Diagnosis of pediatric obstructive sleep apnea hypopnea syndrome using a risk score based on polysomnography sleep video recordings: a pilot study

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Abstract

Objectives: Because access to sleep recordings is limited, there is a need for new reliable diagnostic tools for pediatric obstructive sleep apnea-hypopnea syndrome (OSAHS) diagnosis. A score calculated from a 30 minutes-home sleep videotape recording has already been proposed in 1996 with interesting results. The main objective of this pilot study was to assess the reliability of a similar score applied to reference PSG video recordings and calculated on two different time windows (30 and 10 minutes). **Methods:** Sixteen children suspected of OSAHS, aged between two and ten years, underwent video recording during overnight PSG. Video analysis was made during the second complete sleep cycle. A 30-minute risk score (RS30) and a 10-minute risk score (RS10) were established by analyzing seven parameters. The RS30 and RS10 were correlated with clinical examination data, a sleep questionnaire, the obstructive-apnea-hypopnea index (OAHI) and the oxygen desaturation index (ODI) from synchronized PSG results. **Results:** There was a significant correlation between both the RS30 and RS10, the OAHI and ODI. A RS30 [?] 6.09 was predictive of an OAHI [?] 5 per hour with a sensitivity of 83% and a specificity of 90%. A RS10 [?] 6.50 was predictive of an OAHI [?] 5 per hour with a sensitivity of 67% and a specificity of 100%. **Conclusion:** A risk score based on PSG video recordings shows a good correlation with PSG results, confirming previous reports. Further work should focus on applying this risk score to home sleep video recordings for the diagnosis of pediatric OSAHS.

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Results: There was a significant correlation between both the RS30 and RS10, the OAHI and ODI. A RS30 [?] 6.09 was predictive of an OAHI [?] 5 per hour with a sensitivity of 83% and a specificity of 90%. A RS10 [?] 6.50 was predictive of an OAHI [?] 5 per hour with a sensitivity of 67% and a specificity of 100%.

Conclusion: A risk score based on PSG video recordings shows a good correlation with PSG results, confirming previous reports. Further work should focus on applying this risk score to home sleep video recordings for the diagnosis of pediatric OSAHS.

Introduction

Pediatric obstructive sleep apnea-hypopnea syndrome (OSAHS) is the most severe clinical entity in the obstructive sleep disordered breathing (OSDB) spectrum [1]. In children without comorbidities (type 1 OSAHS [2]), it is often caused by adeno-tonsillar (AT) hypertrophy, leading to upper airway obstruction [3, 4].

OSAHS is responsible for fragmented nonrestorative sleep, which affects the child's behavior and school performance, delays somatic growth, and causes cardiovascular and metabolic problems that can last into adulthood [5, 6].

Clinical assessment of OSAHS is based on clinical findings (physical examination and medical history collection) preferably supplemented by a sleep questionnaire [4]. In 2012, Spruyt *et al* devised a severity hierarchy score (SHS) based on a set of six questions [7]. The SHS provides an easy-to-apply clinical tool, which correlates with PSG results and accurately predicts moderate to severe OSAHS (AHI [?] 5) with the score value of $\frac{1}{2}$ 2.75. A French version of the SHS was validated in 2017 [8].

In children without comorbidities presenting with OSDB, an objective assessment of OSAHS is mandatory prior to tonsillectomy in the following situations [9]: (i) doubt about procedure efficiency, especially in case of discordance between tonsillar size on physical examination and the reported severity of sleep-disordered breathing; (ii) additional surgical or anesthetic risk, such as hemostasis disorder or cardiac condition.

Overnight polysomnography (PSG) in a sleep laboratory is the reference method for the diagnosis of OSAHS. Respiratory polygraphy (RP) is a reliable alternative to in-lab PSG [10, 11], but tends to underestimate OSAHS severity, and also raises the issue of its availability. Sleep recordings allow identification of central and obstructive apneas, hypopneas, and sleep arousal due to respiratory effort. These events are assessed by the 2012 and 2015 criteria of the American Academy of Sleep Medicine (AASM) [12, 13]. OSAHS severity is defined by the obstructive apnea-hypopnea index (OAHI) per hour of sleep: mild OSAHS for an OAHI between 1.5 and 5, moderate between 5 and 10 and severe for over 10 [14, 15].

