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Original Investigations

Children and Nocturnal Snoring: Evaluation of the Effects of Sleep Related Respiratory Resistive Load and Daytime Functioning

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Abstract. Twenty-five children, age range 2 to 14 years (mean age = 7), were referred to the Stanford University Sleep Disorders Clinic for various clinical symptoms, including excessive daytime somnolence, heavy nocturnal snoring, and abnormal daytime behavior. All children (10 girls and 15 boys) were polygraphically monitored during sleep. No sleep apnea syndrome or oxygen desaturation was revealed. However, each child presented significant respiratory resistive load during sleep associated with electrocardiographic R-R interval and endoesophageal pressure swings. The most laborious breathing occurred during REM sleep. Second degree atrioventricular blocks were also noted. Tonsillectomy and/or adenoidectomy was performed in every case and resulted in a complete disappearance or substantial amelioration of the reported symptoms. Objective evaluation by Multiple Sleep Latency Test and Wilkinson Addition Test confirmed the beneficial effect of surgery.

Key words: Snoring - Respiratory resistive load - Tonsillectomy - Sleep - 24 h Holter ECG

Introduction

Over the past decade we have seen children referred to us with increasing frequency for a common set of symptoms which have become progressively more severe despite regular medical consultations and therapeutic trials. Given the size of the patient population seen with these symptoms, it seems appropriate to draw the attention of specialists to the impact of various breathing changes during sleep on daytime somatic and psychological symptoms and on heart function during sleep.

This report will focus on 25 children selected for this report because we were able to obtain sufficient collaboration from parents, pediatricians, and educators to allow for long term follow up.

Patient Population and Cause of Referral

Between 1973 and 1979, 25 children referred to the Stanford University Sleep Disorders Clinic for evaluation were studied before and after tonsillectomy and /or adenoidectomy. There were 10 girls and 15 boys, age range 2 to 14 years (mean age = 7); 11 were between 2 and 6 years of age, 9 between 6 and 11, and 5 between 11 and 14. All children (including the oldest group) were prepubertal and were classified, after evaluation for secondary sexual characteristics, at Tanner stage 1 [11]. Twenty children were Caucasian, 4 were Black, and 1 was Asian.

The causes of the patients' referral varied over the years. Some children were referred by psychiatrists or neurologists because of excessive daytime sleepiness. School teachers also often initiated the consultation because they noted abnormal behavior, such as noticeable somnolence in the classroom. Most of the children with these reported symptoms were between 7 and 14 years of age. We also received referrals of younger children for sleep apnea syndrome [5, 6] screening because of heavy nocturnal snoring. Oto-laryngologists, pediatric allergists, and pulmonary medicine specialists were the primary sources ot these referrals.

Clinical Symptoms

Although daytime somnolence and suspicion of sleep apnea syndrome [5, 6] led to consultation with our subspecialty clinic, all subjects also presented other clinical symptoms for which they had been receiving medical attention for a minimum of 12 months prior to referral.

Table 1A indicates symptoms which led to initial pediatric consultation. In 8 (out of twelve) cases, hyperactivity had led to prescription of methylphenidate. Aggressive and rebellious behavior or social withdrawal and pathological shyness were believed to impair the well-being of 20 children and to interfere with their performance at school. Nineteen of these 20 children had had psychiatric and/or psychological evaluations. Fifteen children were, or had been, followed by a psychiatrist or clinical psychologist during the 12 months prior to referral. All school age (over 7) children had been placed in "special education" programs (children are referred to these programs when, in the opinion of teachers in the state school system, they are unable to pursue a normal curriculum and need specialized attention).

