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The role of sleep laboratory polygraphy in the evaluation of obstructive sleep apnea syndrome in Robin infants

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Abbreviations: American Academy of Sleep Medicine (AASM), arousal index (AI), central apnea index (CAI), electroencephalogram (EEG), gestational age (GA), mixed apnea index (MAI), mixed obstructive apnea hypopnea index (MOAHI), non-invasive ventilation (NIV), naso-pharyngeal tube (NPT), non-rapid eye movement sleep (NREM), obstructive apnea index (OAI), oxygen desaturation index > 3% (OD3%), oxygen desaturation index > 4% (OD4%), obstructive apnea hypopnea index (OAHI), obstructive hypopnea index (OHI), obstructive sleep apnea (OSA), obstructive sleep apnea syndrome (OSAS), polygraphy (PG), prone positioning (PP), Robin sequence (RS), polysomnography (PSG), transcutaneous

carbon dioxide pressure (P(tcCO(2))), rapid eye movement sleep (REM), sleep period (SP), supine positioning (SUP), time in bed (TIB), total sleep time (TST)

Contributor's Statement Page

Dr Coutier conceptualized and designed the study, designed the data collection instruments, collected data, drafted the initial manuscript, and reviewed and revised the manuscript.

Dr Bierme carried out the initial analyses, drafted the initial manuscript and reviewed and revised the manuscript.

Ms Thieux carried out the initial analyses and revised the manuscript.

Dr Guyon designed the data collection instruments and collected data.

Dr Ioan reviewed and critically revised the manuscript for important intellectual content.

Prof. Franco conceptualized and designed the study, supervised data collection, and critically reviewed the manuscript for important intellectual content.

Prof. Reix conceptualized the study and critically reviewed the manuscript for important intellectual content.

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Abstract

Objective/background Obstructive sleep apnea syndrome (OSAS) management in Robin sequence (RS) infants has not been standardized. Sleep laboratory polysomnography (PSG) is the gold standard for OSAS diagnosis, however, access is restricted. This study aimed to compare the respiratory indexes measured in a sleep laboratory using PSG as well as polygraphy (PG), a possible alternative.

Patients/Methods Retrospective study conducted between 2015 and 2017 in a tertiary hospital. PSG performed in RS infants in the sleep laboratory was analysed by a single reviewer. After sleep data removal, anonymized raw data were analysed to obtain only PG data. Respiratory indexes were compared for (i) PSG and PG and (ii) patients with or without OSAS clinical signs.

Results Among the 20 RS (median [IQR] age: 43 [25–114] days at evaluation), 70% of the patients had OSAS clinical signs but all of them had severe OSAS. The median mixed obstructive apnea hypopnea index was not significantly different between PSG and PG (27/h [18–38] versus 26/h [18–56], $p=0.43$). The median obstructive apnea index was higher with no significant difference between PG and PSG (19/h [15–31] versus 7/h [4–25], $p=0.05$). The median obstructive hypopnea index was significantly lower on PG than on PSG (2/h [0–3] versus 8/h [8–19], $p=0.01$). No difference on PSG or PG was observed for patients with and without clinical signs of OSAS.

Conclusion Although PSG remains the gold standard for OSA evaluation, a PG seems to be a useful alternative to measure OSA in RS infants because of their OSAS severity. This evaluation should be recommended in all RS infants, even in the absence of OSAS clinical signs.

