Case Report

Body Rolling in Adults Associated to Obstructive Sleep Apnea

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Abstract

Background: Sleep related rhythmic movement disorder (RMD) is not frequent in adults, and probably its pathogenesis is different to children. In this study we show the possibility of association of Body Rolling and Obstructive Sleep Apnea (OSAS) in adults, as an infrequent but remarkable fact.

Methods: We reviewed the databases of the Sleep Units in two different hospitals looking for patients with RMD in adulthood. All the patients included in the database were studied with full polysomnography and very often video (if clinically indicated). Adult patients with Body Rolling were selected.

Results: Only two adult patients were found to have RMD, both of them with the form of Body Rolling. The two patients also suffered Obstructive Sleep Apnea. They were a 49 year-old woman and a 29 year-old man. In one of them, CPAP was tried with improvement of the apneas but not change in the episodes of Body Rolling.

Conclusions: Body rolling in adults can be associated to Obstructive Sleep Apnea syndrome. This association can be important in order to indicate the polysomnograms in patients with OSAS and also in patients with suspicion of RMD.

INTRODUCTION

As OSAS is very prevalent in the general population, the possible association of other disorders with this process has to be taken with precaution. OSAS has been implicated in the presence of hypertension, coronary artery disease, congestive heart failure, stroke, arrhythmias, diabetes, depression etc [1]. Some reports have linked OSAS to other primary sleep disorders as REM Sleep Behaviour Disorder, sleepwalking, periodic limb movements, confusional arousals, sleep related eating disorder or sleep terrors [1,2]. The study of these or other associations could be crucial for a deeper knowledge of its path physiology and also to better understand and treat those patients who do not improve as expected with the correct treatment.

Few reports show the association of OSAS and the Sleep related rhythmic movement disorder (RMD) [1,3-5]. This disorder consists of clusters of repetitive and stereotyped movements of head, trunk and/or legs during wakefulness or any stage of sleep, even REM, but mostly in the wake-sleep transition. It is a sleep disorder mainly present in infants and children that only exceptionally is observed in adulthood. According to the information given in the ICSD-3, about 50 cases have been reported in adults. Kohyama summarized 33 published cases and only 3 were older than 30 years-old [3]. Other authors think that the frequency of the disorder in adulthood could be underestimated because of the paucity of symptoms [6]. Diagnosis is clinical, by history; the family normally explains the movements and, often, provides a video recording. Polysomnography is rarely necessary for diagnosis. There are several clinical forms of the disorder given the parts of the body involved in the movements: body rocking, head banging, head rolling etc [3,4,7-10]. Body rolling is one of the less frequent forms, only 4 of the 33 cases presented in the revision of Kohyama had this type of RMD [3]. Clinically it consists on moving the trunk from side to side, normally in a supine position. Etiology is unknown and there is no explanation for the mechanism. An alteration in the central motor pattern generator has been advocated as related to its genesis[11]. Although there is no definitive treatment, clonazepam, or other benzodiazepines, have been useful in some cases[12-14], but not in others [15].

It has been suggested that OSAS might trigger RMD and cases of improvement of RMD with CPAP have been reported [1, 5, 6,16]. The goal of this paper is to present the only 2 adult patients with Body Rolling confirmed by polysomnography. Remarkably, both of them suffered also OSAS. We think that this association has not been aleatory, and in our opinion adult patients with RMD should be considered as candidates for having OSAS, and a full-polysomnogram should be performed.

PATIENTS AND METHODS

We reviewed the databases of patients who underwent polysomnographic studies in our Sleep Units from two different hospitals (CUN, Hospital de la Ribera), looking for patients with RMD in adulthood. The patients gave their informed consent to use their clinical data and results of the sleep study for research, eliminating their names and personal data. The Ethical Committee of the university hospital (CUN) evaluated the study and found it suitable and followed the ethical and deontological rules. The tests were not performed specifically for the study but as part of the routine clinical workups. These databases include 11,000 and 13,000 patients respectively, being the patients identified only with a record number. We only found two adult patients with RMD, both of them with the form of Body Rolling.

Patient #1

She is a 49 year-old woman, married, intellectually normal, with diagnosis of Diabetes Mellitus type 2 and hypothyroidism. The patient referred being tired in the morning, and her husband confirmed snoring and breathing stops. She was referred for a polysomnography due to the suspicion of sleep apnea. She had two risk factors for Obstructive Sleep Apnea Syndrome: hypothyroidism and obesity (BMI: 34.4). Other relevant data in the medical history were migraine and livedo reticularis. At admission, she indicated that since the last 2-3 years she moved a lot while sleeping, mainly at the beginning of sleep but also during the night and when awakening. The physical exam was unremarkable. She was on levothyroxine, metformin and cefediol.

Patient #2

The second patient is a 29 year-old male, single, with low academic performance in the elementary school (he attended a special school, being borderline but not clearly retarded). He had been working as mason for ten years. The physical exam was normal. He was not receiving any medication at the time of the visit.

He was referred to the Sleep Center due to movements during sleep. His mother indicated that he had some movements since childhood but he never was evaluated by a Sleep specialist. His mother said that he moves the whole body and sometimes he gets the pillow and rhythmically moves his head. When he moved he produced a loud noise that annoyed the family, and this was the reason to be sent to the doctor. The family forced the patient to place the bedroom downstairs to avoid the noise. He was aware of having episodes of movements but he did not complain of sleep problems.

Polysomnnographic studies

The patients underwent a standard nocturnal polysomnography with full montage including EEG channels, EOG, EKG, EMG of chin and tibialis anterior, body position, oxygen saturation, airflow and thoraco-abdominal bands. Patient #1 was studied with a 36-channel Stellate System, and patient #2 with a 57 channels Profusion PSG E-series System (Compumedics, Australia). Both patients were recorded with video and audio.

