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Polygraphic respiratory events during sleep with noninvasive ventilation in children: description, prevalence, and clinical consequences

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Abstract *Purpose:* The optimal
monitoring during sleep with nonin-
vasive positive pressure ventilation
(NPPV) has not been validated in
children. The aim of the study was to
describe on polygraphic (PG)
recordings the respiratory events and
associated autonomic arousals (AA)
and/or 3 % desaturations (DS3%)
during nocturnal NPPV. *Meth-
ods:* This was a prospective
descriptive study performed in the
pulmonology unit of a pediatric uni-
versity hospital. Consecutive patients
admitted for routine follow-up of
long-term NPPV were enrolled.
Nocturnal PG during sleep with
NPPV was performed. A second PG
was performed after adjustment of the
ventilatory settings when a respira-
tory event occurred more than
50 times/h. *Results:* The PG trac-
ings of 39 patients (age range
1–18 years) were analyzed. Underly-
ing diagnoses included
neuromuscular disease ($n = 13$),
obstructive sleep apnea ($n = 15$), and
lung disease ($n = 11$). Unintentional
leaks, patient–ventilator asynchro-
nies, decrease in ventilatory drive,
upper airway obstruction with or
without reduction of ventilatory
drive, and mixed events were
observed in 27, 33, 10, 11, 12, and
3 % of the patients, respectively. A
predominant respiratory event was
observed in all patients. The mean
duration spent with respiratory events
was 32 ± 30 % (range 3–96 %) of
total recording time. Unintentional
leaks were the most frequently asso-
ciated with AA, whereas patient–
ventilator asynchronies were rarely
associated with AA or DS3%. In eight
re-evaluated patients, a decrease in
the main event was observed
($p = 0.005$). *Conclusion:* Respira-
tory events during sleep with NPPV
are common in children treated with
long-term NPPV. Consequences of
respiratory events vary according to
the type of event with unintentional
leaks being associated preferentially
with AA.

Keywords Polygraphy · Sleep ·
Noninvasive positive pressure
ventilation · Patient–ventilator
asynchrony · Arousal · Desaturation

Introduction

Although long-term noninvasive positive pressure ventilation (NPPV) is increasingly used in children with chronic alveolar hypoventilation, the optimal monitoring has not been validated. The main aim of NPPV is to correct nocturnal gas exchange but restricting monitoring to this therapeutic goal may not be sufficient as normal gas exchange during sleep does not exclude poor sleep quality or abnormal sleep structure [1]. Moreover, NPPV is usually started and set during the day when the patient is awake. Sleep-related changes in ventilatory drive, respiratory, and upper airway mechanics as well as respiratory muscles activity are likely to influence the efficacy and tolerance of NPPV [2]. In addition, NPPV itself may induce undesirable events and residual respiratory events may persist even under optimal NPPV settings. These events were recently described by Gonzalez-Bermejo et al. [3] and comprise unintentional leaks, patient–ventilator asynchronies, decrease in ventilatory drive, upper airway obstruction with or without reduction of ventilatory drive, and mixed events. A polygraphic (PG) or polysomnographic (PSG) recording is mandatory for the detection of these events. Although this strategy has been recommended [4], it may not be feasible on a routine basis in every pediatric NPPV center. Also, before recommending a systematic PG or PSG recording in children treated with long-term NPPV, a description of respiratory events during NPPV is necessary. Moreover, the clinical consequences of a specific event have to be taken in account. Indeed, it is possible that the potential deleterious effects may vary according to the type of NPPV-associated events and underlying disease.

Within this context, we performed a systematic sleep PG recording with NPPV in a consecutive cohort of children treated with long-term NPPV at home. The aims of the study were first to describe the respiratory events according to the SomnoNIV group definitions [3] and second to analyze each event's association with an autonomic micro-arousal (AA) and/or a 3 % oxyhemoglobin desaturation (DS3%) [5, 6].