Keywords: polysomnography; polygraphy; respiratory management; infants; obstructive sleep apnea syndrome, Robin sequence

1. Introduction

Robin sequence (RS) is a congenital anomaly defined by retrognathia, glossoptosis and cleft palate ¹. Its incidence is reported to be 1/8500 births in the United Kingdom ², and as little as 1/10,000 births in France ³. RS is complicated by feeding difficulties and a variable degree of upper airway obstruction with or without hypoxemia, which could become life-threatening ³⁻⁷. Airway obstruction may occur during wakefulness and sleep, and obstructive sleep apnea syndrome (OSAS) is very frequent in RS ⁸⁻¹⁰. OSAS is associated with high morbidity including impaired cognitive and behavioural development ^{4,10}, and predisposition to pulmonary hypertension ¹¹. However, OSAS is underestimated by the clinical picture ^{4, 10-13}. Snoring, a cardinal symptom of OSAS, is present in only 50% of infants with severe OSAS ¹¹, and the first symptoms appear only during the 2nd month of life for the majority of RS patients ¹⁴. This highlights the importance of physiological tools to diagnose OSAS during the first months of life. Polysomnography (PSG) in a sleep laboratory is the gold standard for the diagnosis of OSAS and to assess its severity ^{15,16}. However, access to sleep-laboratory-based PSG is limited. Moreover, PSG is more time-consuming than polygraphy (PG) in terms of installation (electroencephalogram [EEG], electrooculogram, electromyogram) and reading (sleep stage scoring). Several PGs can be performed in the time required for a single PSG. Consequently, an overnight recording of respiratory movements, PG combined with pulse oximetry (SpO₂), and transcutaneous carbon dioxide pressure (P(tcCO₂)), whether or not performed in a sleep laboratory, is generally used to diagnose obstructive sleep apnea (OSA) ¹⁵⁻¹⁷. However, there is no international consensus on the use of this physiological tool ¹⁵⁻¹⁷.

Once confirmed in RS, treatment of OSA is selected according to severity, classified as mild (apnea hypopnea index [AHI] > 1–5 episodes/h), moderate (AHI > 5–10 episodes/h), or severe (AHI > 10 episodes/h) according to the American Academy of Sleep Medicine (AASM) ¹⁸. Although there is no consensus, OSAS management in RS patients is based on the combination of non-surgical treatments (i.e., prone positioning [PP], non-invasive ventilation [NIV], or placement of a naso-pharyngeal tube [NPT]) and surgical treatments for cleft palate and glossoptosis, depending on the severity of OSA ^{17, 19-21}, and varies among hospitals and countries ¹⁶. In our centre, in young infants, continuous positive pressure (CPAP) is chosen for severe/moderate OSAS ^{9,21} and PP for mild/moderate OSAS ^{5,17,22,23}. The aim of the present study was to compare the respiratory measures obtained after PSG and PG in a sleep laboratory in RS infants and between patients with and without respiratory signs to determine the utility of proposing the sleep laboratory PG as an alternative to PSG in RS infants. Given that the prone position (PP) is a therapeutic option for these patients and this position could change the sleep structure (more non-rapid eye movement [NREM] sleep, fewer awakenings ^{24,25}) and increase de risk for Sudden Infant Death Syndrome, the impact of sleep position on the respiratory results was also evaluated.

We hypothesised that the respiratory measurements obtained using PG were the same as those obtained with PSG as performed in a sleep laboratory.

2. Material and Methods

2.1. Study design

2.1.1 Patients

All infants aged < 4 months with a diagnosis of RS who were referred from January 2015 to October 2017 to the sleep unit of the Lyon Paediatric University Hospital (Hospices Civils de Lyon, France) for a complete sleep study using PSG were included. Data were analysed retrospectively. PSG in patients with RS is our current practice. RS type was defined according to the classic RS classification: isolated RS, and associated or syndromic RS¹⁶. The main clinical characteristics and respiratory support (i.e. high flow nasal cannula [HFNC], CPAP or NPT) were collected from the patients' medical files. Presence or absence of clinical signs of OSAS (i.e. snoring, noisy breathing, apneas, an apparent life-threatening event, frequent arousals because of respiratory difficulty, desaturation and/or stridor) were noted.