Despite professional efforts, none of the children had shown any symptomatic improvement during the semester preceeding the initial sleep clinic consultation. Six children presented with a clear history of respiratory allergy for which intermittent seasonal treatment had been given. Our initial interviews revealed the nocturnal symptoms outlined in Table 1B. Most of these symptoms had existed more than 24 months

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Table 1

A. Clinical Symptoms

- Excessive daytime somnolence 32% (8 patients)
- Hyperactivity 48% (12 patients)
- Aggressive and rebellious behavior 40% (10 patients)
- Pathological shyness and social withdrawal 40% (10 patients)
- Learning problems (and/or question of learning disability) * 40% (10 patients)
- Report of clumsiness and/or "lack of coordination" 44% (11 patients)
- Lack of appetite and problems with food intake 64% (16 patients)
- Frequent morning headaches [1] 24% (6 patients)
- Frequent nausea with or without vomiting 24% (6 patients)
- Frequent upper airway infections 48% (12 patients)

B. Noctural Symptoms

- Heavy snoring at night 100% (25 patients)
- Unusually profuse nocturnal sweating ** 84% (21 patients)
- Special sleeping position ** 40% (10 patients)
- Restless sleep and abnormal movements ** 64% (16 patients)
- Enuresis 64% (16 patients)

* Only children 7 years and older have been included here–this represents 71% of the surveyed population.
[1] A bias may exist, as young children do not frequently report headaches.
** These percentages may be underestimations, because parents did not always check children, especially older children, during sleep.

prior to consultation, but had not attracted the attention of the medical teams involved in the care of the child.

Clinical Examination

At examination, 18 patients (76%) were underweight or underdeveloped (height and weight). As mentioned previously, the four children aged 11, 12, 12, and 14 years were still at Tanner stage 1 [11] for secondary sexual characteristics. The 14 year old was a black girl who presented complete absence of secondary sexual characteristics and a general underdevelopment. (Permission to perform sleep-related endocrine research studies was not granted in this case.)

Four children between 4 and 8 years of age presented significant limb hypotonia. In three cases, this was more prominent in the upper limbs and involved major shoulder hypotonia. The fourth child had generalized hypotonia which previously had led to several electromyographic studies (case 4). Three of these four children had had muscle biopsies at the request of a neurologist prior to referral to us. Only one biopsy (case 4, a six year old boy) was read as "unusual pattern–probably abnormal"; no clear diagnoses were obtained, despite in-depth histological and electron microscopic analyses.

As part of the sleep clinic evaluation, all children were seen at the oto-laryngology clinic; all presented enlarged tonsils and/or adenoids, the enlargement ranging from borderline to significant at visual examination (scored from 2+ to 4+). Soft tissue X-rays of the neck confirmed lymphoid tissue hyperplasia and the clinical "score" 2+ to 4+.

Nocturnal Evaluation and Monitoring

All children were monitored polygraphically in a quiet awake supine state and during nocturnal sleep. Variables recorded included electroencephalogram (EEG) C3/A2–C4/A1 of the international placement system; electro-oculogram (EOG); chin electromyogram (EMG); electrocardiogram (12 lead ECG while awake, lead II ECG while asleep, and a 24 h Holter ambulatory ECG); respiratory effort monitored by abdominal and thoracic strain gauges; and airflow measured by nasal and buccal thermistors. In 64% of cases, expired CO_2 concentration was measured during sleep using an LB1/CO_2 or LB2/CO_2 Beckman analyzer. Oxygen saturation was measured by different means. At first, a Waters Instruments ear oximeter was used. More recently, depending on the size of the child's ear, a Hewlett Packard ear oximeter or transcutaneous PO_2 electrode was used. The most recent studies used a finger oximeter not commercially available at this time (Corning, New York).

An endoesphageal pressure transducer or balloon monitored endoesophageal pressure during sleep in 20 (80%) of the children. Endoesophageal pressure may vary during sleep, particularly with different sleep states. A slight increase frequently occurs during REM sleep, and slight pressure irregularities (both increases and decreases) also may be seen. Decreases usually are associated with bursts of rapid eye movement.

Controls

Twenty-five children, whose ages ranged from 2 to 14 years, and who did not present snoring or obstructive sleep apnea syndrome, were monitored during the same period in the same laboratory as controls. This control group was not exactly age matched, but the age range was similar; the greatest age difference between a patient and a control was 12 months.