Materials and methods

Patients

All consecutive patients treated with home nocturnal NPPV were prospectively enrolled during their routine follow-up between October 2011 and April 2012 if (1) they were in a stable condition, (2) they were using NPPV for at least 1 month, and if (3) they had no clinical symptoms of nocturnal hypoventilation such as frequent arousals, nocturnal agitation, or daytime fatigue and sleepiness. Patients were divided into a neuromuscular (NM) disease group, a lung disease group, and an obstructive sleep apnea syndrome (OSAS) group. The study was conducted in agreement with the French regulations and had received appropriate legal and ethical approval from the Institutional Review Board of the French Learned Society for Respiratory Medicine (Société de Pneumologie de Langue Française).

The home NPPV program

Criteria for long-term NPPV were a maximal nocturnal transcutaneous carbon dioxide tension ($PtcCO_2$) greater than 50 mmHg and/or a minimal pulse oximetry (SpO_2) less than 90 % for at least 2 % of night-time [7, 8]. All the patients were ventilated with a pressure-targeted mode, with ["pressure support–volume targeted ventilation (PS-VT)"] or without ("bilevel ventilation") a target volume, with a back-up respiratory rate depending on the age of the patient. The interface was a nasal or face mask, with or without a manufacturer leak, according to the ventilatory mode and settings and patient's tolerance [9]. Objective compliance was assessed by the ventilator's software. None of the patients included in the study required supplemental oxygen therapy during NPPV.

Study design and procedure

Overnight recordings of nocturnal PG, gas exchange, and actigraphy were performed during a single night hospitalization while respecting the child's habitual sleep. Synchronization was ensured between the devices.

Nocturnal ventilatory PG

An overnight PG assessment was performed using the Somnoscreen device (SOMNOscreen TM plus PSG+, SOMNOmedics GmbH, Germany) which is an adaptable PG device that can be connected to a $PtcCO_2$ monitor (SenTec Digital Monitor, SenTec Inc, Therwil, Switzerland). The recorded data included airflow using a pneumotachograph, airway pressure included in the NPPV line, body position, thoracic and abdominal movements assessed with piezoelectric belts (SleepSense, Multiple use Inductive Plethysmography Band, S.L.P. Ltd., Israel), SpO_2 and photoplethysmographic pulse wave amplitude (PWA). The pneumotachograph was inserted between the interface and the expiratory valve in case of a non-leak ventilation or between the interface (with an integrated manufacturer leak) and the circuit in case of a leak ventilation.

AA were identified as a reduction in PWA greater than 30 % of the baseline amplitude [10, 11]. Mean and

minimal nocturnal SpO_2 as well as the percentage of night-time with mean SpO_2 less than 90 % were determined. Desaturation index was defined as the number of SpO_2 desaturations above 3 % from baseline value per hour of PG recording (DS3%) and AA related to a DS3% (AA + DS3%) were quantified [5, 6, 10].

Respiratory events (see online supplement)

A respiratory event is characterized by the occurrence of a modification, discontinuity, or instability of ventilation. Respiratory events were defined and scored using the SomnoNIV Group definitions and quantified as the number of events per PG recording time [3]. A single respiratory event was defined as an event lasting at least two respiratory cycles, which varied according to the age and respiratory rate of the patient [12]. When a combination of several events occurred, we checked first for non-intentional leaks. Continuous leaks were systematically scored as the predominant event. In the absence of continuous leaks, the predominant event was assumed to be the most frequent event encountered solely. As an example, if a PG recording displayed numerous upper airway obstructions with increase in respiratory drive, associated from time to time with asynchrony, the predominant event was assumed to be the obstruction and asynchrony the consequent event. Respiratory events associated with a DS3%, an AA, or an AA + DS3% were also quantified by means of the pulse wave amplitude (PWA) [5, 6].

Six types of respiratory events were identified (see online supplement for complete description): unintentional leaks, patient–ventilator asynchrony, decrease in ventilatory drive, upper airway obstruction with or without reduction of ventilatory drive, and mixed events.

Nocturnal carbon dioxide monitoring

An overnight $PtcCO_2$ recording was performed by the combined $SpO_2/PtcCO_2$ monitor (SenTec Digital Monitor, SenTec Inc, Therwil, Switzerland) [13]. The mean, maximal $PtcCO_2$, and the percentage of night-time with a $PtcCO_2$ greater than 50 mmHg data were gathered.