2.1.2 Nocturnal PSG

An overnight PSG was performed in the reference sleep centre. Infants were systematically recorded in the supine position (SUP) and then in the PP for a minimum of two sleep cycles for each condition, with no tilting of the mattress. They were studied with and without respiratory support if this aid was in use. Periods with respiratory support were excluded from the analysis. The nocturnal PSG was performed in the presence of the child's mother or father, using a Morpheus recorder (Micromed, Mogliano Veneto, Italy). Sleep and respiratory events were scored using SleepRT software (OSG, Rumst, Belgium). The PSG recording included frontal, central and occipital leads (FP1, FP2, C3, C4, O1, O2, A1 and A2), video, two electrooculograms, one chin electromyogram, one electrocardiogram, inductance plethysmography chest and abdominal respiratory movements, nasal cannula, oronasal thermistor and saturation values.

PSGs were scored by a single reviewer (L.C., physician). The reviewer was not aware of the position information. Sleep stage was scored according to the standard guidelines¹⁸. The parameters calculated were: the total duration of sleep stages (NREM, REM), time in bed (from lights off to lights on [TIB]), sleep period (from sleep onset to the final awakening [SP]), total sleep time (TST), sleep efficacy (TST/SLP*100), sleep efficiency (TST/TIB*100) and the arousal index.

2.1.3 Respiratory analysis of nocturnal PSG

Respiratory parameters were defined according to the AASM 2012 guidelines¹⁸. The obstructive apnea index (OAI), obstructive hypopnea index (OHI), obstructive apnea hypopnea index (OAHI), central apnea index (CAI), mixed apnea index (MAI), mean SpO₂, time spent with SpO₂ values below 90%, oxygen desaturation index > 3% (OD3%) and oxygen desaturation index > 4% (OD4%) were collected. Indices of events are expressed as the mean number of events per hour. MOAHI was defined as the sum of OAI, OHI and MAI. Respiratory parameters were analysed for each sleep position and according to the TST. P(tcCO₂) values were obtained over the entire PSG recording period using the SenTec system (SenTec Digital Monitor; Therwil, Switzerland). The mean and maximum P(tcCO₂), as well as the proportion of time spent with P(tcCO₂) > 50 mmHg, were calculated. Blood gases were estimated. Hypoventilation was defined as P(tcCO₂) > 50 mmHg for > 25% of TST according to guidelines¹⁸.

2.1.4 Severity of sleep obstructive apnea syndrome

The infants were divided into three groups according to the AASM guidelines¹⁸: mild, moderate or severe OSA. MOAHI was considered normal in healthy infants at under 1 event/h according to studies assessing OSA in very young infants^{26,27}. For each patient, the

OSA severity classification obtained after PG was compared to the classification obtained after PSG in SUP.

2.1.5 Polygraphy analysis

To obtain only PG data, sleep stage information was removed from the records and a second analysis was performed, 1 month later, by the same physician (LC) in a random manner in order to prevent recognising events identified on the first PSG analysis. TST was determined from the visual evaluation of vigilance states reported by the nurses to calculate respiratory event indices ²⁷. These parameters were obtained for PP and SUP. No specific oxygen saturation and P(tcCO₂) analysis was done for the PG study.

2.1.6 Comparison of respiratory indexes recorded on PSG and PG

The respiratory indexes (OAI, OHI, CAI, MAI, MOAHI) were compared after PSG analysis and PG analysis for each infant in an overall analysis, then for the groups with and without clinical signs of OSA and for PP and SUP.

2.2 Regulatory aspects

The study was approved by the hospital's ethics committee and the national data protection agency (Commission nationale de l'informatique et des libertés; number 18-006). According to the most recent French law for conducting retrospective clinical research, informed consent was not required; the parents of the patients included were informed of the study by letter).