Daytime Study

The 5 children whose age ranged between 11 and 14 years of age underwent daytime testing following a protocol extensively used in our clinic over the past 5 years in normal children of a similar age group. Children were polygraphically monitored during night 1 from 2200 (lights out time) until 0700. Total dark time was 9 h. Between 0700 and 0900 of day 2 children had standard activities (toilet, breakfast, etc). At 0845 EEG, EOG, and chin EMG electrodes were checked and children were reinstructed about the daytime testing. Each was seated in a comfortable chair at a table, with a series of additions derived from the Wilkinson Addition Test (WAT) [14] and an electrical switch in front of them. They were instructed to perform additions and to press the switch at the end of each problem. Continuous polygraphic recording occurred during the testing. At the end of the test, each child was requested to lie down with eyes closed in a bed in a quiet, dark room, and they were polygraphically monitored during the following 20 min, as described in the, by now, classical multiple sleep latency test (MSLT) [3]. This sequence of testing was repeated at 1130, 1330, 1530 and 1730 h with a total of 5 testing sessions during the daytime. During the WAT, if polygraphic evidence of sleep was noted (sleep being defined as three continuous minutes of stage I or II Non Rapid Eye Movement (NREM) sleep, the child was stimulated by calling his/her name over an intercom and entering the room. Test results were compared to those of control children with similar pubertal staging. After evaluation, we recommended adenoidectomy and/or tonsillectomy; this recommendation was followed in all 25 cases reported here.

Follow-up and Retesting Procedures

The following procedures were outlined before surgery was performed. We indicated to the parents that we considered Nr. 1 to be normal medical care, but that further follow up was for research project purposes. The informed consent form included a statement that refusal to participate in, or early termination of, the follow-up protocol would not in any way jeopardize the care of the child. The follow-up protocol steps were:

1. Systematic post-surgery polygraphic monitoring, always including respiration and heart measurements, performed no earlier than 4 weeks and no later than 12 weeks post-surgery. When possible, a second polygraphic monitoring was performed one year post-surgery, and, whenever permitted, we monitored patients more frequently. The later follow-up recordings did not use invasive techniques (i.e., no esophageal balloon was used).
2. Complete re-evaluation of clinical symptoms at the time of monitoring and at 3, 6, and 12 months post-surgery.
3. Contact with the referring pediatrician independent of the patient's sleep-clinic visit.
4. When parental approval was given, contact with the child's school teacher during the first post-surgery year.
5. Re-evaluation over the years of children reported here, and ongoing telephone contact with referring pediatricians and related health personnel involved in the child's care for as long as possible.

The 5 children who underwent daytime testing before surgery were restudied under a similar protocol 3 months post-surgery with, after one night of polygraphically monitored sleep, MSLTs and WATs.

Results of Nocturnal Evaluation and Monitoring Before Surgery

A) *Breathing During Sleep:* All children snored heavily throughout the night. However, no evidence of obstructive sleep apnea as defined in the literature [5, 6, 12] was noted. While in the quiet awake supine position, the children's respiratory rate always was under 20 breaths per min. At sleep onset, a pronounced tachypnea was seen in 23 of the 25 children. Eighteen were restless sleepers; they tossed and turned frequently and often slept in an unusual position with the neck hyperextended.

A significant increase in endoesophageal pressure without concomitant drop in O_2 saturation was noted in all cases during sleep. In control subjects, the range of the lowest negative pressure during quiet wakefulness was -8 to -13 cm H_2O; during sleep, it was -11 to -20 cm H_2O. In patients, the range of lowest negative pressure during quiet wakefulness overlapped with controls; during sleep, it was -30 to -53 cm H_2O. The lowest negative pressures were always recorded during REM sleep in both groups. (The differences between groups were statistically significant at $P<.01$ during NREM and at $P<.005$ during REM sleep, two tailed t-test.) Laborious breathing was observed during all sleep stages. At times, a transient drop in O_2 saturation was noted during REM sleep in association with bursts of rapid eye movements (see Fig. 1). Some patients experienced sleep disturbances associated with the tossing and turning, but sleep fragmentation as is seen in obstructive sleep apnea [4] was not noted.

B) *Sleep Monitoring:* Nocturnal total sleep time (TST) was not significantly increased compared to controls, despite "ad libitum" sleep being permitted. Patients had a normal percentage of REM sleep as compared to controls. Similar amounts of stage 3–4 NREM (delta wave) sleep occurred in both groups, or frequently (in 15 children or 60%) patients showed a non-significant trend toward increased stage 3–4 NREM sleep.

