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Evaluation of Primary Screening Tools in Predicting Risk of Obstructive Sleep Apnoea in Children

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ABSTRACT

AIM: To evaluate three primary screening tools in predicting risk of Obstructive Sleep Apnea in children.

METHODS: 60 subjects aged 9-12 years were examined on the basis of history and clinical examination. These subjects were then screened with three screening tools: an eight-item IF-SLEEPY questionnaire, radiological evaluation with lateral cephalogram and overnight at-home pulse oximetry study. Conclusive scores from all three aids were then combined to devise a protocol. Predictive values of Questionnaire and Lateral cephalogram score were derived using pulse oximetry score as definitive.

RESULTS: A significant correlation was found between the scores of all three diagnostic aids. The Positive predictive value and negative predictive value of Questionnaire score was found to be 76.2% and 87.2% respectively and the positive predictive value and negative predictive value of Lateral cephalogram score was found to be 71.4% and 84.6% respectively.

CONCLUSION: Questionnaire and pulse oximetry score were found to have maximum correlation and were better predictors of risk of OSA. 12 out of 60 subjects (20%) had conclusive finding from all the three diagnostic aids. As PSG is not easily accessible outside of major tertiary centres, combined conclusive scores obtained from questionnaire, lateral cephalogram and pulse oximetry can be used to diagnose children at risk of OSA.

Key Words: Apnea, Pulse, oximetry, IF-SLEEPY.

INTRODUCTION

Obstructive sleep apnea syndrome is characterized by repetitive episodes of upper airway obstruction that occur during sleep, usually associated with a reduction in blood oxygen saturation.[1]

Snoring and difficulty in breathing during sleep are the most common complaints of parents of children with Obstructive Sleep Apnea (OSA), with reports of such symptoms in more than 96% of cases.[2,3] Other common complaints reported by parents of children with OSA are mouth breathing, enuresis, frequent upper respiratory tract infections, recurrent ear infections, as well as hearing and speech problems. Excessive daytime somnolence in children has been shown tocorrelate with severity of OSA and with increased body mass index.[4] Whilst these symptoms may be associated with OSA, they are neither sensitive nor specific for diagnosis.[5,6] The presence of adeno-tonsillar hypertrophy on examination also does not correlate to the diagnosis of OSA.[7,8]

Several questionnaires have been developed for paediatric OSA screening. However, it is still a challenge to define a highly sensitive and specific questionnaire that is easy for the patients/parents to complete, and easy to score by clinicians. It has been found that diagnostic accuracy of questionnaire is good enough to warrant its use as a screening method for paediatric Sleep Disordered Breathing (SDB), but not sufficient to be considered a true diagnostic tool (that is, a replacement for full polysomnography) for paediatric SDB.[9]

Many cephalometric studies have shown that children with adeno-tonsillar hypertrophy and obstructive sleep apnea have narrow posterior airway space, retruded mandible, more vertically oriented occlusal and mandibular planes and longer lower face. Many of these relationships are also observed among adults with obstructive sleep apnea.[10]

Home-style nocturnal pulse oximetry provides oxygen desaturation parameters and pulse rate which have been used for assessing oxygen flow during sleep.[11] This method has been validated to diagnose moderate to severe OSA.[12]

The confirmation of presence and severity of paediatric OSA is still obtained with a Polysomnography (PSG). PSG data include the respiratory disturbance index (RDI, the sum of apneas and hypopneas per hour of sleep); oxygen desaturation during sleep, carbon dioxide retention, number of arousals, and the overall sleep architecture.[5,7]

PSG has logistical limitations insofar as there are only a limited number of paediatric sleep laboratories and performing paediatric PSG entails significant cost.

The aim of the study was to evaluate the three primary screening tools in predicting the riskof Obstructive Sleep Apnea in children and to identify children at risk who need further evaluation.

MATERIALS AND METHODS:

The study population included subjects aged between 9 and 12 years, who were referred to the Department of Orthodontics for a regular orthodontic check-up. **Ethical clearance for the study was obtained from the Institution (Institutional Review Board).** Consent/assent was obtained from parents/guardians accompanying the children.