2.3 Statistical analysis

Data are presented as median and interquartile range [IQR]. All analyses were performed using R Software (R Core Team, 2009²⁸). Analyses were conducted using the non-parametric paired Wilcoxon rank test. A *p*-value less than 0.05 was considered statistically significant. Respiratory indexes (OAI, OHI, CAI, MAI, MOAHI) were compared for: i) PSG and PG for the entire group of patients; ii) PSG and PG for patients with or without respiratory signs of OSA and iii) PSG and PG for PP and SUP.

3. Results

3.1 Patient characteristics

PSG was performed in 20 infants with RS (eight boys, 12 girls). Fifteen of these infants (75%) presented isolated RS, and five (25%) associated or syndromic RS (associated: $n = 2/5$; syndromic: Franceschetti $n = 2/5$, *del22q11* = $1/5$). The median [IQR] age at the time of PSG was 43 [25–114] days. A total of 14 (70%) infants had clinical signs of OSAS when the recording took place (i.e. snoring, noisy breathing, apneas, an apparent life-threatening event, frequent arousals because of respiratory difficulty, desaturation and/or stridor). There were six (30%) infants with respiratory support (HFNC, $n = 1$; CPAP, $n = 4$; CPAP+NPT, $n = 1$). The mean positive expiratory pressure setting was 10 cm H₂O. All infants had sucking-swallowing disorders, 17 (85%) had gastroesophageal reflux treatments and 13 (65%) had been fed through a nasogastric tube in the days or weeks before PSG was performed (Table 1).

3.2 Sleep characteristics

PSG in room air was performed in 20 patients (100%). Median TST, sleep efficacy, sleep efficiency, arousal index, respiratory arousal index, proportion of sleep stages and respiratory indexes by sleep stages are presented in Table 2.

3.3 Respiratory measurements on PSG and PG

Respiratory measurements were obtained after PSG and PG recordings in room air in 20 infants. The median MOAHI did not differ significantly between PG and PSG (median [IQR]: 27/h [18–38] versus 26/h [18–56], $p=0.43$). The median OAI was higher on PG than on PSG, with no significant difference (19/h [15–31] versus 7/h [4–25], $p=0.05$). The median OHI was significantly lower on PG than on PSG (2/h [0–3] versus 8/h [8–19], $p=0.01$). The median MAI was significantly lower on PG compared to PSG (2/h [0–6] versus 3/h [2–5], $p=0.03$).

The median [IQR] pulse oxygen saturation was 97% [94–97], P(tcCO₂) was 52 mmHg [47–55] and the maximum P(tcCO₂) was 56 mmHg [51–60] (Table 3, Figure 1).

3.4 Respiratory measurements in infants with and without clinical signs of OSAS

A total of 14 (70%) infants had respiratory signs of OSA at the time of the recording and six (30%) did not. No difference was observed between the group with or without respiratory signs of OSA (i.e. median gestational age at birth, median age at PSG, median weight at PSG, type of RS). Respiratory measurements were compared between patients with and without respiratory signs of OSA based on PSG and PG. On PSG and PG, there was no significant difference in the median values of respiratory measurements between infants with and those without respiratory signs of OSA.

3.5 Respiratory measurements and sleep position

The same analysis was made in the supine and in prone positions. The pattern of results was identical to the overall analysis reported in Table 3.

3.6 Severity classification of OSAS

After PSG, all infants were classified as having severe OSA. After PG, the OSA classification was identical for 19 (95%) infants, who were still classified as having severe OSA, and was different for one (5%) infant, who was classified as having moderate OSA.

4. Discussion

The present study found that in young RS infants the PSG and PG recordings performed in the sleep laboratory showed a significant difference on the OAI and the OHI but had no impact on the MOAHI.

A trend towards overestimation of OAI in PG compared to PSG was observed, irrespective of sleep position. This contrasts with the study reported by Tan et al. in a group of children aged from 2 to 16 years without RS; the authors did not find a significant difference on the OAI as measured by the two methods (4.9/h on PSG versus 5/h on PG)²⁹. This could be explained by the age of the patients: in the current study, only young infants aged less than 4 months were included. Clinical sleep evaluation without EEG data is easier in children than in young infants. In the latter, normal behaviours such as swallowing or crying could be interpreted as apnea during wakefulness on PG and lead to an overestimation of apnea on PG. However, the high MOAHI found by both PG and PSG in RS infants led to the classification of severe OSA and therefore any overestimation of OAI by PG had no impact on OSAS management in our cohort.

