Modified STOP-Bang Tool for Stratifying Obstructive Sleep Apnea Risk in Adolescent Children

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Abstract

Purpose

Obstructive sleep apnea (OSA) is prevalent in children and diagnostic polysomnography is costly and not readily available in all areas. We developed a pediatric modification of a commonly used adult clinical prediction tool for stratifying the risk of OSA and the need for polysomnography.

Methods

A total of 312 children (age 9–17 years) from phase 2 of the Tucson Children's Assessment of Sleep Apnea cohort study, with complete anthropomorphic data, parent questionnaires, and home polysomnograms were included. An adolescent modification of STOP-Bang (teen STOP-Bang) was developed and included snoring, tired, observed apnea, blood pressure $\geq$ 95th percentile, BMI > 95th percentile, academic problems, neck circumference >95th percentile for age, and male gender. An apnea-hypopnea index $\geq$ 1.5 events/hour was considered diagnostic of OSA.

Results

Receiver Operator Characteristic (ROC) curves for parent-reported STOP-Bang scores were generated for teenage and pre-teen children. A STOP-Bang score of < 3 in teenagers was associated with a negative predictive value of 0.96. ROC curves were also generated based upon child-reported sexual maturity rating (SMR; n = 291). The ability of teen STOP-Bang to discriminate the presence or absence of OSA as measured by the AUC for children with SMR $\geq$ 4 (0.83; 95%CI 0.71–0.95) was better than children with SMR < 4 (0.63; 95%CI 0.46–0.81; p = 0.048).

Conclusions

In community dwelling adolescents, teen STOP-Bang may be useful in stratifying the risk of OSA.

Introduction

In children and adolescents, obstructive sleep apnea (OSA) has a prevalence of 1.2–5.7% [1] and has been associated with behavioral, neurocognitive, learning and cardiovascular problems [2]. Diagnosis and management of OSA in children and adolescents can mitigate the adverse effects of OSA [3–5]. Unfortunately, children are infrequently screened in the primary care setting despite the potential benefits of early identification and treatment of OSA [6]. Additionally, pediatric polysomnography is costly and not widely available in areas without specialized pediatric centers. The issues regarding the under-diagnosis of OSA are further aggravated by the obesity epidemic and likely rising prevalence of OSA in children [7]. The validation of a negative predictive tool would allow appropriate selection of children for polysomnography in limited resource settings.

Several tools for the prediction of OSA in children have been developed. The Pediatric Sleep Questionnaire subscale for sleep-related breathing disorders, consists of 22 questions and was designed primarily as a research instrument rather than a clinical prediction tool aimed at reducing the need for polysomnography [8]. Additionally, it is a lengthy questionnaire and may not be easily administered in a busy "real-world" primary care setting. An alternative tool is the OSA-18 quality of life questionnaire, but this instrument does not accurately exclude moderate-to-severe OSA in children [9].

In adults, an instrument derived from eight easily administered questions called the STOP-Bang questionnaire has been shown to stratify the risk of OSA, with a negative predictive value of 93% for moderate OSA and 96% for severe OSA [10]. This tool was originally developed to predict the risk of OSA during pre-operative evaluation in adults [11]. We hypothesized that a modified version of the STOP-Bang tool may be useful as a clinical prediction tool to determine the need for polysomnography in children, particularly adolescents, who may have a more adult-like phenotype of OSA. In this study, we developed a modified STOP-Bang tool (teen STOP-Bang) for adolescent children and assessed the ability of this tool to stratify the risk of OSA in community-dwelling adolescents. This clinical prediction tool may be useful in a primary care setting to determine the need for polysomnography and further evaluation for OSA in adolescents.

Methods

Study Population

This study is obtained from a systematic analysis of phase 2 of the Tucson Children's Assessment of Sleep Apnea (TuCASA) cohort study. A detailed description of the TuCASA study has been previously described [12]. In brief, Caucasian and Hispanic children were recruited from the Tucson Unified School District, a large district representative of the Tucson population. A total of 503 children and their parents were consented and participated in phase 1 of the study. Approximately 5 years later (mean 4.7 years), 348 children participated in phase 2 of the study. We selected Phase 2 for the current analysis because both pre-teens and teenagers are adequately represented. The TuCASA study was approved by the University of Arizona

Institutional Review Board (IRB approval # 0300000227) and the Tucson Unified School District Research Committee. Prior to undergoing study-related procedures, written informed consent was obtained from the parents and minor's assent was also obtained. Subjects underwent home polysomnograms, and both children and their parents filled out sleep questionnaires pertaining to the children's sleep habits. Anthropomorphic measurements were also obtained during the home visit.