Access to sleep recordings is limited because they are cost and time-consuming [16]. Only 10% of snoring children undergoing adenotonsillectomy for OSAHS are offered a preoperative sleep recording [17]. Therefore, there is a need to develop reliable alternative tools for pediatric OSAHS diagnosis. Sivan *et al.* suggested in 1996 that home sleep video recording could serve as a reliable tool for OSAHS screening in children [18]. The analyses of 30-minute video recordings were highly correlated with the PSG results, showing a sensitivity of 94% and a specificity of 68%. Video scores ¿ 10 appeared to be predictive of OSAHS whereas scores ; 5 indicated normal sleep.

The aim of this pilot study was to assess the reliability of a similar scoring system, calculated on two different time windows (30 and 10 minutes), and applied to a complete sleep cycle obtained on PSG video recordings.

Our main objective was to look for a correlation between the risk score obtained on 30 minutes (RS30) and 10 minutes (RS10) from the PSG video recordings and the OAHI from the same synchronized PSG.

Our secondary objectives were to look for a correlation between the RS30/RS10 and: (i) the oxygen desaturation index (ODI) from the same PSG; (ii) the SHS questionnaire results.

Methods

This study was approved by the local ethics committee and registered under the reference 2021/CE. It was conducted in accordance with the principles of the Declaration of Helsinki on medical research involving human subjects.

Population

This was a retrospective study of pediatric patients who underwent PSG in the sleep laboratory of the university hospital of Clermont-Ferrand, France, between September 2018 and March 2019.

All children selected for the analysis were referred for a suspicion of OSAHS. They were aged two to ten years and had tonsillar hypertrophy graded [?] 2 on the Brodsky scale [19]. Patients with OSAHS type 2 and type 3 or with other sleep disorders than OSAHS were excluded. Sixteen children (11 girls and 5 boys) aged two to ten years (mean age 4.8 ± 1.8) were enrolled in the study. With an average body mass index (BMI) of 16.1 ± 2.4 kg/m², they all had a BMI-for-age between the 5th and 95th percentiles.

Prior to sleep recording, all patients had undergone a comprehensive clinical examination performed by an otolaryngologist, consisting in measurements of weight in kilograms (kg) and height in meters (m), calculation of BMI (kg/m²) and tonsil grading according to the Brodsky scale [19]. All parents of included children completed the French version of the SHS questionnaire.

Video and polysomnography

All children underwent overnight PSG in a sleep unit. Heart rate, electrocardiographic signals, electroencephalographic (EEG) and electrooculographic activities, thoracic and abdominal respiratory movements, respiratory rate, arterial oxygen saturation (SaO2) and airflow were continuously measured throughout the sleep period. Along with the PSG recording, a video was recorded with a Sony \mathbb{R} Ipela SNC-ER550 rotating camera coupled to an infrared device to allow recording in low light conditions without disturbing the child's sleep. The video was synchronized to PSG. The camera was placed on the wall opposite the child's bed, at 2.5 meters and positioned to focus on the child's head and chest. Sheets and blankets were avoided as often as possible, and if needed for the child to fall asleep, removed as soon as he slept. A sound recording, assessed in decibels, was also made at the same time.

Interpretation of the PSG data, blinded to the clinical examination and SHS, was carried out according to the AASM criteria of the AASM [12, 13]. Patients were classified in four OSAHS severity groups: absence (OAHI < 1.5 per hour), mild (OAHI 1.5-5), moderate (OAHI 5-10) and severe (OAHI [?] 10). The ODI was calculated as the number of decreases in SaO2 [?] 3% [20].

The PSG videos were evaluated throughout the second sleep cycle identified by EEG signals as the period between the end of the first rapid eye movement (REM) sleep and the end of the second one. The analysis was blinded to OAHI, ODI and SHS values. The videos were interpreted by an otorhinolaryngologist investigator according to a slightly modified version of Sivan's scoring system. The following parameters were considered: presence of inspiratory noise, type of inspiratory noise, movements during sleep, number of waking episodes, number of apneas, chest retractions, and mouth breathing (Table 1). The movements of more than four body parts (four limbs, head, and chest) were considered as a whole-body movement. Any opening of the eyes or a shift to a sitting position were counted as a waking episode. All events characterized by breathing interruption with persistent respiratory effort for more than two respiratory cycles were counted as apneas. Diaphragmatic paradoxical movements were analyzed together with the chest retractions. For the third item (movements during sleep), we set the threshold for numerous movements at 2 rather than 3 in Sivan's original scoring system. Likewise, for the fifth item (number of apneas), we set the threshold of numerous apneas at 2 rather than 3.