A significant underestimation of the OHI on PG was observed in comparison to PSG, irrespective of sleep position. This was also reported by Tan et al. among children with severe OSA²⁹. This is known to be due to the definition of OHI: flow reduction $\geq 30\%$ during two breathing cycles, with $\geq 3\%$ oxygen desaturation or an arousal¹⁸. As EEG is not recorded on PG, arousals cannot be evaluated and hypopneas causing arousals without desaturation cannot be identified^{29,30}. Underestimation of the OHI and the arousals related to respiratory events could be of importance: Montgomery et al.³¹ reported a relationship between the respiratory arousal index and mental development at 6 months.

Although differences between PSG and PG were observed for the OAI and OHI, there was no significant difference in the MOAHI between PG and PSG. The overestimation of the OAI was compensated by the underestimation of the OHI (or vice-versa). Importantly, the MOAHI

for all patients was substantially above the threshold of 10/h (27/h on PSG and 26/h on PG), corresponding to severe OSA¹⁸. No data on the median OAHl in RS infants are available in the literature.

In the present study, there was no relationship between the clinical signs of OSAS and the MOAHl. In other words, the absence of clinical signs of OSAS was not predictive of the MOAHl or other respiratory outcomes on either PSG or PG. This result must be considered with caution because of the small size of the study sample. No studies are available in the literature presenting results on RS OSAS and PSG or PG respiratory outcomes. Only Abel et al. have suggested an approach for RS OSA evaluation and respiratory management. This team successfully used a McGill oximetry score > 2 to identify children with the most severe OSA which required NPT management³². In our cohort, the McGill score was not used because of the lack of sensitivity in infants aged less than 1 year because of the difficulty distinguishing active sleep and wakefulness³³. This lack of a relationship between clinical signs of OSAS in young RS infants and the OAHl must be noted. A PG or PSG recording seems to be indicated systematically for all RS infants younger than 4 months of age regardless of the clinical signs of OSAS.

The limitations of this study include the sample size, which precluded multivariate analysis, yet infants were all systematically assessed in a similar manner in the sleep laboratory before 4 months of age. A selection bias could be considered because some infants were referred to the sleep laboratory by the paediatric or the neonatal intensive care unit, favouring the recruitment of the most severe patients. However, all RS infants of the Auvergne-Rhône-Alpes region in France were referred to the sleep laboratory and were recorded, with or without clinical signs of OSAS. One-third of these infants presented no clinical signs of OSAS, which is concordant with data reported elsewhere^{9,11} and suggests that the sample was representative. No analyses concerning the PG or PSG recording with respiratory support

were carried out: only a few data were collected in this condition. Another limitation is that all PSGs and PGs were reviewed by a single reviewer (LC), so assessing interrater variability was impossible. Moreover, it must be taken into account that the recordings (PSG and PG) were made in a sleep laboratory directly monitored by trained paediatric nurses.

5. Conclusion

An overnight PG recording t seems to be a valuable alternative for OSA evaluation in RS infants younger than 4 months of age because of the severity of OSAS in these infants regardless of clinical signs of OSAS. In future studies, additional tools such as videopolygraphy, heart rate spectral analysis ³⁴ or simplified EEG recording could contribute to increasing the precision of PG. Since PSG is difficult to access and time-consuming, this exam could be reserved for the evaluation of RS infants who have mild or moderate OSA on PG to improve their management.