Subjects with an apparent retrognathic mandible; history of mouth breathing habit; history of recurrent naso-respiratory infections; history of snoring; features of adenoid facies and with vertical growth tendency were included while subjects who have undergone previous nasal and oropharyngeal surgery; with diagnosed upper aerodigestive malignancies and syndromes or with neuro-muscular abnormalities were excluded from this study.

60 subjects aged 9-12 years were identified on the basis of inclusion and exclusion criteria.

These subjects were then evaluated using three different diagnostic means-

1) The Eight-Item Questionnaire (IF-SLEEPY)[13]
2) Radiological evaluation
3) Overnight athome Pulse-oximetry

All questions were answered using a yes/no response. Three positive response or more were suggestive of risk of OSA and were scored as 1. Positive response less than three or negative response was scored as 0.[13]

TABLE 1. - IF-SLEEPY QUESTIONNAIRE

	QUESTIONS	YES	NO
I	Is your child often **I**rritated or angry during the day?		
F	Does your child often **F**idget and/or is hyperactive?		
S	Does your child usually **S**nore?		
L	Does your child sometimes have **L**aboured breathing at night?		
E	**E**ver noticed a stop in your child's breathing at night?		
E	Does your child have **E**nlarged tonsils and/or adenoids?		
P	Does your child have **P**roblems with concentration?		
Y	Does your child often **Y**awn or is often tired/sleepy during the day?		

After the filling up of questionnaire by the parents, lateral cephalograms were evaluated. The lateral cephalograms obtained as a part of routine investigation for orthodontic diagnosis were used in this study. All cephalometric radiographs were obtained with Sirona Galileos imaging unit. The radiographs were manually traced and the following angular and linear measurements were measured from the lateral cephalogram(FIGURE 1)–

1. **Position of mandible with respect to cranial base (SNB)**: Measured as an angle formed by the sella-nasion line and line N-point B. The normal value is 80˚. Value <78˚ was considered suggestive of OSA.[14]
2. **Width of posterior airway space (PAS):** Measured as linear distance from the point of intersection of the posterior border of the tongue and the inferior border of the mandible to the closest point on the posterior wall of the pharynx. The average value is 11-14 mm. Obtained value < 11 was considered suggestive of OSA.[14]

3. **Soft palate length (PNS-P)**: Measured from posterior nasal spine (PNS) to the tip of the soft palate (P). The average value is 30-33 mm. Obtained value >33 mm was considered suggestive of OSA.[14]
4. **Mandibular plane – Hyoid bone distance (MP-H)**: Measured as perpendicular linear distance between H, the most anterosuperior point of the hyoid bone, and the mandibular plane taken as lower border of mandible. The average value is 12-18 mm. A value > 18 mm was considered suggestive of OSA.[14]
5. **Inclination of mandibular plane (MPA)**: Measured as angle formed by the sella-nasion line and mandibular plane obtained by drawing tangent to the lower border of mandible. The average value is 32˚. A value > 32˚was considered suggestive of OSA.[14]
6. **Adenoidal – Nasopharyngeal Ratio (AN)**: The adenoidal measurement A represents the perpendicular distance from the point of maximal convexity along the inferior margin of the adenoid shadow, to a line drawn along the straight part of the anterior margin of the basioccipital. The nasopharyngeal space N is measured as the perpendicular distance between the posterior-superior edge of the hard palate, and the anteroinferior edge of the spheno-basioccipital synchondrosis. AN ratio was obtained by dividing the measurement for A by the value for N. An AN ratio of ≥0.80 was considered suggestive of OSA.[15]

