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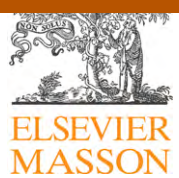

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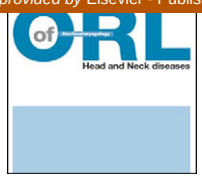

ORIGINAL CLINICAL RESEARCH

Questionnaire and nocturnal oxymetry in children with adenotonsillar hypertrophy☆

P. Contencin[a,*], E. Malorgio[b], S. Noce[b], V. Couloigner[a], A. Vigo[b]

[a] *Service d'ORL, hôpital Necker—Enfants-Malades, AP—HP, université Paris-5, 149, rue de Sèvres, 75015 Paris, France*
[b] *Centro SIDS, Ospedale Infantile Regina Margherita, Regione Piemonte, Turin, Italy*

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KEYWORDS
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Summary
Objectives: Draw up an evaluative approach to the diagnostic contribution of nocturnal oximetry associated with a parental questionnaire in children with adenotonsillar hypertrophy (ATH).
Patients and methods: Analysis of a parental questionnaire on sleep patterns and oximetry recording made in children with ATH. The results of the oximetry were compared to the data gathered from the questionnaire.
Results: Of the 342 children (age range, 3 months to 14 years), 209 permanent snorers, 115 occasional snorers, and 18 non-snorers were identified. The proportion of positive oximetry readings varied from 31.6 to 0% and the difference was significant between the first group and the two others ($p < 0.001$). The data were correlated for four symptoms, including snoring and sleep apnea. The absence of snoring always gave a negative oximetry reading.
Conclusions: In cases of ATH, a negative questionnaire can predict that oximetry will not be useful and if necessary advise for a polysomnography for an exclusion diagnosis. In contrast, a positive questionnaire followed by a positive oximetry argues in favor of the polysomnography not being useful.

Introduction

In children, sleep-disordered breathing (SDB) is a frequent pathology. Depending on whether obstructive sleep apnea syndrome (OSAS) or simple chronic snoring is considered, the prevalence — based on the clinical description — varies from 1% to more than 30% and the repercussions range from problems concentrating and learning difficulties to growth impairment to the very rare cardiac deficiency [1].

The most frequent cause of SDB in the child is hypertrophy of the palatine tonsils and adenoids. Moreover, adenotonsillar surgery substantially improves breathing or even, in many cases, cures these children [2]. The existence of SDB cannot be confirmed with parental questioning only, even if it is made more sensitive with anamnesis, the study of diurnal behavior and expressed in scores [3]. Yet the disadvantages and perioperative risks of adenotonsillectomy are not insignificant, whether in terms of pain, hemorrhage

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* Corresponding author. Phone: +33 1444 94454; Fax: +33 1444 94690.
E-mail address: p.contencin@nck.aphp.fr (P. Contencin).

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of varying intensity, transitory aggravation of postoperative apnea (the first night), or even death (1/40,000). Complementary exams evaluating the intensity of SDB are therefore necessary to detail the indications for adenotonsillectomy and to detect severe cases of OSAS that contraindicate ambulatory surgery because of the immediate postoperative risks [4,5].

The reference test to confirm the diagnosis of OSAS in children is the polysomnograph (PSG), carried out in the sleep laboratory, which can also detect other, less severe SDB such as an increase in respiratory effort, hypercapnia, a high number of micro-arousals, or nasal airflow limitation [6,7]. The harmful consequences of this SDB on the child's development have been demonstrated [8,9]. PSG nevertheless has two major drawbacks: it is very expensive and access is difficult. Given the impossibility — except in very particular circumstances — of generalizing this exam, it would seem useful to test simpler forms of exploration based on observations of the child's sleep and simplified recordings. Nocturnal pulse oximetry has demonstrated insufficient sensitivity but good specificity in characterizing children with the most serious forms of OSAS and situations at risk for postoperative respiratory impairment [3,10]. The objective of this study was to investigate the value of nocturnal oximetry associated with a parental questionnaire to specify the importance of SDB in children with adenotonsillar hypertrophy (ATH).