Polysomnography

A single, unattended overnight polysomnogram was obtained with the Compumedics PS-2 system (Abbotsford, Victoria, Australia). The following signals were acquired as part of the TuCASA montage: C3/A2 and C4/A1 electroencephalogram, right and left electrooculograms, a bipolar submental electromyogram, thoracic and abdominal displacement (inductive plethysmography), airflow (nasal/oral thermistor), nasal pressure cannula, finger pulse oximetry, electrocardiography (single bipolar lead), snoring microphone, body position (Hg gauge sensor), and ambient light levels [12]. A previous study examined the characteristics of the Compumedics PS-2 system in the TuCASA cohort. The initial pass rate was 91%, which improved to 97% when 9 children who failed on the first night of recording completed a second study which was acceptable. The Compumedics system was validated by performing conventional polysomnography in a subset of subjects, which showed good correlation between the two studies [12].

Scoring of sleep was performed by a single registered polysomnographic technologist using standard criteria[13]. Apneas were scored if the amplitude of the thermistor airflow decreased below at least 25% of the amplitude of baseline breathing, and lasted for more than 6 seconds or 2 breath cycles [2, 12, 14]. Hypopneas were designated if the amplitude of any respiratory signal decreased below 70% of the amplitude of baseline, was associated with a 3% oxygen desaturation, and the thermistor signal did not meet the criterion for apnea [2, 12]. Central events were marked if no displacement was noted on both the chest and abdominal inductance channels. However, central events that occurred after movement were not included. The Apnea Hypopnea Index (AHI) was defined as the number of apneas and hypopneas per hour of total sleep time. For data analysis, an AHI $\geq$1.5 was considered to indicate OSA, based on prior research identifying this threshold as significantly abnormal [15].

Anthropometry

All anthropomorphic measures were obtained during the home visit, the evening of the subject's polysomnograms. Weight was obtained as the average of 3 measurements, to the nearest 0.1 Kg. Height was obtained using a folding ruler on a level surface after removal of shoes, and the head in the Frankfort plane. The average of three measurements to the nearest 0.1 cm was used for data analysis. Body mass index (BMI) was calculated as weight in kilograms divided by height in meters squared. BMI percentiles for age and gender were calculated using Center for Disease Control growth charts [16]. To obtain blood pressures, arm circumference was first measured, and then an appropriate sized blood pressure cuff was selected. Seated blood pressures were obtained in triplicate. The average of the 2nd and 3rd measurements was used for data analysis. Blood pressure percentiles based on height, age, and gender were determined using CDC data [17]. The method of Lohman, Roche and Martorel was used to measure the circumference of the neck [18]. The participant sat upright with the head in the Frankfort Horizontal Plane and an inelastic tape was applied around the neck just below the laryngeal prominence. Then the neck circumference measurement was made perpendicular to the long axis of the neck (which was not necessarily in the horizontal plane). The pressure on the tape was kept

to the minimum required to maintain skin contact. The neck circumference measurement was completed in less than 5 seconds, to avoid participant discomfort and was measured to the nearest 1/2 cm and rounded up. Neck circumference percentiles for age and gender were determined through use of a reference data set of healthy weight children [19]. Self-reported sexual maturity ratings (SMR) were also collected using a validated pubertal self-assessment questionnaire [20] without parental supervision that consists of simple line drawings based on photographs of the Tanner and Marshall standards [21, 22].