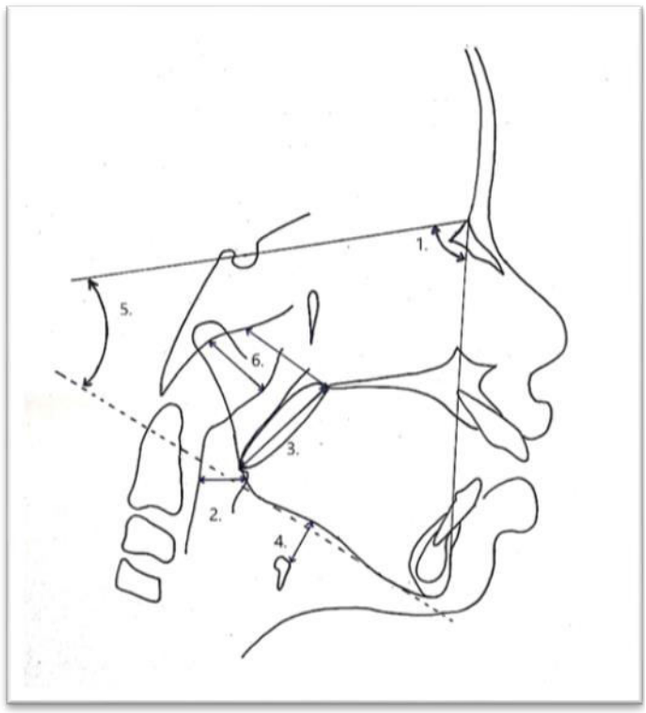

FIGURE 1. VARIBLES MEASURED IN THE STUDY - *1*.SNB; *2*. Posterior airway space (PAS); *3*. Length of soft palate (PNS-P); *4*. Mandibular plane to hyoid (MP-H); *5*. Mandibular plane angle (MPA);
6. Adenoidal-Nasopharyngeal ratio (AN)

Out of the six measurements, when three or more measurements were suggestive of OSA, a score of 1 was assigned. A score of 0 was scored when less than three measurements were suggestive or no measurements were suggestive of OSA.

Following radiological evaluation, all subjects underwent over-night at home pulse-oximetry study where a portable pulse-oximeter with 8-hour recording time was used. The guardian as well as the subject were explained the working of the pulse-oximeter. The pulse-oximeter used was SCURE PULSE-OXIMETER FTP 401 (Silverline Meditech Pvt. Ltd., Gujarat, India)which had 8-hour recording and software to interface features. This overnight recorded pulse oximeter readings were analysed through a computer software (FIGURE 2).

A desaturation is defined as a decrease in peripheral capillary oxygen saturation (SpO2) of 4% or more. A cluster of desaturations is defined as 5 or more desaturations occurring in a 10- to 30-minute period.Each oximetry reading was classified as positive, negative, or inconclusive using the following definitions and criteria:

1) A positive oximetry trend graph had 3 or more desaturation clusters and at least 3 desaturations to 90%;

2) A negative oximetry trend graph had no desaturation clusters and no desaturations to 90%; and

3) An inconclusive oximetry trend graph was one that did not meet the criteria for positive or negative.[11]

Based on the oximetry trend graph, a positive oximetry reading was taken as suggestive for risk of OSA and was scored as 1. Negative or inconclusive oximetry trend was scored as 0.

Data was tabulated to co-relate the scores obtained from all 3 diagnostic tests with one another.

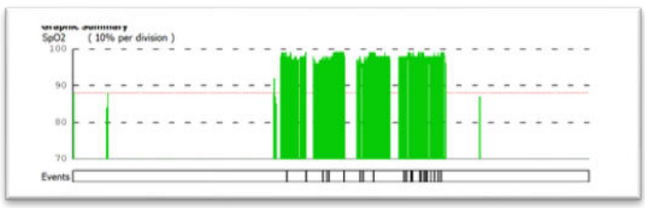

Figure 2. Graphic Interpretation Of Overnight Pulse Oximetry Study.

STATISTICAL ANALYSIS

Data comprising of scores of Questionnaire, Radiological evaluation and Pulse-oximeter study obtained from 60 subjects was tabulated in Microsoft excel. This data was then analysed using statistical software (IBM SPSS V23.0, IBM Corp., Armonk, NY) and co-related with Spearman's correlation test. Chi-square test was then used to find association between the

means of these scores. A probability(P-value) of <0.05 was considered statistically significant.

RESULTS

Conclusive and non-conclusive scores for the risk of OSA from all the three screening tools in 60 subjects were obtained.

For the Questionnaire response (Q Score), 21 subjects had conclusive score and 39 subjects had a non-conclusive score. For the Radiological evaluation of lateral cephalogram (LC Score), 24 subjects had a conclusive score and 36 subjects had non-conclusive score.

For the Pulse-oximetry study (Pulse-ox Score), 21 subjects had conclusive score and 39 subjects had a non-conclusive score.