Overnight actigraphy

An overnight actigraphy recording was performed simultaneously (Actiwatch, Cambridge Neurotechnology Ltd., Cambridge, UK) [14, 15]. Sleep efficiency was defined as the percentage of time spent asleep whilst in bed. The movement and fragmentation index (MFI) was defined as the sum of the minutes of movement expressed as a percentage of the total recording time (movement index) and the number of times that sleep was terminated

after 1 min expressed as a percentage of the total estimated sleep time (fragmentation index) [16–18]. The sum was expressed as a percentage.

Capillary arterialized blood gases

Capillary arterialized blood gases were performed in the morning following the sleep study [19].

Re-evaluation PG study after setting adjustments

When the predominant respiratory event exceeded 50 events/h on the baseline PG recording, an adjustment of the NPPV setting was performed and a second nocturnal PG was performed after 2–4 weeks.

Statistical analysis

Data are presented as mean $\pm$ SD. Baseline characteristics and rate of respiratory events associated or not with DS3% or AA were compared among the three groups using a one way analysis of variance or a Kruskal–Wallis one-way analysis of variance on ranks if necessary. A Chi-square analysis was performed to compare the distribution of the different respiratory events associated or not with a DS3% or an AA. Comparisons of data before and after resetting of NPPV were done using a paired t test or Wilcoxon signed rank test when appropriate. The correlation of the respiratory event indexes with nocturnal carbon dioxide monitoring, actigraphy, and capillary arterialized blood gases was performed using the Pearson correlation test or Spearman correlation test when appropriate. A p value of less than 0.05 was considered statistically significant.

Results

Thirty-nine patients with NM disease ($n = 13$), lung disease ($n = 11$), or OSAS ($n = 15$) were included in the study (Table 1). The mean age of the total population was 10.0 ± 5.8 years with no difference between the three groups. All the patients with an NM or a lung disease were ventilated with a PS-VT mode and all the patients but one with OSAS were ventilated with a bilevel ventilation mode. All the patients used a single circuit coupled either with an expiratory valve in eight patients or an intentional leak valve in the remaining patients. For the total population, the mean inspiratory positive airway pressure was 14.3 ± 2.2 cmH$_2$O. Eight patients had no expiratory positive airway pressure set; the mean expiratory positive airway pressure was 4.2 ± 2.8 cmH$_2$O for the other 31 patients. The patients with lung disease had a significantly

lower mean SpO_2 and higher maximal $PtcCO_2$ during NPPV as compared to the two other groups with no difference in daytime blood gases (Table 1). Sleep efficiency was within the normal limits among the three groups with a fragmentation index slightly above normal values [20–22]. Compliance with NPPV was good and not different between the three groups (Table 1).

Table 2 shows the scoring of the PG tracings for the total population and among the three groups. Respiratory events were extremely common and all the patients had at least one. Patient–ventilator asynchrony and unintentional leaks were the most common events in the patients with NM or lung disease, whereas unintentional leaks, decrease in ventilatory drive, and upper airway obstruction with or without reduction of ventilatory drive were almost equally observed in the OSAS group.

For a given patient, there was a predominant respiratory event representing 75 ± 19 % of total time spent in respiratory events. The mean duration spent in a respiratory event was 32 ± 30 % (range 3–96 %) of total recording time.

Figure 1 shows the association between the different respiratory events and a DS3%, an AA, or an AA + DS3% for the total population, the NM disease group, the lung disease group, and the OSAS group. Unintentional leaks

Table 1 Patients characteristics with daytime and nocturnal gas exchange and actigraphic sleep quality