C) *Heart Monitoring:* A 24 h ambulatory Holter ECG showed some cardiac arrhythmias during sleep (tachy-bradycardia and 1st and 2nd degree atrioventricular block) in 10 children. Computer plotting of the R-R interval revealed a significant change during sleep as compared to wakefulness (see Figs. 2 and 3)

D) *Other Observations:* Hyperhidrosis during the nocturnal recording was noted in all patients.

Daytime Test Results

All five children slept a minimum of 478 minutes (mean 499) the night preceding daytime testing. They all fell asleep during MSLT. Their mean MSLT scores (see Table 2A) differed significantly from Tanner stage 1 control children ($P<.001$). The WAT was analysed in two ways: the number of problems resolved during each test and the number of errors noted during each test. As each child was retested post-surgery within 4 months of the initial testing, comparison was made with each child as his own control. Results of pre- versus post-surgery testing are given below.

Post-surgery Results

The 25 children reported here were, by definition, followed for at least 12 months post-surgery.

Clinical Symptoms: In all cases, the reported symptomatology either disappeared or was ameliorated greatly. School teachers and parents reported at the 3 month follow-up evaluation that the children had demonstrated noticeable behavioural changes and that school performance had improved dramatically. By the 6 month follow-up visit, none of the children were taking previously prescribed methylphenidate or pemoline. Of all symptoms, enuresis was least affected by surgery; of the 16 patients presenting pre-surgery enuretic episodes, 8 continued to be enuretic at one year post-surgery. Only two children remained in special education classes; the other children had returned to regular school programs.

Examination: Height and weight changed dramatically post-surgery. All children had reached at least the 75th percentile for height and weight within one year after surgery. These gains, as compared to pre-surgery values, were significant at $P<.005$, one tailed t-test. The four children who had been noted to have muscle hypotonia demonstrated clear improvement, although hypotonia was still noticeable in the child whose biopsy had been judged "unusual" (case 4). The 14 year old black girl who had been grossly underweight and who had demonstrated no sexual maturation pre-surgery reached Tanner stage III before the end of the post-surgery year. (All control girls age 12 and over were at least at Tanner stage II [11].

Monitoring: Sleep was much quieter for all children, with complete disappearance of the restlessness, snoring, and profuse sweating noted pre-surgery. The sleep-related tachypnea had disappeared completely (see Figs. 4A and B), and neither the R-R interval swings nor the 2nd degree atrioventricular block associated with REM sleep were found on the post-surgery 24 h Holter ECG. Abnormal endoesophageal pressure