Patient and methods

This study investigated all the children seen consecutively in the ORL department at the Regina Margherita Hospital in Turin (Italy), and selected between January 2002 and December 2005 for their ATH and not for their functional signs. ATH was diagnosed clinically on the oropharynx examination: ''kissing'' or voluminous palatine tonsils adjoining the median line. In absence of tonsillar abnormality, adenoid hypertrophy was demonstrated with nasendoscopy. It is known that in absence of patent tonsillar hypertrophy, adenoid obstruction is the leading cause of SDB in this age group [11]. All the children in the study underwent nocturnal pulse oximetry recording after their parents had completed, with a nurse, a questionnaire on the existence of diurnal and nocturnal symptoms, including snoring (Table 1). The child's weight stagnation (lasting several months), thinness, and constant mouth breathing (subjective aspects during the exam) were noted in consultation by the pediatrician in charge of the study. During this consultation, consent for the study was obtained from the parents, who where trained in observing the functional signs that needed to be noted during the days and nights preceding the oximetry recording.

Oximetry was studied for 8 h at home using a VitaGuard VG 3000® with a Masimo Set® (Getemed, Teltow, Germany) equipped with a multichannel recording device to record a measurement every 6 s. The light sensor for pulse oximetry was lightly attached to the child's index finger during the evening by a specialized hospital nurse and the child then returned home; the parents were instructed to note the times the child awoke and any abnormal movements the child made. The recording was analyzed in the morning on a personal computer using Vitawin® software. The graphs were read on the screen and the data verified meticulously, after manual exclusion of any common artifacts that the software may not have taken into account (mean reading time, approximately 30 min). The results of the analysis were interpreted based on criteria reported in the literature [10]. To characterize the abnormal recordings, the following criteria were retained. Positive oximetries were those that included at least three clusters comprising at least five episodes per 30-min period of lowered SpO_2 by at least 4% compared to baseline and at least three episodes at 90% SpO_2 or less. The negative recordings were characterized by the absence of any desaturation cluster and a recording that never went below 90% SpO_2 [10]. All the test results that were neither positive nor negative were labeled intermediate. Their separate analysis contributed no intrinsic value to the oximetry data and they were grouped with the negative results so that the positive results could be compared with all the other results. The data from these double-blind investigations were compared and analyzed searching for diagnostic and prognostic factors that could improve the management of these children. Every comparison between the questionnaire data and the oximetry data was subjected to univariate analysis. The chi-square test, the odds ratio, and, if necessary, the Fisher exact test were used for the statistical analysis of the data. A result was considered significant when the risk of error was less than 5%.

Results

Of the 342 children analyzed, there were 231 males and 111 females (M/F ratio, 2.08/1), aged between 3 months and 14 years (mean, 4.65 years). The questionnaire differentiated four groups: 209 permanent snorers (group I), 115 occasional snorers without ($n = 68$) or with ($n = 47$) rhinitis (groups II and III, respectively), and 18 non-snoring children (group IV), 16 of whom had disturbed sleep (14 cases of agitation, eight cases of apnea) according to their parents. The oximetry results were positive in 31.6%, 8.8%, 0%, and 0% of each group, respectively. The difference was significant between the first group and the three others ($p < 0.001$). There was a clear correlation between the questionnaire data and the positive oximetry data on the following points: snoring (permanent or intermittent), apnea,

Table 1 Contents of the medical questionnaire for parents.

Daytime symptoms		
Failure to thrive	Yes	No
Thinness (general aspect)	Yes	No
Rhinolalia clausa	Yes	No
Permanent mouth breathing	Yes	No
Recurrent rhinitis/rhinopharyngitis	Yes	No
Sleepiness	Yes	No
Nighttime symptoms		
Observed ''apneas'' (respiratory obstructions)	Yes	No
Snoring: in case of URTI	Yes	No
Intermittent (even with no infection)	Yes	No
Habitual, permanent	Yes	No

URTI: upper respiratory tract infection.

Table 2 Distribution of cases by snoring or no snoring and oximetry.

Symptom	Groups	Oximetry +	Oximetry −	*P*	Sensitivity (%)
Snoring	I + II + III	75	249	< 0.05	100
No snoring	IV	0	18		
Predictive value		NPV = 100%			

Table 3 Distribution of cases by permanent snoring or no permanent snoring and oximetry.

Symptom	Groups	Oximetry +	Oximetry −	*P*	Sensitivity (%)
Permanent snoring	I	66	143	< 0.0001	88
No permanent snoring	II + III + IV	9	124		
Predictive value		NPV = 93.2%			

NPV: negative predictive value of the absence of symptoms.

Table 4 Distribution of cases by permanent or intermittent snoring and oximetry (non-snorers excluded).