	NM disease ($n = 13$)	Lung disease ($n = 11$)	OSAS ($n = 15$)	Total population ($n = 39$)	p value
Anthropometrics					
Gender (M/F)	7/6	4/7	10/5	21/18	
Age (years)	10.0 ± 7.7	11.0 ± 4.7	9.7 ± 4.7	10.0 ± 5.8	0.979
NPPV					
NPPV mode	PS-VT	PS-VT	Bilevel ventilation ($n = 14$) PS-VT ($n = 1$)	Bilevel ventilation ($n = 14$) PS-VT ($n = 25$)	
Objective compliance (h/night)	7.7 ± 1.7	8.5 ± 1.2	7.3 ± 1.8	7.8 ± 5.8	0.216
Diurnal gas exchange					
PaO_2 (mmHg)	87.0 ± 8.2	76.4 ± 15.6	88.0 ± 7.6	84.3 ± 11.5	0.065
$PaCO_2$ (mmHg)	38.8 ± 5.2	43.4 ± 3.4	41.0 ± 4.3	40.8 ± 4.8	0.105
Nocturnal gas exchange					
Minimal SpO_2 (%)	90.3 ± 6.3	85.8 ± 6.0	88.0 ± 7.7	88.2 ± 6.9	0.150
Mean SpO_2 (%)	96.7 ± 1.5	94.5 ± 3.9^a	97.3 ± 1.9	96.3 ± 2.7	**0.026**
% time spent with SpO_2 <90 % (%)	0.15 ± 0.30	0.29 ± 0.69	0.27 ± 0.73	0.23 ± 0.59	0.489
Maximal $PtcCO_2$ (mmHg)	43.5 ± 6.7	49.3 ± 7.9^b	45.0 ± 3.8	45.8 ± 6.3	0.083
Mean $PtcCO_2$ (mmHg)	38.6 ± 6.0	46.0 ± 7.8	39.4 ± 4.3	41.0 ± 6.7	**0.012**
% time $PtcCO_2$ >50 mmHg (%)	5.0 ± 17.3	9.5 ± 24.4	0.2 ± 0.8	4.4 ± 16.0	0.189
Actigraphy data					
Sleep efficiency (%)	82.5 ± 13.4	83.9 ± 10.3	82.7 ± 10.9	83.1 ± 10.9	0.959
Fragmentation index (%)	35.0 ± 20.8	35.0 ± 14.3	33.0 ± 17.5	34.1 ± 16.7	0.805

NM neuromuscular, *OSAS* obstructive sleep apnea syndrome, *NPPV* noninvasive positive pressure ventilation, *PS-VT* pressure support–volume targeted ventilation, *PaO₂* partial arterial oxygen pressure, *PaCO₂* partial arterial carbon dioxide pressure, *SpO₂* pulse oximetry, *PtcCO₂* transcutaneous carbon dioxide tension

[a] Statistical difference between the lung disease and OSAS groups
[b] Statistical difference between lung disease versus OSAS and NM disease groups

Table 2 Description of the respiratory events during a polygraphy recording: percentage of recording time spent with a single respiratory event (mean $\pm$ SD) and number of subjects with the single events

	Unintentional leaks	Patient–ventilator asynchronies	Decrease in ventilatory drive	Upper airway obstruction with decrease in ventilatory drive	Upper airway obstruction	Mixed events
NM disease ($n = 13$)	31 ± 38 % ($n = 7$)	38 ± 46 % ($n = 9$)	8 ± 16 % ($n = 6$)	11 ± 23 % ($n = 5$)	4 ± 7 % ($n = 6$)	0 ± 0 % ($n = 0$)
Lung disease ($n = 11$)	37 ± 41 % ($n = 8$)	50 ± 39 % ($n = 10$)	4 ± 8 % ($n = 5$)	8 ± 12 % ($n = 5$)	16 ± 18 % ($n = 7$)	0.1 ± 0.1 % ($n = 1$)
OSAS ($n = 15$)	17 ± 31 % ($n = 6$)	11 ± 24 % ($n = 6$)	17 ± 33 % ($n = 10$)	20 ± 34 % ($n = 7$)	16 ± 25 % ($n = 9$)	9 ± 26 % ($n = 8$)
Total population ($n = 39$)	27 ± 36 % ($n = 21$)	33 ± 40 % ($n = 25$)	10 ± 23 % ($n = 21$)	11 ± 28 % ($n = 17$)	12 ± 10 % ($n = 22$)	3 ± 16 % ($n = 9$)