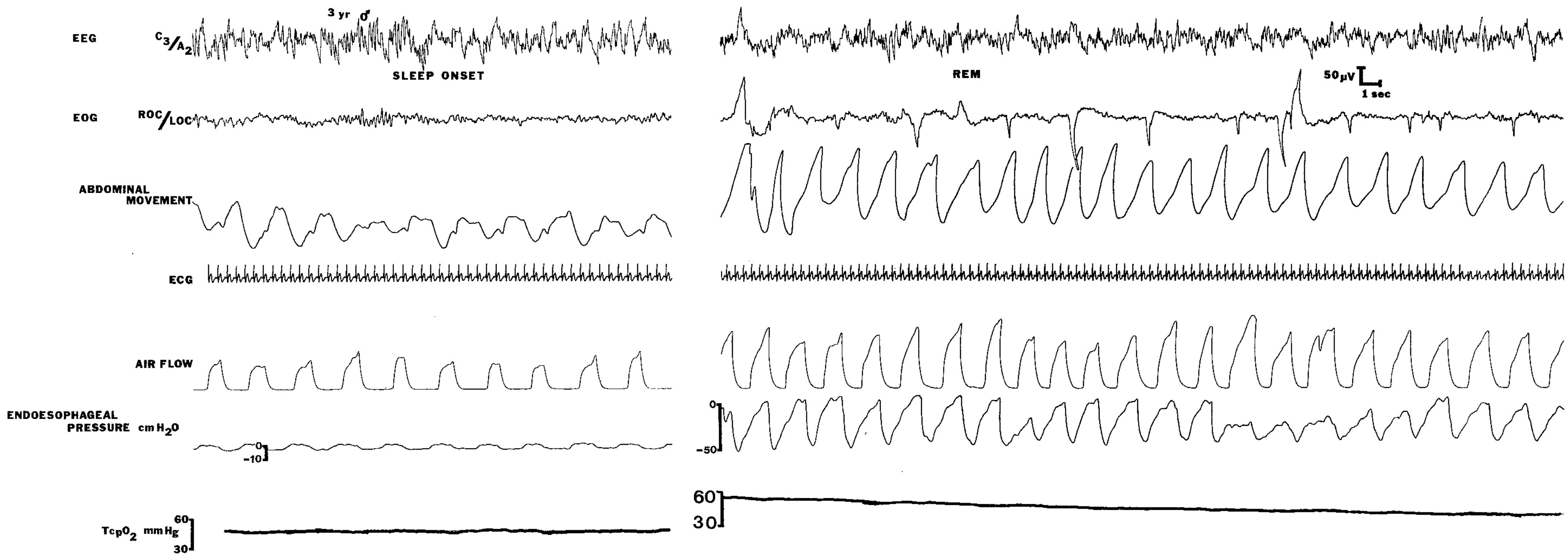

Fig. 1. Monitoring during sleep performed in a 3 year old boy. From top to bottom:

Channal 1 = Electroencephalogram, C3/A2 from the 10–20 international placement system
Channal 2 = Electro-oculogram
Channal 3 = Abdominal movement, monitored by strain gauge
Channal 4 = Electrocardiogram (lead II)
Channal 5 = Air flow, monitored by 3 thermistors placed in front of nostrils and mouth
Channal 6 = Endoesphageal pressure, monitored by endoesophageal balloon
Channal 7 = Transcutaneous oxygen electrode monitoring (Litton C)

Left side = sleep onset, *Right side* = rapid eye movement (REM) sleep near 0400

Note: The transutaneous oxygen electrode had been relocated less than 10 minutes before the presented sample of the recording on the right side. Values indicated may not be completely accurate, as an adaptation period is needed to reach specific skin temperature needed for correct measurement. The figure shows an increase in endoesophageal pressure during sleep. Variations of pressure are seen particularly in association with REM during the segment of REM sleep presented