Modified STOP-Bang questionnaire

The following were included as the teen STOP-Bang tool: Snoring (How often does your child snore loudly?), Tired (Is your child sleepy during the daytime?), Observed apnea (Does your child stop breathing during sleep?), systolic or diastolic blood pressure greater than or equal to 95th percentile for height and age, BMI greater than 95th percentile for age, Academic problems (Does your child have learning problems?), Neck circumference greater than 95th percentile for age, and male Gender. Answers to the questions had choices that included—don't know, never, rarely, occasionally, frequently or almost always. The responses were collapsed into a positive or negative response in the following manner. Frequently or almost always was considered as a positive response and responses of don't know, never, rarely, or occasionally were all considered as a negative response. The number of positive responses was calculated as the subject's teen STOP-bang score. In the teen STOP-Bang questionnaire the question regarding academic problems was inserted in the place of age greater than 50 years that was used the original version of the STOP-Bang questionnaire for use in adults, because learning problems have been shown to be a symptom of sleep-disordered breathing in children [23].

Data Analysis

Statistical analysis was performed using SPSS Version 22 (IBM, Armonk, NY). Data was assessed for a normal distribution using a one sample Kolmogorov–Smirnov test. Receiver operating characteristic (ROC) curves were generated based on teen STOP-Bang scores. Separate ROC curves were generated and compared based on age as well as SMR status. This was done to test the hypothesis that the teen STOP-Bang would be more accurate in adolescent children. Both age and SMR status were used as age is a more useful clinical measure, while SMR is a more accurate measure of progression through puberty. An SMR of 4 was used as a split point to differentiate children in earlier vs. later phase of puberty. This SMR correlates with an average age of approximately 13–14 years [21]. Sensitivity, specificity, negative predictive value, positive predictive value, and likelihood ratios were calculated by generating 2x2 contingency tables. Prevalence-adjusted likelihood ratios were also calculated, as the prevalence of OSA in the cohort was 0.08 and non-adjusted likelihood ratios may be misleading when sample prevalence is substantially different from 0.50 [24, 25]. Wilcoxon-signed rank tests were used to determine significance for comparisons in paired data, and Mann-Whitney U tests were used for un-paired data, as the teen STOP-Bang scores were not normally distributed. A p-value of less than 0.05 was considered significant.

Results

Of the 348 children in the second phase of the TuCASA study, 312 had complete anthropomorphic data, complete questionnaires and acceptable polysomnograms, and were included in data analysis. Demographic data is described in **Table 1**. Twenty-five children were found to have OSA defined as AHI $\geq$ 1.5/hour measured by polysomnography at the participants'

Table 1. Demographics of participants.

	No OSA AHI <1.5/hour	OSA AHI ≥ 1.5/hour	P value
	(n = 287)	(n = 25)	
Ethnicity			0.43
Caucasian	184 (64%)	18 (72%)	
Hispanic	103 (36%)	7 (28%)	
Age, years	13.3 (9.9–17.6)	13.6 (10.5–17.4)	0.48
Gender			.07
Male	141 (49.1%)	17 (68%)	
Female	146 (50.9%)	8 (32%)	
BMI Percentile	67.4 (35.2–89.7)	94.3 (81.6–97.8)	<0.001
Neck percentile	95 (75–95)	95 (82.5–95)	0.9
Teen STOP-Bang score			
Parental report	1 (1, 2)	3 (1.5, 3)	<0.001
Self report	1 (1, 2)	2 (1.5, 3)	0.003
AHI	0.2 (0.1, 0.4)	2.2 (1.8, 3.4)	<0.001
ODI	0.3 (0.1, 0.6)	2.4 (1.9, 3.5)	<0.001
Arousal Index	5.9 (4.6, 7.3)	7.4 (5.3, 10.9)	<0.001

OSA = obstructive sleep apnea; AHI = apnea-hypopnea index; BMI = body mass index; ODI = 3% oxygen desaturation index; Arousal index calculated as total number arousals divided by hours of total sleep time. Data is presented as proportion or median, 25th and 75th quartiles.

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home. OSA was mild (AHI between 1.5 and 5) in all but five cases, with one case of severe OSA (AHI of 31).

