnostic and Statistical Manual of Mental Disorders, 4th Edition, (*DSM-IV).*

In the current study, to assess various emotional and behavioral disorders related to POSA, the Pediatric Symptom Checklist (PSC) was administered. The PSC with only 35-item screening questionnaire can be easily completed by parents as opposed to 108 question survey. Furthermore, it is designed to help pediatricians in an outpatient setting identify school-age children with difficulties in psychosocial functioning. The PSC is a previously validated tool with a specificity of 68% and a sensitivity of 95% when compared to pediatrician ratings.[25] As stated earlier, unlike adults, children with obstructive sleep apnea may exhibit hyperactivity in behavior. Therefore, it was thought that the PSC may possibly be used as a shorter adjunct tool for dental clinicians to use when screening for POSA if there is a suspected behavior manifestation of the disorder. It was considered that a positive score may allow for increased ability to capture high risk children for the disease and make appropriate referrals to a sleep physician. In a similar thought process, screening with the PSC may also help identify children

early enough to prevent a misdiagnosis of ADHD. In an effort to evaluate the merits of using the PSC as an adjunct screening tool that is sensitive enough to notice these behavior manifestations of SRBD, PSC was completed for all study participants for statistical evaluation.

According to the PSC results of the current study, 2 of the 10 subjects were identified at high risk based on their reported emotional or behavioral problems (PSC score≥28). Both of these subjects were also at high risk for SRBD based on the PSQ instrument. However, neither subject had a PM-STOP-Bang score of 3 or higher nor was either positive for sleep apnea based on the home sleep apnea test. Therefore, it is plausible to conclude that with a high false positive rate, the PSC is not reliable to use as an adjunct in evaluating SRBD.

Maxillary morphological differences were previously reported between OSA and control subjects, indicating a potential etiological role in OSA.[27] However, the relationship between maxillary constriction and the etiology of OSA is not clear. Also, although the posterior transverse discrepancy and its effects on OSA have not been shown, statistically significant differences in palatal heights between OSA and control subjects have been previously reported.[27] Interestingly, within its limitations, this study reported posterior transverse discrepancy for 4 of the 5 patients who were also identified as “high risk” with the PM-STOP-Bang questionnaire. Furthermore, there was a significant difference in the distribution of adequate and inadequate transverse occlusion, with a higher percentage of inadequate transverse occlusion among those who were classified as POSA cases from the PM-STOP-Bang. However, the comparison of the demographic and orthodontic characteristics to the HSAT results did not show any statistically significant association. It is possible that due to the small sample size, the relationship between the maxillary morphological factors and the sleep apnea was not determined.

In future studies with a large sample size, in addition to administering the questionnaire, it would be useful to record clinical findings of malocclusion to assess a possible relationship between the presence of POSA and malocclusion, and its related features such as Class II malocclusion with a retrognathic mandible. If an association is present, then the PM-STOP-Bang instrument should be modified to include an additional question about clinical presentation, specifically maxillary and mandibular morphology such as retrognathic chin.

There were several limitations to the study. The most important limitation of the study was the small sample size. Due to the single center design, there was a possible sample selection bias. Also, although the study aimed to enroll 15 cases and 15 controls, due to COVID-19 closures, the number of subjects recruited were only 5 cases and 5 controls. The study had an IRB approval to begin in January of 2020 but was halted when clinics closed in March of 2020. The graduate orthodontic clinic did not re-open until July 2020. Furthermore, patients with urgent orthodontic needs were given priority when scheduling appointments for the opening. Finally, new patients were not accepted to the clinic for several months. Therefore, the actual predetermined number of subjects could not have been recruited. A future study with a larger sample size is warranted to validate the PM-STOP Bang as a potential screening instrument for accurately identifying children at high risk for sleep apnea.

An additional limitation of the study was regarding the use of the NOX-T3 home sleep monitor device in this sample population. While overall, the patients were able to use the device, some pulse oximetry data was limited. Also, the traditional pulse oximeter for children in this sample was found to be a significant challenge for compliance; therefore, in future studies the use of a wrist only HSAT would be a better option. Additionally, because the study sample

consisted of children, there were some subjects who turned off the device too soon. Although four hours of data recording was requested, two subjects completed only 3 hours and 30 minutes of sleep time. Therefore, for more definitive diagnosis, it would be ideal to repeat the HSAT testing with a different device preferably with a wrist pulse oximeter so that strict adherence to required sleep time (at least 4 hours) would be achieved.

In summary, the current study showed promising results that if administered in dental offices, the PM-STOP-Bang questionnaire may be an easy and reliable screening tool for the identification of pediatric patients at high risk for sleep apnea. Future studies with increased sample size and longer home sleep recording data are warranted.

Conclusion

Within its limitation the current study indicated that the PM-STOP-Bang with a sensitivity of 100% and a specificity of 71%, when compared with HSAT, may, one day be an alternative to the PSQ, a validated instrument.

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