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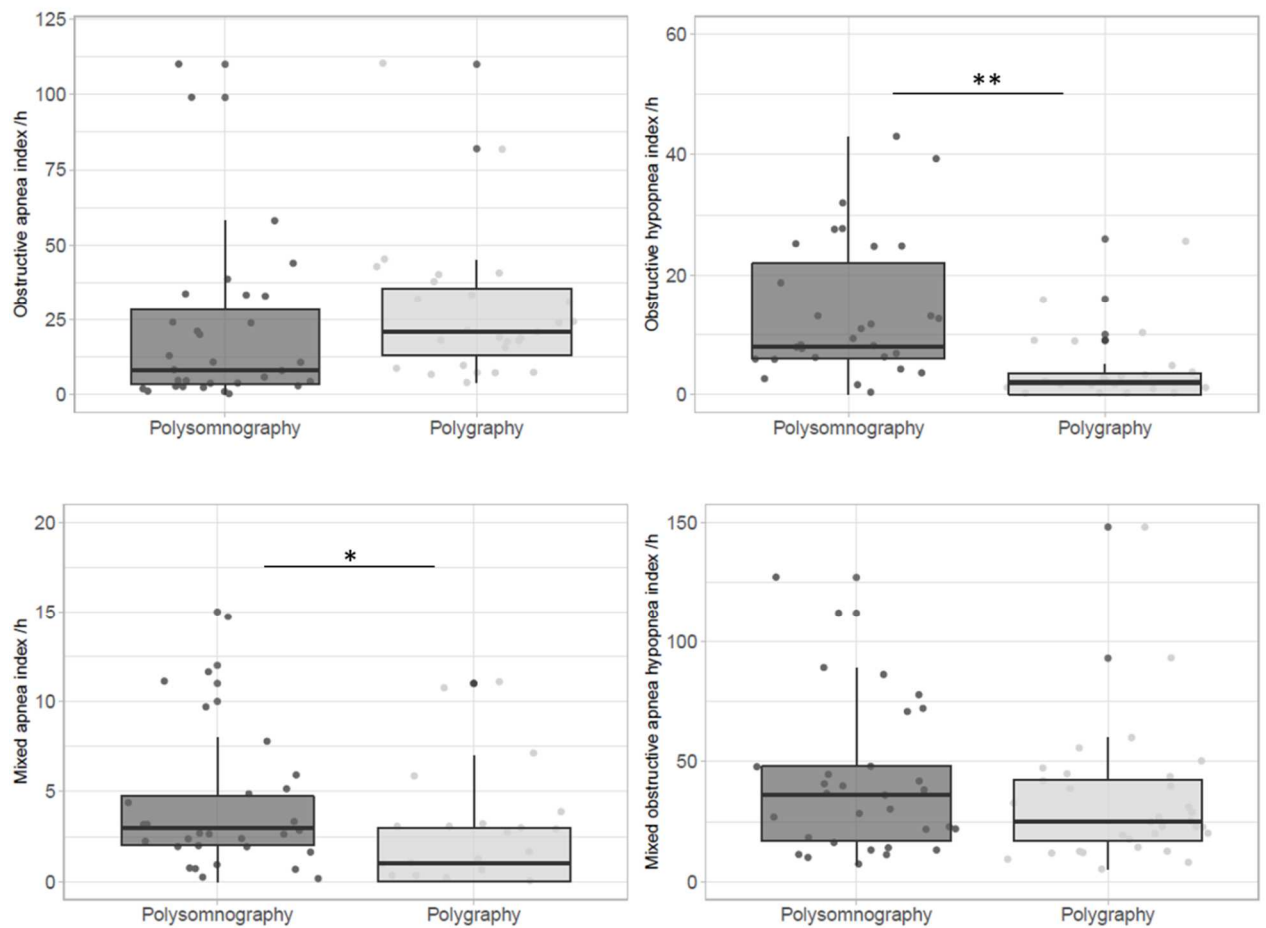
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Figure 1. Comparison of respiratory indexes (Obstructive apnea index, Obstructive hypopnea index, Mixed apnea index, Mixed obstructive apnea hypopnea index) between polysomnography and polygraphy in 20 infants with Robin sequence without regard to sleep position.