The score obtained for the second sleep cycle of each child was then brought back first to a 30-minute duration, leading to a 30-minute risk score (RS30), then to a 10-minute duration, leading to a 10-minute risk score (RS10). The number of apneas, waking episodes and movements was averaged for a 30-minute period using the following formula: (number of apneas + number of waking episodes + number of movements) over 30 minutes = (number of apneas + number of waking episodes + number of movements) over the entire second sleep cycle*30/ length of the second sleep cycle in minutes. The same calculations were performed for a 10-minute period. For the other variables (inspiratory noise, intercostal retractions, mouth respiration), only their intermittent or continuous nature was noted, hence their value was unchanged irrespective of the duration.

Statistical analyses

Statistical analyses were performed with Stata software (version 15, StataCorp, College Station, Texas, USA) and R 3.5.1 (http://cran.r-project.org/). All tests were two-sided with an alpha level set at 5%. Categorical variables were expressed as number of children and associated percentages, and continuous variables as mean +- standard deviation or as median [25th; 75th percentiles], according to the statistical distribution and the results previously reported in the literature. Factors associated with OAHI, RS30 and RS10 were studied with: Mann-Whitney test for binary variables, Kruskal-Wallis test for categorical variables with more than two categories and Spearman's rank correlation coefficient for continuous variables. To assess whether RS30 and RS10 can be predictive of an OAHI [?] 1.5 and [?] 5, receiver operating characteristic (ROC) curves were plotted. The area under the ROC curves were presented with a 95% confidence interval (CI) obtained with the technique of DeLong *et al*[21]. Finally, Youden's index was calculated to study the "optimal" threshold value of RS30 and RS10 to predict an OAHI [?] 1.5 and [?] 5. It was presented with its specificity, sensitivity, and negative and positive predictive values.

Results

The mean score for tonsillar hypertrophy on the Brodsky scale was 2.9 + 0.7. Ten children had an SHS above the 2.75 threshold (mean 2.72 + 1.22).

PSG analysis showed a median OAHI of 3.4 [0.9; 9.2] per hour. Of the 16 children, 10 were diagnosed with OSAHS (OAHI [?] 1.5). Four patients had mild OSAHS, two had moderate OSAHS and four had severe OSAHS (Table 2). The median ODI for the study population was 2.8 [1.8; 8.3] per hour.

A full analysis of the second sleep cycle (mean time of 72 + 13 minutes) was performed in all children. All the videos were of good quality and centered on the child's head and chest throughout the recording. Analysis of all the videos yielded median values of 4.5 [0.5; 9.5] apneas, 6 [4; 10] whole body movements and 1 [0; 3] awakening. In one of the five children with observed intercostal retractions, the movement was continuous. Seven children were occasional mouth-breathers. The mean RS30 was 4.70 +- 3.04 and the mean RS10 was 3.72 + 2.96 (Table 3).

Children without OSAHS had a median RS30 of 3.00 [1.00; 4.50]. Patients with OSAHS had a median RS30 of 3.01 [1.23; 5.88] if mild, 7.62 [6.09; 9.15] if moderate and 7.25 [4.75; 9.26] if severe. Children without OSAHS had a median RS10 of 1.71 [1.00; 3.00]. Patients with OSAHS had a median RS10 of 1.17 [0.08; 4.29] if mild, 6.37 [4.36; 8.38] if moderate and 7.17 [4.25; 7.92] if severe. Associations between OSAHS severity and RS30 or RS10 values were not significant (p=0.09 for both).

The correlation coefficient between the RS30 and the OAHI was 0.71 (p = 0.002) and -0.63 (p = 0.008) between the RS30 and the BMI. The correlation coefficient between the RS10 and the OAHI was 0.59 (p = 0.02) and -0.58 (p = 0.02) between the RS10 and the BMI. No link between the RS30 or RS10 and patient age was identified.