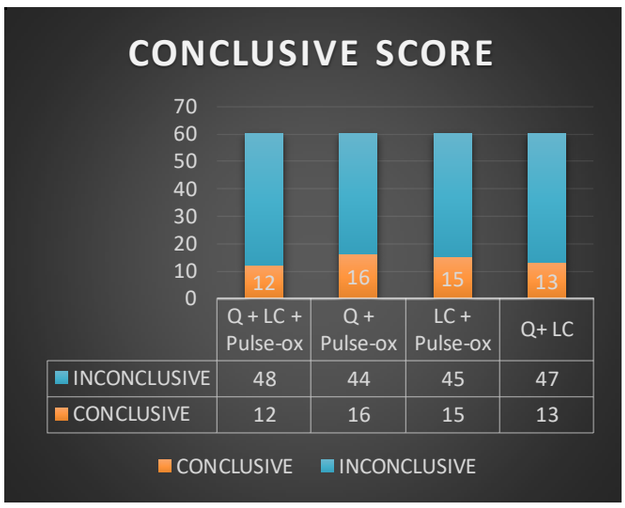

Figure 3. Combined Conclusive And Non-Conclusive Scores For The Risk Of Osa Obtained From The Three Screening Tools

12 out of 60 subjects (20%) had conclusive finding from all the three diagnostic aids. 16 out of 60 subjects (26.6 %) had conclusive finding from Questionnaire and pulse-oximetry combined. 15 out of 60 subjects (25%) had conclusive finding from Lateral cephalogram and pulse-oximetry combined. 13 out of 60 subjects (21.6%) had conclusive finding from Questionnaire and lateral cephalogram combined (Figure 3).

Table 2. Correlation Of Questionnaire, Lateral Cephalogram And Pulse-Oximetry Scores To One-Another Using Spearman's Correlation Test

	Q SCORE	LC SCORE	PULSE-OX SCORE
Q Score Correlation Coefficient		0.341**	0.634**
LC Score Correlation Coefficient	0.341**		0.560**
PULSE-OX Score Correlation Coefficient	0.634**	0.560**	

**** Correlation is significant**

Spearman's correlation test was then used to correlate the scores obtained from these three screening tools. It was found that there was a significant correlation between all the three tools. The correlation between Pulse-oximetry score with Questionnaire score was found to be 0.634 or 63.4%, that between Pulse-oximetry score and Lateral cephalogram score was 0.560 or 56% and between Lateral cephalogram-score and Questionnaire-score was 0.341 or 34.1% (TABLE 2).

Maximum correlation was found between Q score and Pulse-ox score (63.4%). A moderate correlation was found between LC score and pulse-ox score (56.0%) and a minimum correlation was found between Q score and LC score (34.1%).

Pulse-oximetry score was found to have more accuracy in predicting OSA. So, the predictive value of Questionnaire score and Lateral cephalogram score was derived from the scores of Pulse-oximetry. The association between Q score and pulse-ox score was determined using chi-square test.

Positive predictive value was obtained by dividing the

TABLE 3. PREDICTIVE VALUE OF Q-SCORE WITH PULSE-OX SCORE AS STANDARD.

		Total	PULSE-OX SCORE	
			Conclusive	Non-Conclusive
Q SCORE	Conclusive	21	16	5
	21/60	35%	76.2%	12.8%
	Non-conclusive	39	5	34
	39/60	65%	23.8%	87.2%

conclusive score of Questionnaire by total conclusive score of Pulse-oximetry. The Positive predictive value of Questionnaire score was found to be 76.2%.

Negative predictive value was obtained by dividing the non-conclusive score of Questionnaire by total non-conclusive score of Pulse-oximetry. The Negative predictive value of Questionnaire score was found to be 87.2% (TABLE 3, FIGURE 4).

Positive predictive value was obtained by dividing the conclusive score of Lateral cephalogram score by total conclusive score of Pulse-oximetry. The Positive predictive value of Lateral cephalogram score was found to be 71.4%.

Negative predictive value was obtained by dividing the non-conclusive score of Lateral cephalogram score by total non-conclusive score of Pulse-oximetry. The Negative predictive value of lateral cephalogram score was found to be 84.6% (TABLE 4, FIGURE 4).

TABLE 4. PREDICTIVE VALUE OF LC-SCORE WITH PULSE-OX SCORE AS STANDARD.