Legend of Figure 1: The central line of the boxplot represents the median; the superior and inferior lines represent respectively the third and the first quartiles. **: p = 0.01; *: p = 0.03

Table 1: Characteristics of Robin infants before and during polysomnography

	Study population n = 20
Median GA at birth, weeks [IQR]	39 [37-40]
Median birth weight, g [IQR]	3170 [2378-3430]
Sex ratio M/F	8/12
Median age at PSG, days [IQR]	43 [25-114]
Median weight at PSG, kg [IQR]	4[3-4]
Prenatal diagnosis, n (%)	4 (20)
Type of PRS, n (%)	
- Isolated	15 (75)
- Associated or syndromic (type 3)	5 (25)
Digestive characteristics	
- Sucking-swallowing disorders, n (%)	20 (100)
- Gastroesophageal reflux treatments, n (%)	
Thickened milk + proton inhibitor or anti acid	13 (65)
Thickened milk + proton inhibitor or anti acid	4 (20)
Missing data	3 (15)
- Nasogastric tube, n (%)	
Before PSG	13 (65)
During PSG	0 (0)
Respiratory characteristics	
- Respiratory signs*, n (%)	14 (70)
- Respiratory support at time of PSG, n (%)	
None	15 (71)
Non-invasive ventilation **	5 (24)
Nasopharyngeal tube + CPAP	1 (5)

*GA: gestational age, PSG: polysomnography, RS: Robin sequence, *respiratory signs of OSAS: snoring, noisy breathing, apneas, apparent life-threatening event, frequent arousals because of respiratory efforts, desaturation, and/or stridor, **: CPAP or high flow nasal cannula, CPAP: Continuous Positive Airways Pressure*

Table 2. Sleep characteristics of Robin infants

	Study population n= 20
TST, minutes	170 [101-239]
Sleep efficacy, %	80 [62-91]
Sleep efficiency, %	77 [56-90]
Arousals index, n/h	24 [17-35]
Respiratory arousal index, n/h	20 [11-29]
Proportion of sleep stages, % of total sleep time	
REM	36 [26-45]
NREM	29 [12-56]
Respiratory indexes, n/h	
MOAHI in REM	67 [39-98]
MOAHI in NREM	21 [7-38]

All data are expressed as median [IQR]. REM: rapid eye movement stage; NREM: non rapid eye movement stages; TST: total sleep time

Table 3. Respiratory characteristics in polysomnography and polygraphy of Robin infants

	Polysomnography n = 20	Polygraphy n = 20	p value
	Median [Q1-Q3]	Median [Q1-Q3]	
Obstructive apnea index (OAI), n/h	7 [4-25]	19 [15-31]	0.05
Obstructive hypopnea index (OHI), n/h	8 [8-19]	2 [0-3]	0.01 **
Mixed apnea index (MAI), n/h	3 [2-5]	2 [0-6]	0.03 *
Mixed obstructive apnea hypopnea index (MOAHI), n/h	26 [18-56]	27 [18-38]	0.43
Mean SpO ₂ , %	97 [94-97]	-	
Time with SpO ₂ < 90%, %	1 [0-10]	-	
Oxygen desaturation index > 3% (OD3%), %	34 [19-59]	-	
Mean P(tcCO ₂), mmHg	52 [47-55]	-	
Maximum P(tcCO ₂), mmHg	56 [51-60]	-	
Time spent with P(tcCO ₂) > 50 mmHg, %	49 [2-100]	-	

*SpO₂: pulse oxymetry, TST: total sleep time, P(tcCO₂): transcutaneous carbon dioxide pressure. p value <0.05 was considered as statistically significant. **: p = 0.01; *: p = 0.03*