The RS30 and RS10 area under the ROC curve for an OAHI [?] 5 was 0.88 (95%CI: 0.70 to 1.00).

According to Youden's index, a RS30 threshold of 6.09 was predictive of an OAHI [?] 1.5 with a sensitivity of 60% (95%CI: 26 to 88%) and a specificity of 100% (95%CI: 54 to 100%). The positive and negative predictive values were equal to 100% and 60% respectively. A RS10 threshold of 6.40 was predictive of an OAHI [?] 1.5 with a sensitivity of 50% (95%CI: 19 to 81%) and a specificity of 100% (95%CI: 54 to 100%). The positive and negative predictive values were equal to 100% and 55% respectively.

When considering an OAHI [?] 5, a RS30 threshold of 6.09 had a sensitivity of 83% (95%CI: 36 to 99%) and a specificity of 90% (95%CI: 56 to 99%). The positive and negative predictive values were equal to 83% and 90% respectively. A RS10 threshold of 6.50 had a sensitivity of 67% (95%CI: 22 to 96%) and a specificity of 100% (95%CI: 69 to 100%). The positive and negative predictive values were equal to 100% and 83% respectively.

The correlation coefficient between the RS30 and the ODI was 0.70 (p = 0.003), 0.57 (p = 0.02) between the RS10 and the ODI, and 0.29 (p = 0.28) between the ODI and the OAHI. Greater OAHI values were not significantly associated with greater ODI, SHS, number of apneas, number of movements, number of waking episodes, RS30 and RS10 values (Figure 1).

Fifty percent (5/10) of patients with an SHS [?] 2.75 had an OAHI [?] 5 per hour. The correlation coefficient between the OAHI and the SHS was 0.43 (p = 0.09), 0.62 (p = 0.01) between RS30 and the SHS, and 0.65 (p = 0.007) between RS10 and the SHS.

Discussion

The use of video recordings has already been assessed as a diagnosis tool for sleep conditions such as parasomnias [22]. Its application to pediatric OSDB has only been reported in two publications [18, 23], of which only one was available by the time we started this work [18]. Indeed, as early as 1996, Sivan *et al* had demonstrated the interest of a standardized method of sleep scoring based on home video analysis in children suspected of OSAHS. However, since then, the idea didn't spread wider, probably limited by the availability of efficient home video registration equipment. Nowadays, any family with a smartphone possesses a simple technical solution to record a sleep video at home, putting in a new light the diagnostic value of this tool [23].

Our study was designed based on that of Sivan et al[18], with some modifications we consider as strengths:

-we used PSG videos rather than home recordings, to get rid of the parent-dependent video quality issue

-the videos were synchronized to PSG, which allowed to compare our video risk scores and the PSG results on the same night. It also allowed a complete sleep cycle video analysis based on EEG. Like Thomas *et al* [23] in their recent study, we chose to consider OAHI rather than AHI, which is more in line with current scoring guidelines.

-the video score obtained was brought back to two different time windows: 30 minutes as in Sivan's study, and 10 minutes. Ten minutes seemed a more relevant video duration to use in potential future works as it is more compatible with an everyday practice. Indeed, the underlying purpose of this pilot study was to prepare a wider prospective work using smartphone home sleep video recordings. This explains our choice to slightly modify items 3 and 5 compared with Sivan's original score. Of note, there is only one similar publication based on smartphone recordings in the literature [23], in which the authors used a different scoring system based on 1-minute videos.

We acknowledge some limitations in our study:

-we included fewer patients than Sivan et al, therefore there was only two patients with moderate OSAHS

-with only one investigator, we could not assess a potential interscorer variability. Of note, there was few disagreement between the 3 investigators in Sivan's study [18].

Despite these limitations, our results are consistent with those of Sivan *et al.* The threshold value retained in their study was a 30-minute score greater than or equal to 8, which was predictive of an AHI [?] 1 per hour with a sensitivity of 94% and a specificity of 68% [18]. In our cohort, we were unable to identify a reliable threshold for the diagnosis of mild OSAHS. Indeed, despite a perfect specificity (100%), sensitivity remained low for both RS30 (60% for a threshold of 6,09) and RS 10 (50% for a threshold of 6,40) when considering an OAHI[?] 1.5. Our results were more accurate when considering an OAHI[?] 5. Indeed, a RS30 threshold of 6.09 was predictive of a moderate to severe OSAHS with a sensitivity of 83% and a specificity of 90%. Similarly, a RS10 threshold of 6.50 was predictive of a moderate to severe OSAHS with a lower sensitivity (67%), but a higher specificity (100%).