		Total	PULSE-OX SCORE	
			Conclusive	**Non-Conclusive**
LC SCORE	**Conclusive**	**21**	**15**	**6**
	21/60	35%	71.4%	15.4%
	Non-conclusive	**39**	**6**	**33**
	39/60	65.0%	28.6%	84.6%

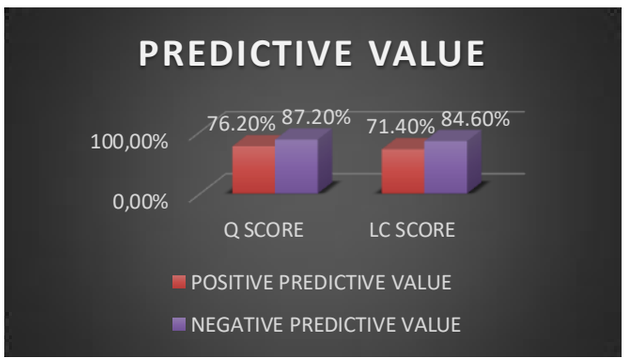

Figure 4. Predictive Value Of Questionnaire Score And Lateral Cephalogram Score When Compared With Pulse-Oximetry Score

DISCUSSION

There is an increasing interest in the role of the orthodontist for screening OSA and as a practitioner who may be valuable in the multidisciplinary management of OSA in both children and adults. As experts in the science of facial growth and development, combined with our knowledge of oral devices, orthodontists are well suited to collaborate with physicians and other allied health providers in the treatment of OSA. Although OSA can be definitively diagnosed only by a physician, the orthodontist may be called on to screen for OSA, contribute to the identification of underlying dentofacial components, and assist the physician in managing the disease.

In this study, a significant correlation was found between the scores of all three diagnostic aids.Maximum correlation was found between the conclusive scores obtained from Pulse-oximetry study and Questionnaire response. A moderate correlation was found between the conclusive scores obtained from Pulse-oximetry study and lateral cephalogram evaluation and a minimum correlation was found between questionnaire response and lateral cephalogram evaluation.

This is in accordance with Davies et al who studied correlation between clinical symptoms, lateral cephalogram measurements and pulse-oximetry in adults suspected of having OSA and found that clinical symptoms such as neck size, obesity and soft-palate length, hyoid bone distance and retroglossal airway space were all significantly correlated with >4% SpO2 dip rate. They also suggested that the relationships of soft palate length and mandibular plane to hyoid distance with the severity of OSA, both appear to be secondary to variation in neck circumference. [16]

Chang et al performed a retrospective study to develop a screening process for OSA using a combination of symptoms, questionnaire and the oxygen desaturation index as determined by nocturnal pulse oximetry. They demonstrated that the oxygen desaturation index and the occurrence of observable apnea or mouth breathing during sleep or restless sleep were significant predictors of OSA in PSG tests.[17]

Various studies have determined sensitivity and specificity of overnight at-home pulse oximetry in comparison with PSG. Brouillette RT et al ina cross-sectional study have concluded that when a child is suspected of having OSA, a positive nocturnal

oximetry trend graph has at least a 97% positive predictive value. They further stated that given the high positive predictive value (close to 100%) of a nocturnal oximetry recording demonstrating clusters of desaturations events in a child with suspected OSA, performance of polysomnography does not appear to be required to refer the patient for adenotonsillectomy.[11]

Kirk et al found the sensitivity and specificity of the portable monitor "SnoreSat" for the identification of moderate sleep apnea (polysomnography AHI > 5/h) were 67% and 60%, respectively.[18]

Nixon et al has found an abnormal oximetry had 82-86% sensitivity and 10-13% positive predictive value for detecting children who required major interventions for respiratory compromise postoperatively.[19]

Kladitis et al in their systematic review stated that children without clusters of desaturation events have low risk of major respiratory complications following adenotonsillectomy. They concluded that nocturnal oximetry is a valuable tool that can facilitate treatment decisions when polysomnography is not available.[20]

In this study, we found a significant correlation between the scores of all three diagnostic aids. The Positive predictive value and negative predictive value of Questionnaire score was found to be 76.2% and 87.2% respectively and the positive predictive value and negative predictive value of Lateral cephalogram score was found to be 71.4% and 84.6% respectively. The predictive value for Questionnaire score and lateral cephalogram score were found to be significant individually.