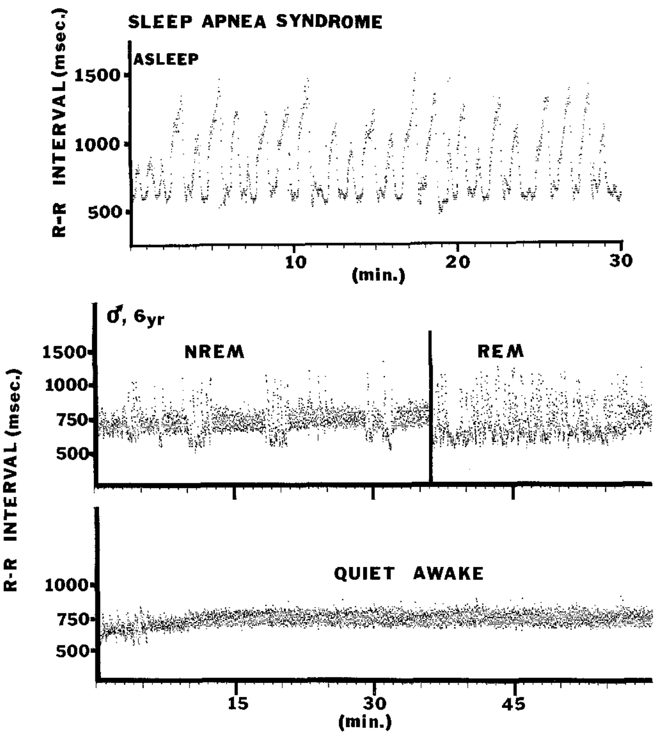

Fig. 2. Computer plottings of the R-R interval obtained from a Holter ECG monitoring. The top of the figure gives an example of the type of R-R interval variations seen during obstructive sleep apnea. One can see clearly the progressive bradycardia which occurs during the apneic event, and the abrupt tachycardia occurring with resumption of ventilation. When the Holter ECG is used as a screening device for sleep apnea syndrome, the peak to peak measurement between each tachycardia "spike" is used to approximate the duration of the sleep apnea. The two bottom tracings show the changes noted in our children with respiratory load but no apnea. During quiet wakefulness, (supine, just before sleep onset), the R-R intervals are well grouped and very regular. During Non REM sleep (when R-R intervals are most regular in normal humans) and during REM sleep, one can see that repetitive changes in heart rate occur. These are related to heavy (inspiratory) snoring. The autonomic nervous system changes which occur with Non REM sleep and REM sleep, and the inspiratory resistance increase–which probably lead to a very incomplete Mueller maneuver–most likely are factors in the R-R interval changes. Once again, 24 h Holter ECG monitoring may be used as a screening device when a breathing abnormality during sleep, such as the one outlined here, is suspected clinically

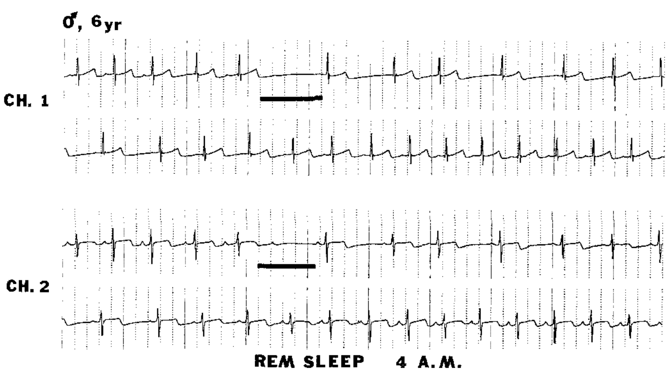

Fig. 3. Recording of the same subject, a 6 year old boy, as shown in Fig. 2. This is a computer print-out of the 24 h Holter ECG segment monitored at 0400 during REM sleep. Note the existence of 2nd degree atrioventricular block in association with very heavy snoring and laborious breathing. Normal oxygen saturation is, however, maintained. In this subject, 2nd degree atrioventricular block was noted in association with each REM period during the monitored night

Table 2. Multiple sleep latency test scores

A. Pre-surgery

Naps	Case 1	Case 2	Case 3	Case 4	Case 5
Nr. 1	12	11	12	9	12
Nr. 2	8	7	7	6	8
Nr. 3	10	12	10	8	9
Nr. 4	10	10	9	10	15
Nr. 5	13	12	11	9	11
$\bar{X}$	10.6	10.4	9.8	8.2	11

B. Post-surgery

Naps	Case 1	Case 2	Case 3	Case 4	Case 5
Nr. 1	20	20	20	20	20
Nr. 2	18	15	19	16	20
Nr. 3	17	20	20	20	20
Nr. 4	20	20	17	16	20
Nr. 5	20	18	20	20	20
$\bar{X}$	19	18.6	19.2	18.5	20

Table 3. WAT: Number of problems accurately solved

A Pre-surgery

Tests	Case 1	Case 2	Case 3	Case 4	Case 5
Nr. 1	12	11	8	4	9
Nr. 2	5	9	8	3	6
Nr. 3	6	7	8	4	9
Nr. 4	4	4	5	5	6
Nr. 5	4	6	6	4	6
$\bar{X}$	6.2	7.4	7.0	4	7.2

B. Post-surgery

Tests	Case 1	Case 2	Case 3	Case 4	Case 5
Nr. 1	12	16	17	9	10
Nr. 2	14	12	17	11	10
Nr. 3	15	14	14	11	12
Nr. 4	12	15	17	12	11
Nr. 5	14	15	17	9	10
$\bar{X}$	15.4	14.2	16.4	10.4	10.6

swings had ceased. All of these changes were noted at the time of the first post-surgical nocturnal recording (4 to 12 weeks after surgery).