In comparison with the SHS questionnaire, the RS30 and the RS10 had a similar sensitivity for the diagnosis of moderate to severe OSAHS, but a higher specificity. Indeed, a threshold of SHS [?] 2.75 had a sensitivity of 88% and a specificity of 63% to predict an AHI [?] 5 per hour in Spruyt *et al* study [7]. Of note, we found a lower correlation coefficient (r = 0.43) between the OAHI and the SHS, compared to RS30 (r = 0.71) and RS10 (r = 0.59). Just like in our study, Spruyt *et al* had difficulty in establishing a predictive threshold for mild OSAHS.

It is important to emphasize that comparison of our results with both those of Sivan *et al* [18] and Spruyt *et al* [7] must be taken with caution, as we didn't follow the same rules for scoring respiratory events. Indeed, their studies were carried out before the edition of the 2012 and 2015 rules for scoring of the AASM [12, 13].

Both SHS and our risk scores focus on the noise (snoring) and movements components of OSAHS. An imprecise observer-dependent evaluation of these two parameters may contribute to lower the accuracy of these scores, raising the question of the interest of an automated analysis. Two recent studies in adults using an automatic analysis system of the patients' thoracic movements to determine the severity of OSAHS showed interesting results [24, 25].

We chose to study the second sleep cycle of children for several reasons. We avoided the first sleep cycle as it may include less respiratory events due to predominance of slow wave sleep, thus distorting analysis. Besides, there is an increase in the proportion of time spent in REM sleep with each successive sleep cycle [26]. As the number of central and obstructive apneas is higher during REM sleep, the interpretation of the last sleep cycles could lead to an overestimation of the number of respiratory events. Finally, as this pilot study was a prelude to a wider prospective work using smartphone home sleep video recordings, it seemed relevant to analyze a rather early in the nighttime window, when children are asleep but parents still available to capture the video.

Conclusion

This study demonstrates the reliability of a clinical score calculated from 30 minute-sleep video recordings for the diagnosis of moderate to severe OSAHS, consolidating Sivan *et al* previous reports. In addition to sleep questionnaire such as the SHS, it could allow a better evaluation of type 1 OSAHS, thus restricting the need for sleep recording.

We were also able to assess this clinical score on a shorter timeframe (RS10), more compatible with an everyday practice. Consequently, we have set up a prospective multicenter study to assess this RS10 applied to home sleep video recordings in children with suspected OSDB and AT hypertrophy: "SMARTSAS: Screening of Obstructive Sleep Apnea by Smartphone Homemade Video in Childhood Snoring Population (ClinicalTrials.gov identifier NCT03743558).

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Legends of the figures

Figure 1: Scattergram of 30-minute and 10-minute risk scores according to the severity of obstructive sleep apnea syndrome over the second sleep cycle. The lines represent the medians. OAHI: Obstructive Apnea Hypopnea Index; OSAHS: Obstructive Sleep Apnea Hypopnea Syndrome; RS10: 10-minute Risk Score; RS30: 30-minute Risk-Score. OSAHS was defined as followed: absence if OAHI < 1.5, mild if OAHI between 1.5 (included) and 5 (not included), moderate if OAHI between 5 (included) and 10 (not included), and severe if OAHI [?] 10.

Table 1: Point scoring method used for video analysis for calculating the 30-minute risk score (RS30). RS30 was calculated as the sum of the seven questions.

1 Inspiratory noise	0 None		
	1 Low		
	2 Loud		
2 Type of inspiratory noise	0 Episodic		
	1 Continuous		
3 Movement during sleep	0 No movement		
	1 Little movement $([?] 1)$		
	2 Numerous movements $([?] 2)$ of the whole body		
4 Number of waking episodes	1 Point for each episode		
5 Number of apnea	0 None		
	1 One or two		
	2 Numerous $([?] 2)$		
6 Chest retraction	0 None		
	1 Intermittent (periodic)		
	2 All the time		
7 Mouth breathing	0 None		
	1 Intermittent (periodic)		
	2 All the time		