Kadmon et al validated the questionnaire in a cohort of 150 children and found that the eight-item (parent-response) displayed a sensitivity of 78% and a specificity of 40% for the diagnosis of OSA.[13]This is in accordance with a retrospective cross-sectional study done by Abumuamar AM et al where they reviewed all the paediatric questionnaires and found that the eight-item scale (IF-SLEEPY) displayed 82% sensitivity and 28% specificity.[21]

The positive predictive value found in our study was in correlation with these previous studies, however the negative predictive value was found to be higher. The results can be explained on the premises that both the studies mentioned have validated the questionnaire with gold-standard polysomnography whereas in our study it was done with pulse-oximetry score.

A retrospective review of 50 children from a Hong Kong sleep clinic combined a lateral neck roentgenogram with history and physical findings and showed good sensitivity (>90%) in detecting OSA, but the specificity was only about 50%. Statistically significant association was found between scores of lateral cephalogram and pulse-ox and questionnaire.[22]The Positive predictive value of lateral cephalogram was found to be in good correlation with this study, however the negative predictive value was found to be higher.

Out of the 60 subjects screened, 12 out of 60 subjects (20%) had conclusive finding from all the three diagnostic aids. This conclusive score helps to identify children at definite risk of OSA who need further investigation. These children can then be subjected to polysomnography which is currently considered gold standard for a definite diagnosis of OSA in children.

15 out of 60 subjects (25%) had conclusive finding from two diagnostic aids. Children with one or two conclusive score should be educated about the disorder and should be evaluated periodically for any further complication. The protocol for screening evaluated in this study can narrow down the number of children who actually need to be further investigated with a polysomnography, thereby reducing the cost and inconvenience to a large number of children.

Lateral cephalogram is part of the routine investigation of an orthodontic practice. So, an eight-item Questionnaire along with pulse oximetry can be easily be incorporated in an orthodontic office for further evaluation of children found to be at risk of OSA on the basis of history and clinical examination. As PSG is not easily accessible outside of major tertiary centres, combined conclusive scores obtained from questionnaire, lateral cephalogram and pulse oximetry can be used to diagnose children at risk of OSA. Further studies are needed to determine if these results are maintained in larger cohorts.

The limitation of our study was that the validation of the Questionnaire and lateral cephalogram measurements was done with pulse-oximetry readings as definite and conclusive. However, pulse-oximetry is not accepted as confirmative diagnostic tool for diagnosing paediatric OSA.

CONCLUSION

Following conclusions can be drawn from the study –

1. 20% of the subjects had conclusive finding from all the three diagnostic aids. This conclusive score helps to identify children at high risk of OSA who need further investigation. These children can then be subjected to polysomnography which is currently considered gold standard for a definite diagnosis of OSA in children.
2. A significant correlation was found between the scores of all three diagnostic aids. Questionnaire and pulse oximetry scores were found to have maximum correlation in our study and were found to be better predictors of OSA.

3. The predictive value for Questionnaire score and lateral cephalogram score in comparison with pulse oximetry score were found to be significant individually. The Positive predictive value and negative predictive value of Questionnaire score was found to be 76.2% and 87.2% respectively and the positive predictive value and negative predictive value of Lateral cephalogram score was found to be 71.4% and 84.6% respectively.
4. Subjects with significant correlation with 2 of the 3 diagnostic aids, may be observed for further deterioration of symptoms or subjected to periodic evaluation.

LIST OF ABBREVIATIONS

1. OSA – Obstructive Sleep Apnea
2. SDB – Sleep Disordered Breathing
3. PSG – Polysomnography
4. RDI – Respiratory Disturbance Index
5. SNB – Sella-Nasion-pt. B
6. PAS – Posterior Airway Space
7. PNS – Posterior Nasal Spine
8. MPA – Mandibular Plane Angle
9. SpO2 – Peripheral Capillary Oxygen Saturation

DECLARATIONS

Ethical approval and Consent to participate: Obtained

Consent for publication: Obtained

Availability of data and material: Obtained with consent taken beforehand

Competing interests: None

Acknowledgment: None.

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Conflicts of interest: There are no conflicts of interest.

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