Symptom	Groups	Oximetry +	Oximetry −	*P*	Sensitivity (%)
Permanent snoring	I	66	143	< 0.001	88
Intermittent snoring	II + III	9	106		
Predictive value		NPV = 92.2%			

Oximetry −: negative or intermediate oximetry; NPV: negative predictive value of the absence of symptoms.

rhinolalia clausa, and frequent rhinitis. Two data from the questionnaire appeared to be highly significant. The absence of any type of nocturnal snoring and the existence of snoring exclusively related to rhinitis were closely correlated with a negative oximetry recording (negative predictive value [NPV], 100%). The ''frequent rhinitis'' factor was the most closely correlated with oximetry. On the other hand, the absence of the notion of ''apnea'' noted during sleep and recurrent rhinitis was quite strongly predictive of normal oximetry (NPV, 81%). Overall, the specificity and sensitivity of the other parameters studied were low.

Tables 2—4 provide the details of the relations found between oximetry and the main symptoms indicated by the parents. Table 5 provides a summary of all the results obtained.

Discussion

Although SDB has a clinical definition, today only PSG can rigorously assess its severity. Questioning the family of the child with ATH and suspicion SDB is not sufficient [10]. However, systematic PSG for every child is not possible because this sleep laboratory examination is costly and unfortunately its access is difficult in many regions.

Yet complementary exams are contributive for several reasons in cases of suspected SDB in children because:

- they provide information arguing in favor of traditional hospitalization when they show significant abnormalities that may aggravate SDB in the immediate postoperative period;
- in identifying the most severe cases of OSAS, they can aid in prioritizing patients on a waiting list when the current delays to adenotonsillectomy can reach several months;
- most importantly, they reinforce the surgical indication as soon as the results are abnormal. This aspect presents two clear advantages:
  - the contribution of arguments providing reinforced information for the patients' families (the validity of the intervention is objectively demonstrated),
  - prevention of complaints or medicolegal assistance in case of a serious undesirable postoperative event.

Among the alternatives to PSG proposed, nocturnal oximetry — the subject of the present study — is the most accessible and the least expensive. However, its main drawback is its lack of sensitivity. Performed alone or after screening using a simple questionnaire in a non-selected population, it does not contribute sufficient information, as underscored by several studies [12,13]. Our results are in agreement with these data: despite the constant presence of ATH and the high prevalence of symptoms suggestive of SDB (209 children who were permanent snorers out of 342 [61.1%]), only 21.9% of the children tested had a positive, i.e., abnormal, oximetry recording.

Oximetry's specificity is good provided that rigorous analysis technique is respected with manual reading of each recording, which required a mean of 30 min per patient in the present study. Optimal precision in the search for and elimination of the artifactual parts of the recording is crucial so as not to overestimate the existence of positive oximetry recordings [14].

The present study, conducted in a population of children referred by their ENT physician for suspected SDB, is original on many counts. Contrary to other series reported in the literature including the study of oximetry in children recruited for their symptoms or functional signs, this study investigated children selected for their ATH, therefore those seen

Table 5 Summary of nocturnal oximetry data and symptoms (comparison of presence and absence of sign or symptom).

Symptoms	Oximetry + (*n*)	Oximetry − and intermediate (*n*)	PPV (%)	NPV (%)	Sensitivity (%)	*P*	Odds ratio (95% CI)
All types of snoring (Gr. I + II + III)	75	249	23.1	100	100	< 0.02	∞ (1.98—∞)
Permanent snoring (Gr. I)	66	143	46.15	93.2	88	< 0.0001	8.09 (3.74—17.43)
Intermittent snoring (Gr. II + III)	9	106	31.6	92.2	88	< 0.001	7.97 (3.68—17.17)
Snoring with rhinitis (Gr. III)	3	44	6.4	75.6	4	0.0055	0.21 (0.06—0.7)
Apneas	64	182	26	90.4	90.1	0.003	4.4 (1.88—10.27)
Recurrent rhinitis	60	151	28.4	88.5	80	0.0002	3.76 (1.95—7.24)
Mouth breathing	66	209	24	86.6	88	NS	2.04 (0.96—4.34)
Thinness	23	59	45.1	66.7	30.7	NS	NS
Failure to thrive	16	38	45.7	65.7	21.3	NS	NS
Parents snorers	49	161	38.3	65.1	68	NS	NS
Daytime sleepiness	23	56	53.1	67.1	30.7	NS	NS

NPV: negative predictive value; PPV: positive predictive value; NS: not significant.