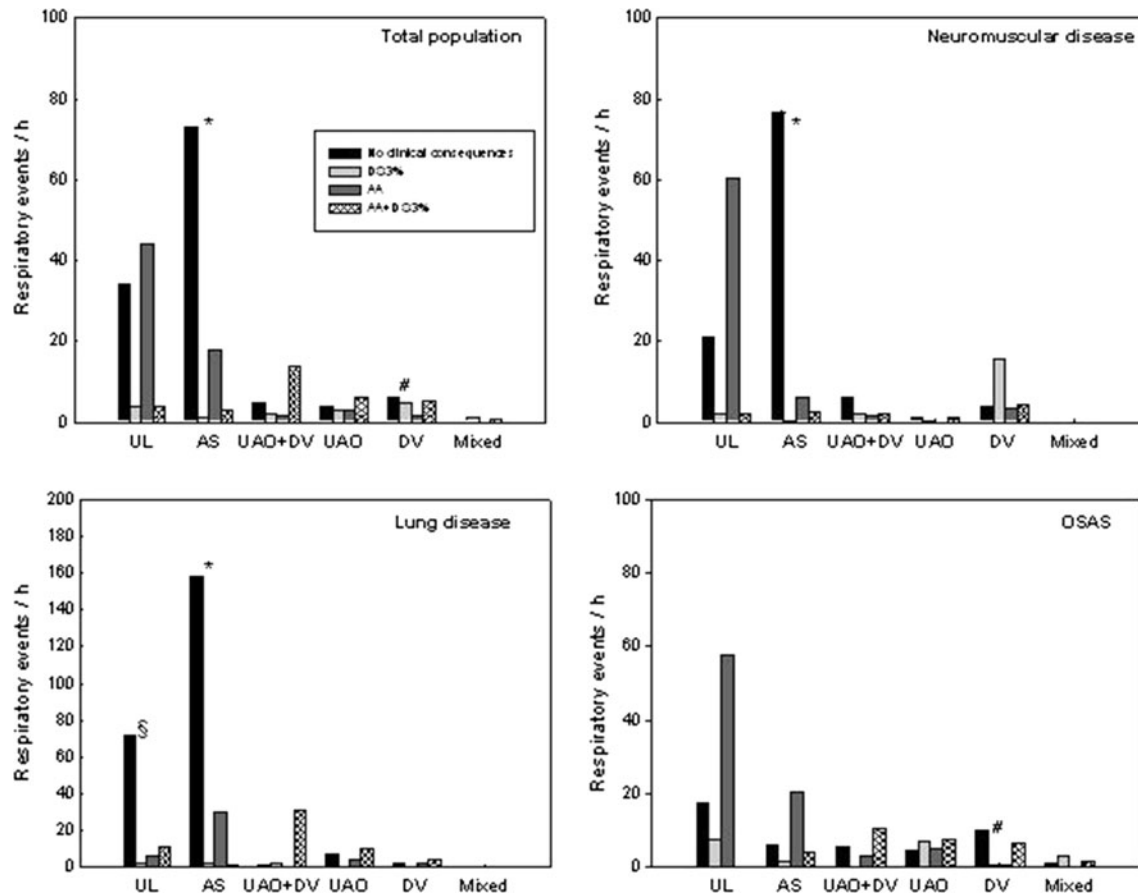*NM* neuromuscular, *OSAS* obstructive sleep apnea syndrome

Fig. 1 Incidence of respiratory events and their association with autonomic arousals (AA), 3 % desaturation (DS3%) or autonomic arousals with 3 % desaturation (AA + DS3%), in the total population, the patients with neuromuscular disease, lung disease, and obstructive sleep apnea syndrome (OSAS). *UL* unintentional leaks, *AS* patient–ventilator asynchronies, *UAO + DV* upper airway obstruction with decrease in ventilatory drive, *UAO* upper airway obstruction, *DV* decrease in ventilatory drive, *Mixed* mixed events. *$p < 0.05$ between "No clinical consequences" and "3%DS", "AA", and "AA + DS3%", #$p < 0.05$ between "No clinical consequences" and "3%DS", and "AA" §$p < 0.05$ between "No clinical consequences" and "3%DS"

were very frequent. The majority of these unintentional leaks were associated with AA in the total population, the NM and OSAS groups but not in the lung disease group. Patient–ventilator asynchronies were also very common but conversely, the vast majority of these asynchronies were not associated with an AA or a DS3%, except for the OSAS group. Upper airway obstruction with decrease in ventilatory drive, although not common, was frequently associated with an AA + DS3% in the total population and particularly in patients with lung disease or OSAS. Decrease in ventilatory drive was frequently associated with a DS3% in NM patients.