Receiver operating characteristic (ROC) curves were generated for both parent-reported as well as child-reported teen STOP-Bang scores (Fig 1). ROCs were also generated separately for pre-teen (9–12 yr) and teenage children (≥13 yr). For teenage children, the area under the curve (AUC) for the ROC was 0.77 (95% confidence interval [CI]; 0.65, 0.90) for parent-reported score, and 0.70 (95% CI; 0.56, 0.84) for child-reported score. For pre-teen children, the AUC was 0.68 (0.51, 0.85) for parent-reported score and 0.64 (0.48, 0.81) for child-reported score. The p-value for the difference between the AUC of ROCs parent-reported score for pre-teens and teenagers was 0.42. A teen STOP-Bang score of ≥ 3 in teenagers was associated with a sensitivity of 64%, specificity of 82%, positive predictive value of 0.24 and negative predictive value of 0.96. Sensitivity and specificity were higher in teenage children when compared to pre-teen children. Test characteristics for different teen STOP-Bang scores for are listed in Table 2.

Sensitivity analysis was performed for AHI thresholds of 1 and 3 events per hour. For both thresholds, the teen STOP-Bang AUC was inferior. The AUC for a threshold of 1 was 0.68 (95% CI; 0.60–0.77), with 46 children with an AHI ≥ 1. The AUC for a threshold of 3 was 0.73 (95% CI; 0.53–0.92), with 7 children with an AHI ≥ 3. We also performed sensitivity analysis evaluating different BMI percentile cut points of 90th and 99th percentile, but found that these did not improve test characteristics.

To determine whether pubertal status influences the test characteristics of the teen STOP-Bang, we generated ROC curves stratified by self-reported sexual maturity rating (SMR threshold of 4). For children with a SMR ≥ 4, the parent-reported teen STOP-Bang AUC was 0.83 (95%CI; 0.71, 0.95). For children with a SMR < 4, the parent-reported teen STOP-Bang AUC was 0.63 (95% CI; 0.46–0.81). The AUC for the ROCs stratified by sexual maturity were significantly different (p = 0.048).