RESULTS

Patient #1

The polysomnography showed the presence of obstructive apneas occurring predominantly in supine position. The apnea/hypopneas index was 30.26, but the involvement of oxygen saturation was mild (Figure 1A,2). Sleep structure was within normal limits, with 5 REM periods.

She presented 7 episodes of body rolling with duration of 48, 22, 3, 2, 7, 2.5 and 17 minutes respectively. During the long episodes, the movements could stop for less than 2 minutes, and then continue. The episodes occurred in somnolence, stage N1 and stage N2, but never in REM sleep (Figure 1A). The video showed the typical movements of body rolling. All the episodes were in the same position. It was striking the long duration of the episodes at the beginning of sleep and at the end of the night. The recording shows a typical rhythmic artefact in all channels; during most of the episodes the breathing was irregular but the flow limitation was not long enough to be considered as apnea (Figure 3).

In the study with auto-CPAP the apneas and hypopneas were reduced but not completely corrected (residual AHI was 10) and most of them were central events. In this night, she had 6 episodes of body rolling (Figure 1B), the first one being a little longer than in the previous night (50 minutes). The duration of the other episodes was 4, 3, 7, 3 and 17 minutes respectively.

Patient #2

The PSG confirmed the diagnosis of RMD. Obstructive apnea and hypopneas were also recorded. AHI was 11.25, with a minimum oxygen saturation of 78%. He was suffering very frequent and severe RMD, having the movements during 31.3% of the total sleep time. They appeared in all sleep stages, REM and no-REM. One of the episodes developed in REM sleep right after an obstructive sleep apnea. The video confirmed the typical movements of body rolling. The recording showed that some of the clusters of RMD started right after the end of an apnea (Figure 4).

This patient did not accept to be treated with CPAP. The OSAS were mild and we did not insist on this treatment.

DISCUSSION

In this report we present the association of OSAS and Body Rolling in adults. It is already quoted that RMD can be associated to OSAS or other sleep disorders [1,6], but the communications have been very exceptional. Prevalence of OSAS is very high, but prevalence of RMD in adults is extremely low [17]. We found 2 cases of this combination in adults, in a sample of more 5.000 patients with OSAS. The prevalence of RMD in adults is probably...
Figure 1 Sleep graphs of the basal (A) and CPAP (B) studies in patient #1. The episodes appear in non-REM sleep and wakefulness in both nights. The respiratory detections included the apnea/hypopnea events that in the second night were fairly controlled using the auto-CPAP.

Figure 2 Polysomnographic recording in patient #1 during one the episodes. In spite of the movement artefact, it is possible to observe the irregular breathing (Flow) and the beginning with a decrease in the airflow (upper row).
Apart from the exceptional but possible association, the cases presented here raise several interesting points to be addressed.

The first point of this association is the age. Both patients were adults. The first patient is a woman in advanced adulthood, and the second a 29-year old man. In both cases it was not possible to know exactly the time of the beginning of the disorder, but according to the responses of the family it could be in the lower than 1/5000, but with only 2 patients it is not possible to sustain any data for incidence or prevalence.
adulthood in the first case, and in childhood in the second one. In any case, we can confirm that adulthood is probably more likely to have this association than childhood. Within the several clinical forms of this disorder, Head Banging and Body Rocking are the most frequent [6,9,18]. Body Rolling is very infrequent and even more exceptional in adults than in infancy. Some cases of Body Rolling since childhood have been published, but OSAS was ruled out [19,20]. So, the patients we present here are a really unusual finding. We understand that only two examples are not enough to extract general conclusions, but this association is rare and it cannot be forgotten.

The second point of interest is the possibility of triggering episodes of typical body rolling with an obstructive sleep apnea. In our second patient, this fact was clearly determined in several occasions. The question about the mechanism is open. The mechanical input or the Valsalva maneuver could induce the movements. However, apneas are not enough to induce RMD in the majority of patients. Another possibility is that arousals or apneas can be the trigger, but none of these hypotheses can be confirmed.

Other point of discussion is the relationship with the treatment with CPAP. Some reports insisted on the possibility of RMD triggered by apneas [5,6]. Gharagozlou reported improvement of RMD with CPAP in a 51-year-old man with Body Rolling associated to OSAS and Restless Limb Syndrome [16]; their hypothesis is that arousals with or without apnea might induce the RMD [16]. In our second patient, during the night with CPAP, the amount, duration, and distribution of the clusters of Body Rolling were similar to the first night, in spite of the almost complete correction of the respiratory problems. In both nights the longest clusters occurred at the beginning and at the end of the sleep. The lack of change with CPAP in our patient might be explained by the similar arousal index in both nights, without and with CPAP.

In conclusion, the goal of this paper was to present the infrequent presence of Body Rolling in adulthood. The most relevant finding has been the association with OSAS: the only two patients that we found with Body Rolling suffered also from Sleep Apnea. Moreover, in one of them some of the episodes were triggered by apneas. These findings suggest that Body Rolling in adulthood can be an unusual manifestation linked to OSAS. Adult patients with complaints of possible RMD should be asked about symptoms related to OSAS. In our opinion, a full polysomnography is a clear indication for adult patients with RMD because OSAS can be very relevant for their treatment and outcome. Finally, our cases confirm that OSAS is a very complex disease with relationships with many other disorders. Cases with some atypical features should be studied in detail.

CONFLICT OF INTEREST

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers’ bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

REFERENCES