Repeat Daytime Testing

As can be seen in Table 2, the MSLT scores were very significantly improved post-surgery. None of the children changed Tanner staging in the three month post-surgical period, but the second set of MSLT scores is within the range of Tanner stage I control children [4]. The WAT results show that after surgery the number of problems tabulated increased by a mean of 35% (range 30–40%). The number of problems solved with accurate results increased by a mean of 48% (range 32–57.5%).

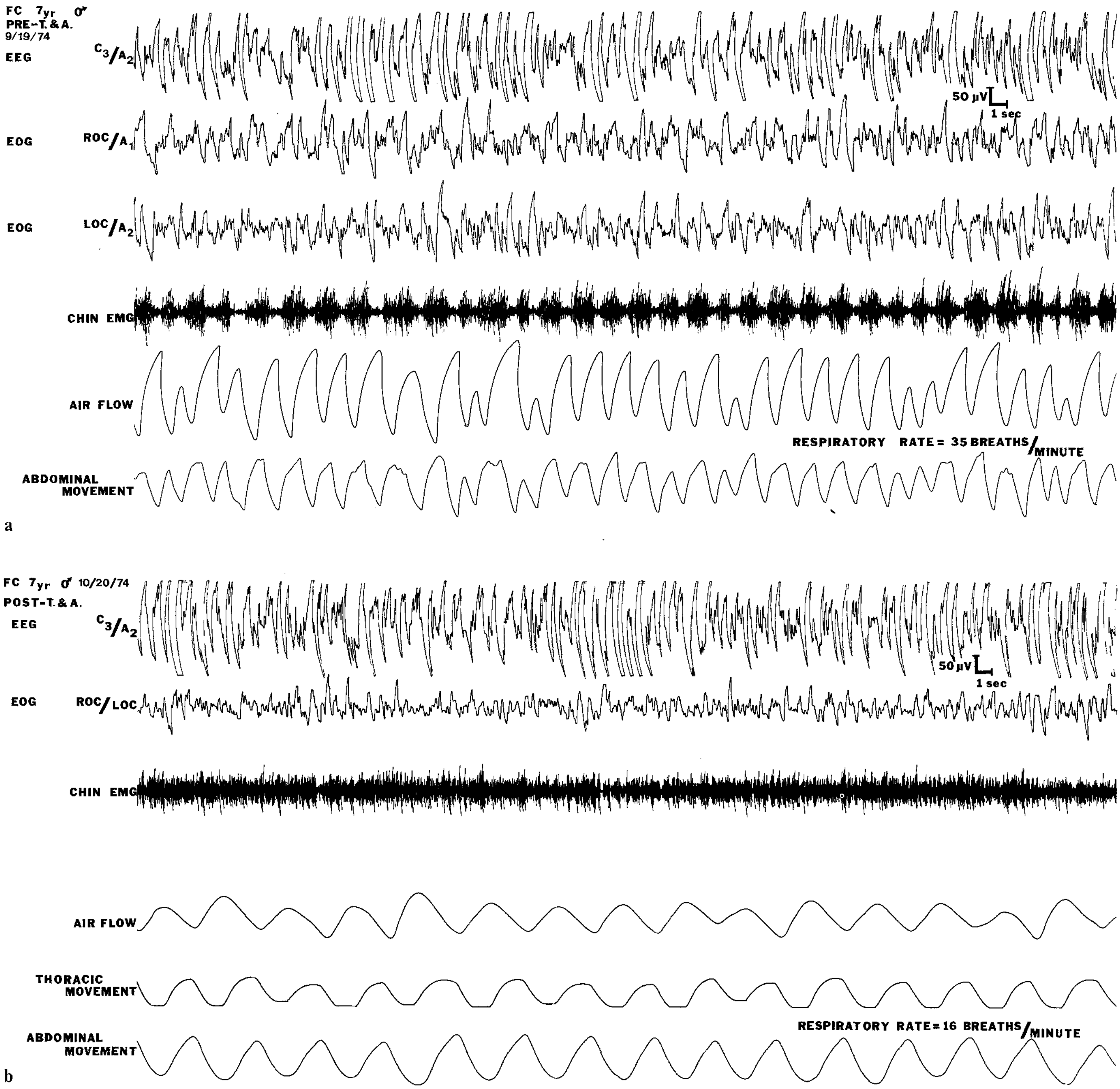

Fig. 4A and **B.** Examples of sleep monitorings of a 7 year old boy during stage 4 Non REM sleep just before and one month after T and A. From top to bottom:
Channel 1 = Electroencephalogram, C3/A2 from the 10–20 international placement system
Channel 2 = Electro-oculogram
Channel 3 = Electro-oculogram
Channel 4 = Chin electromyogram
Channel 5 = Air flow, monitored by 3 thermistors (nasal and buccal)
Channel 6 = Abdominal movement, monitored by strain gauges
Note: Fig. 4B presents only one EOG channel and an extra chest movement channel
Snoring may be noted easily on channel 4 (chin electromyogram) in Fig. 4A, and has completely disappeared in Fig. 4B. The sleep EEG is identical in both cases

Normally one expects to see a trend toward better results in the WAT performed later during the day due to a possible "learning effect" and habituation to the testing condition. However, before surgery analysis of the number of additions without errors performed on each WAT throughout the day indicates that a constant, clear decrease in performance occurs from early morning test to late afternoon test in all patients. The mean number of problems accurately solved was 9.2 during test 1 in the early morning and 5.2 during the last session starting at 1700. Post-surgery, on the other hand, there were no significant differences between the first and last test. (See Table 3)

Discussion

All children presented here were heavy snorers at night. None of them, however, had complete airway obstruction as seen in obstructive sleep apnea syndrome. All of them had an enlargement of the pharyngeal lymphoid tissues. Previous reports have indicated that failure to thrive and development of Cor pulmonale may result [see reviews in 7]. Twenty years ago tonsillectomies and/or adenoidectomies were performed nearly routinely, at times with little clinical justification. Since then a more conservative attitude has developed, and surgery is not recommended unless clearly justified clinically. However, until now there has been little, if any, systematic evaluation of the effect of a narrowed airway on cardiorespiratory functions during sleep, on