All

Pre-teens
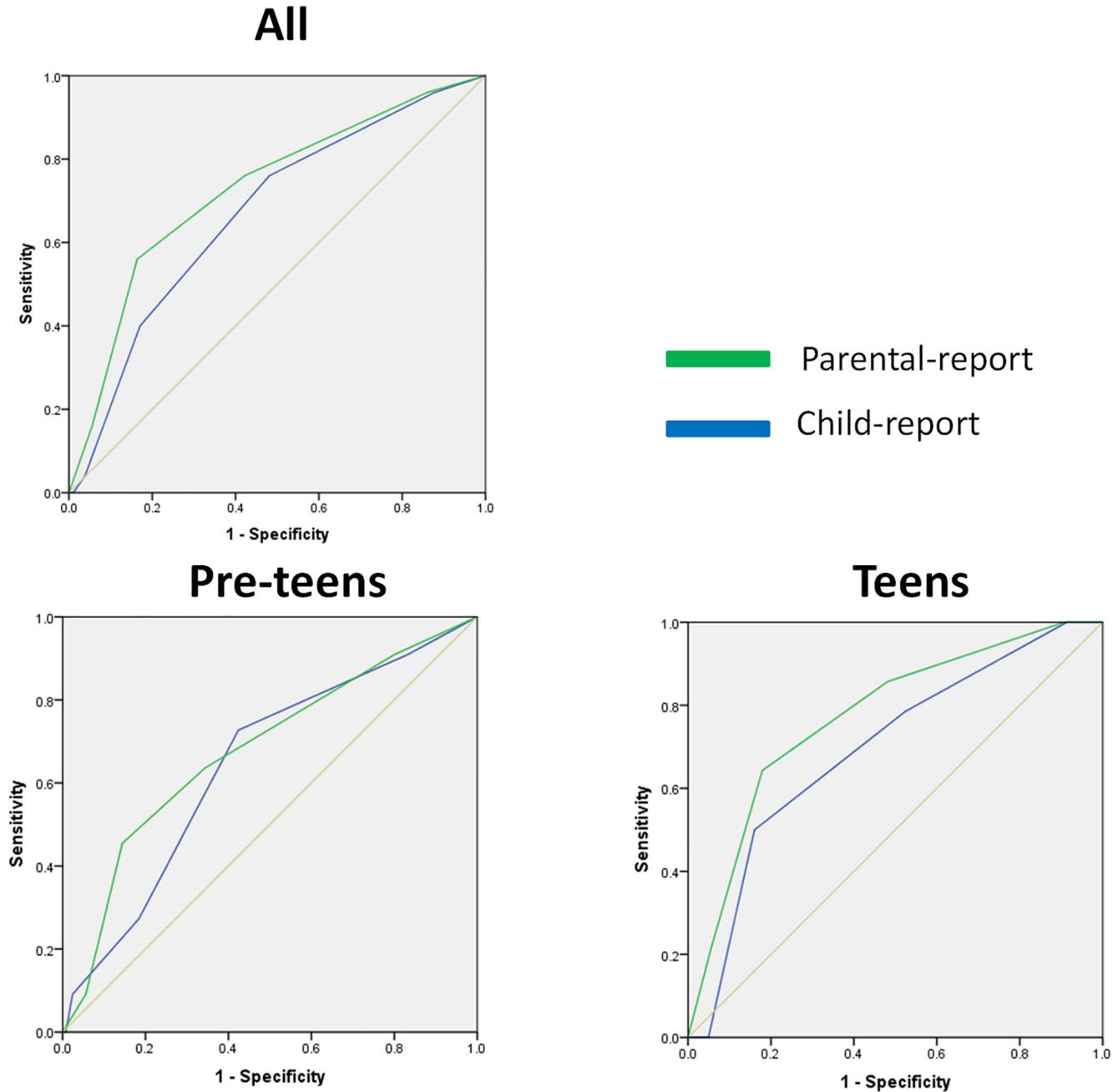
Teens

Fig 1. Receiver operating characteristics of teen STOP-Bang. Receiver operating characteristics (ROC) for modified STOP-Bang scores that were child-reported (blue lines) or parent-reported (green line) in discriminating presence of obstructive sleep apnea (defined as an apnea-hypopnea index $\geq$ 1.5 / hour). The ROC area under the curve for all (top left), pre-teens (bottom left) and teens (bottom right) are shown.

The ability of parent-reported scores to predict OSA was compared to that of child-reported scores. For teenage children, there was no difference between the parent- and child-reported scores (p = 0.97). For pre-teen children, a significant difference was seen between parent and child-reported scores (p = 0.028; Table 3).

Table 2. Test Characteristics of teen STOP-Bang Scores with sensitivity analysis for various thresholds.

Variable	Sensitivity	Specificity	PPV	NPV	LR+	LR-	LR+[P]	LR-[P]
Teen STOP-Bang score of ≥3								
All	**0.56**(0.35–0.75)	**0.84**(0.79–0.88)	**0.20**(0.15–0.24)	**0.96**(0.92–0.98)	**3.4**(2.2–5.3)	**0.5**(0.3–0.8)	**0.3**(0.2–0.5)	**0.05**(0.03–0.08)
Teens	**0.64**(0.36–0.86)	**0.82**(0.75–0.88)	**0.24**(0.12–0.41)	**0.96**(0.92–0.98)	**3.6**(2.2–6.0)	**0.4**(0.2–0.9)	**0.3**(0.2–0.6)	**0.04**(0.02–0.09)
Preteens	**0.45**(0.18–0.75)	**0.86**(0.78–0.91)	**0.21**(0.08–0.44)	**0.95**(0.88–0.98)	**3.2**(1.5–6.9)	**0.6**(0.4–1.1)	**0.3**(0.1–0.6)	**0.06**(0.03–0.12)
Teen STOP-Bang score of ≥2								
All	**0.76**(0.54–0.89)	**0.58**(0.52–0.64)	**0.13**(0.09–0.21)	**0.97**(0.92–0.99)	**1.8**(1.4–2.3)	**0.4**(0.2–0.8)	**0.2**(0.1–0.2)	**0.04**(0.02–0.08)
Teens	**0.86**(0.56–0.97)	**0.52**(0.44–0.60)	**0.13**(0.07–0.23)	**0.98**(0.91–0.996)	**1.8** (1.4–2.3)	**0.3** (0.1–1.0)	**0.2** (0.1–0.3)	**0.02**(0.006–0.09)
Preteens	**0.64**(0.32–0.88)	**0.66**(0.57–0.74)	**0.14**(0.06–0.27)	**0.95**(0.88–0.98)	**1.8** (1.1–3.1)	**0.6**(0.3–1.2)	**0.2**(0.1–0.3)	**0.05**(0.02–0.13)
Teen STOP-Bang score of ≥4								
All	**0.16**(0.05–0.37)	**0.94**(0.91–0.97)	**0.20**(0.07–0.44)	**0.93**(0.89–0.95)	**2.9**(1.0–7.9)	**0.9**(0.7–11)	**0.3**(0.1–0.6)	**0.08**(0.0.05–0.12)
Teens	**0.21**(0.06–0.51)	**0.94**(0.89–0.97)	**0.25**(0.07–0.57)	**0.93**(0.88–0.96)	**3.8**(1.1–12.6)	**0.8**(0.6–1.1)	**0.3**(0.1–0.9)	**0.07**(0.04–0.13)
Preteens	**0.09**(0.005–0.43)	**0.94**(0.88–0.98)	**0.13**(0.007–0.53)	**0.92**(0.86–0.96)	**1.6**(0.2–12.0)	**1.0**(0.8–1.2)	**0.1**(0.02–0.9)	**0.08**(0.05–0.15)

