CASE HISTORY

The patient is a 56-year-old male who complained about snoring and apneas during sleep. He was obese without further abnormalities. He did not use alcohol and denied smoking. During a 48-hour polygraphy a severe sleep apnea syndrome was found.

Although the patient did not complain about excessive daytime sleepiness he frequently napped during the day. He had a polysomnography in our clinical sleep unit in order to complete the diagnostic procedures and to try out nasal CPAP. The hypnogram and other polygraphic features of this night are shown in fig. 1.

Hypnogram and other polygraphic features of the patient mentioned in the case history: sleep apneas during REM sleep when the patient is lying on his back. Successful treatment with nasal CPAP.

Severe periodic limb movements after CPAP was given.
Sleep onset: 10 minutes; REM sleep 9% with REM onset after 2h 32 min.; 31% slow wave sleep; sleep efficiency 69%. 54 apneas, mainly during REM sleep, with a duration of 28-54 sec.
Movement index after CPAP has been given: 46 per hour
In the first part of the night the patient slept reasonably well, except for a period of about 20 minutes in which he was awake. No apneas occurred. They were seen only in REM sleep, when they occurred at high frequency.
In contrast to the period of sleep before REM sleep the patient lay on his back. In the first hour of sleep some movements of the legs were noted. They soon disappeared. Following the period of frequent apneas nasal CPAP was given. At 5 cm. waterpressure no more apneas occurred and the patient stopped snoring. To our surprise the leg movements returned and were seen continuously. They had the typical aspect of periodic limb movements but occurred without arousals. The patient slept well during this last half of the night and was satisfied with this first try-out of nasal CPAP.

This case history turned our attention to mutual aspects of sleep apnea and periodic movement of the limbs syndrome (PMLS):
1. Both syndromes can be seen in the same patient.
2. Treatment of one of the syndromes can have a deteriorating effect on the other one.
Both phenomena were investigated further with an emphasis on prevalence and demographic factors.
METHODS

In our laboratory all patients who were clinically suspected of sleep apnea syndrome had a 48-hour polygraphic examination at home in which EEG, EMG, EOG, respiration and often leg movements were recorded. When a severe sleep apnea syndrome (apnea index higher than 20 per hour and excessive daytime sleepiness) was found the patient underwent a clinical polysomnography in our sleep unit. During that registration leg movements were always recorded. The polygraphic recordings were scored according to the ASDA rules (1). All patients were asked to complete the Sleep Disorders Questionnaire, originally compiled by Douglass et al. (2) and validated for the Dutch language by Sweere (3). This questionnaire results in a score which indicates the probability of certain sleep disorders.

RESULTS

In the period from June 1993 to September 1995 in our Center for Sleep and Wake Disorders 91 patients were seen who fulfilled the inclusion criteria. Twenty-seven (30%) of these patients also had periodic movements of the limbs according to the ASDA criteria (1). Twenty-four of these patients were males. Their ages ranged from 37 to 73 years, with a median of 46 years.

As could be expected from such a group of patients suspected to suffer from sleep
apnea syndrome, excessive daytime sleepiness, snoring and to a lesser extent complaints about apneas were the main clinical features. Thirty percent of the patients complained about unpleasant feelings in the legs during the evening and night. This percentage is approximately as high as that of patients complaining about apneas (fig. 2)!

Figure 2.
Complaints of the patients. Excessive daytime sleepiness (EDS) in 67%, snoring in 78%; apneas -often hetero-anamnestic- in 33% and leg movements during evening and night in 30%.

The mean body-mass index (BMI) was 29, ranging from 22 to 42. Except for being obese no other major abnormalities were found in this group of 27 patients with the combination of sleep apnea syndrome and PMLS.

More particularly, history nor examination gave clues to polyneuropathy or other disorders in the legs.

The Sleep Disorder Questionnaire was completed by all patients. The scores derived from this questionnaire provide clues for the sleep disorder the patient is suffering from, divided in four main categories: sleep apnea syndrome, narcolepsy, depression and abnormal movements during the night. The 27 patients had on average the following percentile scores on the Sleep Disorders Questionnaire: sleep apnea syndrome: 38, narcolepsy 21, depression 23 and PMLS 46. Thus, the average score of this group of patients for PMLS was even higher than that for the main abnormality, which was the apnea syndrome! Twenty-five per cent of the patients investigated had a score higher than the 50th percentile for apnea syndrome and half of the patients scored above the 50th percentile for abnormal movements during the night. At polysomnography all patients had a severe sleep apnea syndrome combined with PMLS according to the ASDA criteria. Apneas and movements of the legs occurred independently. Care was taken that movements which could possibly be related to an apnea period were not used as criterion for the existence of PMLS.
In a few patients there was a mutual influence between both disorders. The patient mentioned in the case history is a good example of such patients. In the group of 27 patients with both sleep apnea syndrome and PMLS two patients were found in which PMLS worsened after successful nasal CPAP; in two other patients PMLS ameliorated in parallel to the apneas. For the other 23 patients no mutual influence could be demonstrated.

DISCUSSION

Combination of sleep apnea syndrome and PMLS has been described before. Its prevalence is not known. From the results of our unselected group it appears that the combination can be seen often. The clinical relevance of the sleep apnea syndrome in these patients is beyond doubt. Whether the patients also had clinically relevant PMLS is not clear. When the ASDA criteria are applied a positive diagnosis of PMLS can be made, but from our experience we are not sure about the clinical value of the cut-off points given in this consensus paper. This aspect warrants further investigation. Accordingly we doubt whether the PMLS in the patients who have a combination of this disorder and sleep apnea syndrome calls for therapy. If so, difficulties arise: when the standard therapy for PMLS is given which in our setting is Clonazepam, perhaps worsening of the sleep apnea syndrome occurs. At the moment we treat those patients who have sleep apnea syndrome and PMLS with a combination of nasal CPAP and -in case of a movement of the legs higher than 20 per hour- with L-DOPA. The results of this combination therapy will be evaluated in the near future.

Up to now no satisfying explanations for the concurrence of both disorders in one patient have been given. It seems unlikely that the combination is based on mere coincidence. A central mechanism leading to diminished inhibition of motorneurons is postulated as an explanation for periodic movement of the limbs syndrome. Whether there is a connection between the sleep apnea syndrome and this disorder of regulation of firing of motorneurons is completely unknown. Intriguing in this respect is the mutual influence found in some of our patients. Perhaps the worsening of PMLS after nasal CPAP as found in the index patient is merely the result of better sleep after the device has been applied. On the other hand, mutual influence of central mechanisms might also be the cause.

CONCLUSION

In patients suffering from the sleep apnea syndrome, PMLS also occurs in a high percentage. In a few patients there is even a mutual influence between both disorders. Pathogenesis and clinical relevance of PMLS is still not clear and needs further investigation.
REFERENCES


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