by an otolaryngologist. The results of comparing symptoms and oximetry data provide the following data. There were significantly more positive oximetry readings in the group of permanent snorers (group I) than in each of the other groups, notably compared to occasional snorers (groups II and III), and, unmistakably, in the groups of snorers compared to the non-snorers (group IV). In particular, the negative predictive value of the absence of snoring was 100% for oximetry. The sensitivity of snoring was 100% in snorers, whereas its specificity was only 6.7%. The results of the positive oximetry were significant for the ''apneic'' children and for those suffering from frequent rhinitis compared to those who did not have the same signs and symptoms (Table 5). This underlines the existence in this population of a relation between positive oximetry and apnea, rhinitis, or obstructive rhinopathy, even if the specificity of this test is not high with regard to the parental observations. Among the occasional snorers, there was also a difference between the children suffering from snoring during rhinitis (group III) and the others (group II), but in the opposite direction (significantly fewer positive oximetry recordings in group III). We assume that group III patients had fewer episodes of rhinitis. This observation suggests that the rhinitis factor should be taken into account differently in terms of inducing SDB, depending on whether it is expressed by the parents as a chronic phenomenon (high incidence of SDB) or intermittent (low incidence of SDB, group III), in children suffering from ATH.

Role of the questionnaire

The problem of the questionnaire's usefulness has not yet been concluded [15]. Yet tonsillar hypertrophy alone cannot justify surgery without precise information on the nature and quality of the child's sleep. It should be remembered that this type of adenotonsillectomy is a procedure that is subject to risk (postoperative hemorrhage in a mean 3% of cases; one death in 40,000 tonsillectomies). The surgical indication should therefore be warranted. Questioning the family is a first lead to investigate. The interview should be rigorously conducted with no suggestion or pressure on the parents, but they should be informed beforehand of the signs to note, as in our study. When the results are normal, it seems prudent to check that the child's sleep has actually been observed.

However, even if it has been shown to be correlated with oximetry in this population, it is highly probable that a single negative questionnaire does not ensure durable absence of SDB. At least clinical follow-up of these cases is advised. When parents have observed constant snoring or apnea episodes in this population, oximetry was positive in less than one case out of four only (predictive value, 23.1%). When it was not, a polysomnograph is strongly recommended in these children. If locally this examination is impossible or long and difficult, regular clinical monitoring by parents and the otolaryngologist should be carried out. The situation can always evolve toward deterioration, which requires occasional monitoring at night spread over several months (one to three times a week, for example), outside of periods with rhinitis or laryngitis. This clinical and parental monitoring seems warranted because SDB risks interfere with psychocognitive and cardiovascular development for the most part related to repeated hypoxia [16,17]. However, there are no arguments in favor of these episodes occurring frequently and having significant clinical repercussions in absence of positive oximetry findings. Moreover, the consequences of increased resistance of the airway and sleep fragmentation, in particular in the psychocognitive domain, are controversial and may be modest [18—20]. All in all, the questionnaire used in this study proved that negative results correlated with negative oximetry, which is therefore pointless. In light of these results, it seems appropriate to not propose oximetry in cases of absence of snoring

(and apnea) reported by parents, assuming the monitoring mentioned above is carried out. However, in children who snore with an intermediate or negative oximetry, the PSG remains the reference exam. While waiting for this exam, parental monitoring of the child's sleep as well as rigorous surveillance of daytime vigilance, growth, and academic performance, depending on the child's age, are advised [8,21]. If daytime or nighttime symptoms appear, surgical treatment of ATH can be discussed on a case-by-case basis.

Role of oximetry recording

With a child whose family reports moderate daytime or nighttime symptoms, what is the contribution of nocturnal oximetry? Patiently analyzed as in this study, it is confirmed that this exam has no ''pathological'' value because, in all the other cases, it does not confirm the absence of SBD. However, if it is positive, it is a decision-making aid in view of surgery, which can be rapidly but calmly planned to the maximum benefit of the child. Moreover, whatever the results of the questionnaire, if it is positive it seems clearly predictive of the severity of the obstruction, a criterion that determines the immediate postoperative prognosis. Many studies have shown the risk of morbidity (and sometimes mortality) associated with severe obstructive sleep apnea [21—23]. Without PSG, oximetry can confirm the existence of a severe form of SBD with desaturations (SpO_2). Pre- and particularly postoperative monitoring can thus be prepared for adenotonsillectomy. It has been shown that children suffering from chronic nighttime intermittent desaturation with periods below 80% SpO_2 have increased sensitivity to opiates and can easily encounter postoperative respiratory complications [5]. Hospitalization with cardiorespiratory monitoring for the first night is strongly recommended. Finally, obtaining positive oximetry can make it possible to explain the expected benefits of surgery to the patient, the family, and the family physician in optimal conditions, or even convince the ENT surgeon of the validity of this pharyngeal surgery if there is initial reticence, notably if the child is quite young.