No correlation was observed between the frequency of any respiratory event and the actigraphic sleep efficiency and fragmentation index, daytime arterial blood gases, mean and maximal nocturnal $PtcCO_2$, or the objective compliance with NPPV.

In eight patients, a second PG during NPPV was performed after the adjustment of the ventilatory settings aimed
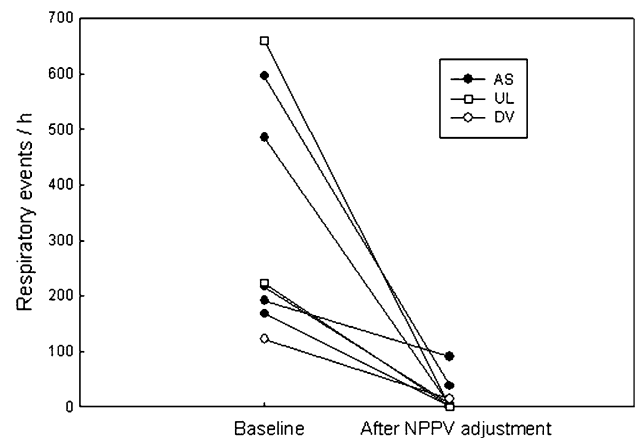

Fig. 2 Evolution of the predominant respiratory event before and after changes of the noninvasive positive pressure ventilation (NPPV) settings in eight patients. *AS* patient–ventilator asynchronies, *UL* unintentional leaks, *DV* decrease in ventilatory drive

at reducing the predominant respiratory event (patient–ventilator asynchrony in 5 patients, unintentional leaks in 2 patients, and decrease in ventilatory drive in 1 patient) (Fig. 2). A significant reduction in the main respiratory event was observed on the second PG tracing ($p = 0.005$).

Discussion

This study is the first to analyze the PG recordings during sleep in children treated with long-term NPPV and to analyze the associated DS3% and AA. The main observations of the study are that respiratory events are extremely common with each patient harboring a predominant event; unintentional leaks were most frequently associated with an AA, whereas patient–ventilator asynchronies were rarely associated with an AA or a DS3%; and finally, the PG recording was helpful to adapt the ventilatory settings in order to reduce the predominant respiratory event.

The optimal monitoring of patients treated with long-term NPPV is still a matter of debate. There is agreement on SpO$_2$ recording as a minimal requisite but two studies from our group have shown that overnight PtcCO$_2$ is also mandatory as patients may remain hypercapnic despite normal SpO$_2$ levels [13, 23]. Some authors recommend a systematic PSG as this investigation represents the gold standard for the qualitative and quantitative diagnosis of sleep abnormalities [4]. But PSG is time-consuming, expensive, technically demanding, and not feasible on a routine basis. Moreover, the analysis of the respiratory events during sleep with NPPV on a PSG requires an extensive knowledge not only of the patient's sleep but also of the setting and functioning of the ventilator and the patient–ventilator interaction [2, 3]. Within this context, a descriptive analysis of all respiratory events observed during long-term NPPV in stable children is a preliminary and essential step.

The main observations of the present study are that respiratory events are common, even in compliant children who do not complain of any residual sleep-disordered symptom, with unintentional leaks and asynchrony representing the most common events. Reports in the literature have mainly dealt with these two types of respiratory events. In a recent systematic study in adults, Rabec et al. [24] also found that unintentional leaks represent the most frequent event occurring under NPPV. Unintentional leaks in adult patients with NM disease receiving noninvasive or invasive ventilation have been shown to be associated with persistent daytime hypercapnia [25]. In the present study, no association was observed between the occurrence of unintentional leaks and daytime PaCO$_2$ levels. This may be explained by the fact that daytime hypercapnia, defined as a PaCO$_2$ greater than 45 mmHg, was observed only in five patients and