Test performance of teen STOP-Bang with various score cut-off thresholds. A teen STOP-Bang score of ≥3 has the best test characteristics. Mean values are in bold font, and 95% confidence intervals are within parenthesis; PPV = Positive Predictive Value; NPV = Negative Predictive Value; LR+ = Likelihood ration of a positive test; LR- = Likelihood ratio of a negative test; LR+[P] = Likelihood ratio of a positive test weighted for prevalence; LR-[P] = Likelihood ratio of a negative test weighted for prevalence. In general an LR+[P] of greater than 5 and LR[–]P of less than 0.2 are considered as indicative of good test characteristics.

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Finally, we evaluated the necessity of including neck circumference as a measure in teen STOP-Bang. While BMI and blood pressure are routinely measured in the pediatric office, neck circumference is not a standard measure, and inclusion of this variable may limit implementation into clinical practice. We generated a new score using all teen STOP-Bang variables except neck circumference (teen STOP-Bag). We then compared the performance of teen STOP-Bag and teen STOP-Bang. We found no significant differences between teen STOP-Bag and teen STOP-Bang. For all children, the AUC of teen STOP-Bag was 0.734 compared to the teen STOP-Bang AUC of 0.728 (p = 0.80).

Discussion

In this community-based cohort of 312 children, the teen STOP-Bang instrument was found to have a high negative predictive value in assessing the likelihood of obstructive sleep apnea. The

Table 3. Parent- versus self-reported Teen STOP-Bang scores.

	Sample size	Parent or caregiver reported score	Child-reported score	P value
All	312	1 (1, 2)	2 (1, 2)	0.17
Preteen	136	1 (1, 2)	1 (1, 2)	0.03
Teen	176	2 (1, 2)	2 (1, 2)	0.97

Data is presented as proportion or median, 25th and 75th quartiles. Child and parent-reported scores are similar based on the median and inter-quartile range, however, a significant difference was seen in pre-teens because individual scores did not match well between parent and child dyads.

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prevalence adjusted negative likelihood ratio is 0.05 (Table 2) which is consistent with an excellent negative predictive value. Given the sample prevalence of 8%, a teen STOP-Bang of less than 3 would reduce the odds of a child having sleep apnea from 1 in 13.5 to 1 in 263. Such a high negative predictive value makes the teen STOP-Bang a useful instrument to mitigate the need for obtaining a polysomnogram to further evaluate the presence of OSA. This may be of particular utility in children with learning problems or symptoms of attention-deficit hyperactivity disorder, as these symptoms may be related to sleep-disordered breathing in children [23, 26]. A low teen STOP-Bang score (less than 3) would suggest that OSA is unlikely. Additionally, a teen STOP-Bang score of 4 or greater did show increased specificity compared to a score of 3 (94% vs. 82–86%), suggesting that increasing teen STOP-Bang scores may be associated with an increased likelihood of OSA.