Saito et al. attempted to verify whether oximetry could specify the indications for adenotonsillectomy in selected children with snoring and apnea, compared to a group of control children [14]. They observed that there were more positive oximetry results in children who were snorers and the poorer the initial results of oximetry, the more the children benefited from the surgery. They also considered that polysomnography was not necessary in cases with positive oximetry (43.5% of the children tested) and therefore substantial savings could be made by the local healthcare system. In this series of children with snoring and apnea, we found double the incidence of positive oximetry compared to the children in our study who were selected based on adenotonsillar volume.

Finally, although oximetry can demonstrate true apneas with multiple episodes of desaturation, it cannot diagnose upper airway resistance syndrome. Useful only if it is positive, oximetry seems valuable as a first-line examination. In the present study, it allowed us to forgo PSG in 21.9% of the patients (75/343) in whom it was positive, a savings of a few hundred euros per child, which makes room in the sleep laboratory for children whose management is clinically more difficult: those whose oximetry results are negative or intermediate.

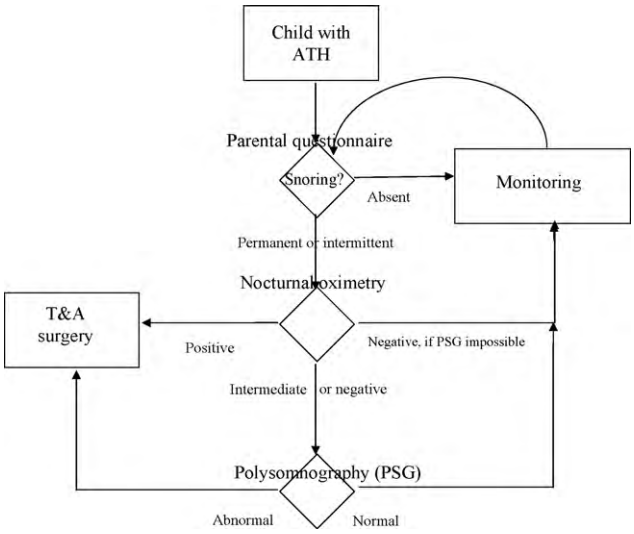

Figure 1 Proposal for a decisional tree for a child suffering from adenotonsillar hypertrophy (ATH).

Use of questionnaire + oximetry

The importance of correlating the questionnaire and oximetry should be highlighted because questioning parents, often done first, was shown to be clearly correlated with nocturnal oximetry in cases where there is no snoring or apnea. In these conditions, oximetry seems pointless. In cases of snoring without precise details, oximetry can identify cases of severe SDB at a lower cost and thereby preclude an expensive polysomnographic exam that has no benefit for these children.

In certain locations and depending on the contingencies of the healthcare system, this process can also be used to prioritize patients on a waiting list for adenotonsillectomy according to the severity of the patients' condition [24]. It should be remembered that the most seriously affected children should also be monitored closely after surgery over the intermediate term [25].

The limitations of this study are related to the principle of oximetry itself as well as the absence of diagnostic and therapeutic follow-up (but the latter point will be treated in a later publication). Oximetry interpreted according to Brouillette et al. [10] can only detect the most seriously affected children. This leaves a vast zone of pathological uncertainty — negative and intermediate oximetry results — in which this exam is not contributive. A decision tree aiming to optimize the diagnostic workup of children suffering from ATH is proposed in Fig. 1.

Conclusion

This study suggests that nocturnal oximetry combined with data from a parental questionnaire can be useful in the diagnostic workup of children with ATH. In presence of snoring or

apneas, oximetry was positive in nearly one-quarter of the cases, thus obviating the need for polysomnography, whose access is often difficult. In absence of these symptoms, use of oximetry was not demonstrated to be useful. In children who are snorers, positive oximetry has made it possible to better inform families by providing an objective document in favor of adenotonsillectomy. Although it does not equal the reference exam (PSG), it seems able to screen the most severe cases of SDB and thus aid in prioritizing patients on a waiting list for surgery.

Conflict of interest

The authors declare no conflict of interest.

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