that the volume of leaks was not calculated. In a step by step strategy monitoring during sleep, the occurrence and the severity of leaks can be addressed firstly by the use of the devices' built-in software [1, 26]. When leaks are detected some simple technical adjustments can be proposed before a PG or PSG [24, 27]. The incidence and consequences of one type of patient–ventilator asynchrony, i.e., ineffective inspiratory efforts (IIE), have been analyzed in 48 adults enrolled in a long-term home NPPV program [28]. A positive correlation was observed between the IIE index and the time spent with an SpO$_2$ less than 90 %, with the patients with an IIE index above 100 having a poor nocturnal gas exchange. Conversely, patient–ventilator asynchrony was not found to be associated with a significant SpO$_2$ desaturation or an increase in PtcCO$_2$ in another study with adult patients with obesity hypoventilation [29]. Again, a comparison with our results is difficult because of the small number of patients with an SpO$_2$ less than 90 % ($n = 2$) under NPPV.

The clinical consequences of respiratory events occurring during sleep with NPPV represent the major issue. If the respiratory event is not associated with any deterioration in gas exchange or sleep architecture and quality and does not influence the patient's compliance with NPPV, its correction and thus detection are not mandatory. It is thus essential to detect and quantify the most deleterious respiratory events, i.e., those associated with poor subjective or objective sleep quality, gas exchange, and/or compliance. We chose to analyze the AA and SpO$_2$ for the following reasons. PSG with electroencephalographic (EEG) recording was not performed in the present study, precluding the identification of EEG defined micro-arousals. As arousals are recognized as a major cause of poor sleep quality, we took the option to use the PWA and actigraphic data to estimate the AA and the awakenings as these two tools have already been validated [5, 10, 11, 15, 30]. Moreover, in children, the arousal threshold is higher than in adults and we have previously demonstrated that autonomic activation is more sensitive than EEG to identify sleep disruption associated with abnormal respiratory events [31]. We observed that the majority of the unintentional leaks were associated with AA in the patients with NM disease and OSAS (Fig. 1). Conversely, patient–ventilator asynchronies, although being very common, were rarely associated with an AA or a DS3%. The consequences of the respiratory events on the PtcCO$_2$ changes were not analyzed in the present study because time-course changes in PtcCO$_2$ are more progressive and less variable than SpO$_2$ changes [27] and because the large majority ($n = 34$) of the patients had a maximal PtcCO$_2$ less than 50 mmHg during sleep.

Our study has several limitations. The PG recording during NPPV was informative and allowed one to decrease the predominant residual respiratory event in the

subgroup of patients reassessed after setting changes. As there is no validated cutoff for respiratory events during NPPV in children, we took arbitrarily a cutoff of 50 events/h because this number emerged as being very high. However, clinically relevant cutoff values for each respiratory event in the different pediatric populations need to be validated in future studies. We used the PWA to identify AA associated or not with a desaturation. We acknowledge that the use of PWA as a marker of AA has not been validated against changes in cortical electroencephalographic activity during NPPV, but a recent study from our group showed the usefulness of this marker to assess the efficacy of NPPV during sleep in children [6]. As underlined by the SomnoNIV Group, the flow and pressure signals during NPPV vary according to the ventilatory modes, the type of circuit, and the ventilatory settings [3, 32]. Seventy percent of the patients in the present study used a PS-VT mode, during which the ventilator is estimated to increase the inspiratory pressure in order to maintain a minimal tidal volume. The use of this new mode may add an additional "ventilator" factor that has not been described for the more "classical" volume or pressure targeted modes by the SomnoNIV Group [3, 32].

In conclusion, this study reports the high frequency of respiratory events during sleep with NPPV in stable children treated with long-term NPPV at home. Unintentional leaks and patient–ventilator asynchronies were the most common events but the latter were well tolerated with regard to AA and DS3%. As PG recording of sleep during NPPV requires a high level of expertise, future studies should assess which step by step strategy should be proposed for the monitoring of NPPV [26] with the final goal to reduce the number of hospitalizations for NPPV in children.

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