One previous study has evaluated the use of a modified STOP-Bang in children [27] and reported a sensitivity of 12% and specificity of 90% (negative predictive value of 0.67, positive predictive value of 0.37). There are several differences that likely explain differences between the study by Kadmon et al and our findings [27]. In the study by Kadmon et al, BMI and neck circumference cut-offs were absolute numbers and not age-adjusted percentiles. Additionally, elevated blood pressure was assessed by self-report, and not measured by American Heart Association guidelines as performed in our study. Also, we included the additional factor of academic problems as a question in substitution of age>50 that distinguishes our study from the prior study. Finally, the study by Kadmon et al included a wide age range (7–18 years old), whereas our data has shown that this tool is more accurate in older children when compared to younger children. The more adult-like phenotype of OSA in adolescents [28] may have contributed to the better test characteristics of the teen STOP-bang instrument in our study.

We found that the teen STOP-Bang instrument was more accurate in children who have progressed further through puberty. This is likely due to differences in the adolescent compared to pre-adolescent phenotype of OSA that more closely resembles that of adults, which is the population in which the STOP-Bang tool was originally developed [29]. Previous studies have shown that post-pubertal children show sex-related differences that are not seen in younger children [30]. A recent cohort study in older adolescents has also shown that risk factors for sleep-disordered breathing are different between younger children and adolescents [28].

An additional finding of our study was that while there was no significant difference in the results of parent report compared to teenage child report, there was a significant difference in pre-teen report compared to parent report. This finding may reflect that older adolescents may be more aware of their symptoms, or are more accurate reporters, although further research would be needed to definitively determine this. Prior research in this area, specifically in health-related quality of life, has shown conflicting results, with some studies showing good correlation between child self-report and parent-report [31, 32], while others have shown significant differences [33, 34].

Our study has several limitations. Our study included only Hispanic and Caucasian children by design, so the validity of these results in other ethnicities is unknown. There was a relatively small population (n = 25) of children with OSA in our study, validation of these results in a larger cohort would be useful. There were few children with moderate or severe OSA, which prevented analysis of teen STOP-Bang performance specifically in moderate-severe OSA, which has been shown to be of use in the adult STOP-Bang[35]. While there was a significant difference between ROC curves generated based on pubertal status, there was no difference in ROC curves by age. This is likely due to children undergoing puberty at varying ages, which results in a heterogeneous sample when age is used as a surrogate for pubertal status. However, age is likely a more implementable clinical measure than measuring SMR in children to screen for sleep-disordered breathing. Additionally, SMR was self-assessed based on a pictorial scale,

and it is possible adolescents may not be accurate in their assessment, however, several studies have shown that adolescents can accurately measure pubertal development to within one sexual maturity rating stage of a trained examiner [36–38]. Finally, while increased neck circumference is a well-known risk factor for OSA in adults[39], and has been shown to be a predictive factor in children[40], there was not a significant difference in neck circumference seen in our population between children with and without sleep apnea. This may explain why the omission of neck circumference did not worsen the teen STOP-Bang test performance.

A major strength of this study was that it was conducted in a community-based cohort. Previous tools have been developed using children referred for polysomnograms, a population with a higher pre-test prevalence of OSA [8, 41]. The use of a community cohort improves the generalizability of our study to a primary care setting. Additionally, our study focused specifically on adolescent children; to our knowledge, there is no previously described tool for assessing the risk of OSA in this age group.

Conclusions

We have developed a modification of the STOP-Bang tool for use in adolescent children. In community dwelling adolescents, the teen STOP-Bang questionnaire may be useful in stratifying the risk of OSA and determining the need for further evaluation for OSA and polysomnography. The implementation potential in general pediatric clinics, however, needs to be determined. Moreover, future prospective studies are needed to determine the validity of our teen STOP-Bang tool, particularly in other races and ethnicities which were not included in our study.

Supporting Information

S1 Dataset. TuCASA data utilized for the development of the teen STOP-Bang.
(SAV)

Author Contributions

Conceived and designed the experiments: SQ JG WM DC SP. Performed the experiments: SQ JG WM. Analyzed the data: DC SP. Contributed reagents/materials/analysis tools: SQ JG WM DC SP. Wrote the paper: SQ JG WM DC SP.

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