sleep itself, and on the daytime functioning of children whose sleep is disturbed significantly on a nightly basis. Evaluation of the effect of enlarged tonsils and adenoids on breathing during sleep may prove an important guide to the treatment of specific cases. Enlargement of lymphoid tissue may not be sufficient to lead to a significant respiratory resistive load during wakefulness but, during sleep, may have a significant impact. Several factors explain why sleep can exacerbate problems to levels leading to daytime impairment.

The fact that humans sleep supine in itself affects respiration (see Sullivan [10] for review). The decrease in muscle tone associated with sleep onset, which leads to an increase in airway resistance [8], and the atonia of intercostal and respiratory accessory muscles during REM sleep are physiological events which recently have been well documented as increasing diaphragmatic workload [1, 2]; this was confirmed by the increase in endoesophageal pressure monitored during REM sleep in our control population.

In children with enlarged lymphoid tissue, a further reduction of the naso-pharynx occurs secondary to normal sleep mechanisms. Despite the fact that no complete airway obstruction occurs, and that diaphragmatic movement can maintain normal oxygenation, we believe that this abnormal narrowing of the airway has an impact on nocturnal airflow, with increased respiratory resistive load, and is therefore responsible for the increased endoesophageal pressure swings monitored in these children. An incomplete Mueller maneuver (inverse Valsalva) can be postulated to explain the noted cardiac arrhythmias [13].

Specific testing with WAT demonstrated that in a subgroup of five school age—Tanner stage I—children with school problems and complaints of daytime fatigue and sleepiness, objective testing can document the complaint. Comparing pre- and post-surgery WAT scores, we have also been able to demonstrate significant improvement of a performance decrement.

Finally, we wish to emphasize that the plotting of the R-R interval obtained from a 24 h Holter ECG can provide inexpensive screening information about a patient's breathing during sleep; this may be a practical approach since long and specialized respiratory recordings during sleep are costly and more difficult to arrange.

In conclusion, the "cost" of breathing during sleep for a child presenting with regular, heavy snoring is a variable which should be evaluated during sleep by appropriate techniques. Daytime behavior and school performances can be adversely affected by nighttime life and poor quality of sleep.

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