Table 2: Characteristics of the sample according to the severity of obstructive sleep apnea syndrome. Data are expressed as mean \pm standard deviation or median [25th; 75thpercentiles]. BMI: Body Mass Index; OAHI: Obstructive Apnea Hypopnea Index; ODI: Oxygen Desaturation Index; OSAHS: Obstructive Sleep Apnea Hypopnea Syndrome; SHS: Severity Hierarchy Score. OSAHS was defined as followed: absence if OAHI < 1.5, mild if OAHI between 1.5 (included) and 5 (not included), moderate if OAHI between 5 (included) and 10 (not included), and severe if OAHI [?]10. * One missing data (n = 15).

	All				
children					
(n = 16)	Absence of				
OSAHS					
(n = 6)	Mild				
OSAHS					
(n = 4)	Moderate				
OSAHS					
(n = 2)	Severe				
OSAHS					
(n = 4)					
${\bf Age} \ ({\bf years})$	4.8 ± 1.8	5.8 ± 2.3	4.3 ± 1.3	5.0 ± 0.0	3.8 ± 1.5
Weight (kg)	19.5 ± 5.7	22.2 ± 5.7	18.3 ± 5.4	22.0 ± 9.3	15.5 ± 2.8
${ m Height} \ ({ m cm})$	109.2 ± 11.2	114.7 ± 9.9	102.3 ± 6.6	117.5 ± 17.7	103.8 ± 10.3
BMI (kg/m^2)	16.1 ± 2.4	16.7 ± 1.9	17.3 ± 3.8	15.4 ± 2.0	14.3 ± 0.4
Brodsky*	2.9 ± 0.7	3.2 ± 0.8	3.0 ± 0.8	2.5 ± 0.7	2.8 ± 0.5
SHS	2.72 ± 1.22	2.05 ± 1.28	3.12 ± 0.99	3.47 ± 0.40	2.96 ± 1.48
OAHI (/hour)	$3.4 \ [0.9; \ 9.2]$	$0.5 \ [0.4; \ 1.1]$	3.4 [2.7; 4.1]	L / J	14.9 [11.6; 17.8]
ODI (/hour)	$2.8 \ [1.7; \ 7.7]$	$2.4 \ [1.3; \ 3.0]$	5.6 [2.3; 9.5]	$12.3 \ [2.6; \ 22.0]$	$4.4 \ [0.9; \ 9.5]$

Table 3: Data of nocturnal respiratory events over the second sleep cycle. OAHI: Apnea Hypopnea Index; ODI: Oxygen Desaturation Index; RS10: 10-minute risk score; RS30: 30-minute risk score; SHS: Severity Hierarchy Score.

Patients	#1	#2	#3	#4	#5	#6
SHS	3.81	1.75	3.63	3.75	3.56	3.63
OAHI (/hour)	4.30	2.55	11.22	7.13	1.11	17.94
ODI (/hour)	10.70	2.80	7.00	22.00	2.80	12.00
Duration of the second cycle (minutes)	75	65	82	78	58	95
Number of apneas during the second cycle	4	1	6	21	5	114
Number of movements of the whole body during the second cycle	5	3	6	4	11	11
RS30: Inspiratory noise	1	0	2	2	1	1
RS30: Type of inspiratory noise	1	0	1	1	0	1
RS30: Movement during sleep	1	1	1	1	1	2
RS30: Waking episodes	1.20	0.46	0.00	1.15	0.50	2.52
RS30: Apneas	1	0	1	2	1	2
RS30: Chest retractions	1	0	2	1	0	1
RS30: Mouth breathing	1	0	1	1	1	1
30-minute risk score (RS30)	7.20	1.46	8.00	9.15	4.50	10.52
RS10: Inspiratory noise	1	0	2	2	1	1
RS10: Type of inspiratory noise	1	0	1	1	0	1
RS10: Movement during sleep	1	0	1	1	2	1

Patients	#1	#2	#3	#4	#5	#6
RS10: Waking episodes	0.40	0.15	0.00	0.38	0.33	0.84
RS10: Apneas	1	0	1	2	1	2
RS10: Chest retractions	1	0	2	1	0	1
RS10: Mouth breathing	1	0	1	1	1	1
10-minute risk score (RS10)	6.40	0.15	8.00	8.38	5.33	7.84

