Clinical Neurophysiology Practice 6 (2021) 229–233

Contents lists available at ScienceDirect

Clinical Neurophysiology Practice

journal homepage: www.elsevier.com/locate/cnp

Research paper

Heart rate change and clinical characteristics in patients with neck myoclonus: An observational study

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ARTICLE INFO

Article history:
Received 13 September 2020
Received in revised form 27 May 2021
Accepted 18 June 2021
Available online 27 July 2021

Keywords:
Heart rate
Neck myoclonus
Autonomic nervous activity
Incomplete arousal
Daytime sleepiness

ABSTRACT

Objective: This study was conducted to evaluate heart rate (HR) change and clinical characteristics in patients with neck myoclonus (NM), a physiological motor phenomenon occurring during sleep.
Methods: For 18 consecutive patients in whom NM was confirmed from video-polysomnography, we analyzed 576 NMs. Change rate of HR at each 1 sec point towards the averaged HR in prior 5 sec period was calculated before and after all NM events.
Results: Findings show NM events as more prevalent during REM sleep than during NREM sleep (83.9% vs. 16.1%). For NM without cortical arousal in REM and NREM sleep, the respective HR increased 20 s before NM ($p < 0.05$); the change rate was up to 13%. For NM with cortical arousal in REM sleep, the HR increased 50 s before NM ($p < 0.05$); the change rate reached 18%. Three NM subjects showed abnormal vocalization or shouting during REM. Six NM subjects had excessive daytime sleepiness without sleep disorder.
Conclusion: HR increased before NM events, which may be associated with pathophysiology of NM. NM may possibly be associated with excessive daytime sleepiness or abnormal behaviors during REM sleep.
Significance: HR increase is associated with pathophysiology of NM and clinical symptoms.

1. Introduction

Neck myoclonus (NM) is regarded as a physiological motor phenomenon occurring during sleep, similarly to periodic limb movements during sleep (PLMS), twitches observed from rapid eye movement (REM) sleep, fragmentary myoclonus, and sleep starts. (American Academy of Sleep Medicine, 2014) NM is observed as one or more short stripe-shaped movement-induced artifacts on polysomnography. These and other physiological motor phenomena are frequently observed in clinical settings. Nevertheless, the clinical / patho-physiological relevance has not been well elucidated. Regarding PLMS, heart rate increase, blood pressure change, sympathetic hyperactivity or cortical activity associated with the motor events have all been reported (Allena et al., 2009, Ferri et al., 2007, Pennestri et al., 2013, Sforza et al., 2005). In addition, PLMS are known to induce or exacerbate sleep fragmentation and subsequently to promote or trigger NREM parasomnia episodes (Castelnovo et al., 2018). On the other hand, the effects of NM on the heart rate and their association with other sleep disorders have not been reported in the literature to date. A recent study showed NM as accompanied by leg movements, arousal or awakening, or stage shift (Wolfensberger et al., 2019). Because of sleep fragmentation caused by NM, NM might be associated with non-restorative sleep and daytime sleepiness.

Considering these earlier findings, this study was conducted to investigate heart rate change, clinical effects, and the NM phenotype.

2. Methods

2.1. Subjects

We retrospectively reviewed consecutive video-polysomnography (v-PSG) recordings accumulated during April 2016 through April 2018. Patients treated using medications possibly influencing motor symptoms at the time of v-PSGs, such as antidepressants, benzodiazepines and Z-drugs and sedating antihistamines, and affecting cardiovascular functions, such as beta blocker, anticoagulant and antihypertensive drugs, were excluded.

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https://doi.org/10.1016/j.cnp.2021.06.005
2467-981X/

Of 18 patients who were shown to have NM, one or more short stripe-shaped movement-induced artifacts on polysomnography, were examined in this study (Frauscher et al., 2010). Fig. 1 represents examples of NM. For all patients, daytime sleepiness was assessed using the Epworth Sleepiness Scale (ESS) (Johns, 1991). Among the 18 patients, 12 patients who were suspected of having central disorders of hypersomnolence underwent multiple sleep latency tests following nocturnal v-PSG. The study protocol was reviewed and approved by the Ethical Committee of the Neuropsychiatric Research Institute.

2.2. Nocturnal video-polysomnography and scoring

Nocturnal video-polysomnography was performed using a standard system (Alice 6; Respironics Inc., Murrysville, PA, USA) including the following: six channels of scalp electroencephalographic data (F3/A2, F4/A1, C3/A2, C4/A1, O1/A2, O2/A1); two channels of electro-oculography; electromyography (EMG) of the submentalis muscle, the flexor digitorum superficialis muscle, and the bilateral anterior tibialis muscle; electrocardiography; nasal/oral airflow data; a percutaneous oximetry sensor for recording oxygen saturation data; a microphone for detecting snoring sounds; and chest/abdominal respiratory effort data. Sleep stages and electroencephalographic arousal, periodic limb movements during sleep, and respiratory events were scored carefully by board-certified sleep technologists according to criteria established by the American Academy of Sleep Medicine (Berry et al., 2012).

2.3. Analysis of heart rate change

For all 18 v-PSG recordings, board-certified sleep technologists visually re-scored NM events on v-PSG recordings. They carefully extracted periods of 700 s including NM from stage-scorable epochs without large artifacts. We firstly detected 587 NM events, of which 576 NM events were able to be analyzed. For all NM events, the 700 s periods before and after NM (350 s) were analyzed (total of 576 periods). In each period, change of HR in each 1 s point compared to the averaged HR in the last 5 s were assessed as increase rate of HR. Then, the increase rate was standardized dividing by the averaged heart rate in the respective 700 s periods to exclude individual variation of HR. Finally, we averaged the standardized rate of heart rate increase for all the subjects. These analyses were conducted using the software (PLMS Event Analysis; Norupro Light Systems Inc., Tokyo, Japan) (Sasai et al., 2013).

2.4. Statistical analysis

Chi-square tests were applied to compare categorical variables. Repeated measured ANOVA was used to compare the increase rate of HR at each point with significance of $p < 0.05$. A point of increase of HR was inferred as the starting time of the time period with significant increase of HR. All statistical analyses were conducted using software (IBM SPSS ver. 24.0; IBM Corp., Armonk, NY, USA).

3. Results

3.1. Demographic information and polysomnographic findings

Table 1 presents demographic characteristics and polysomnographic findings associated with the NM cases. Results show that NM was more predominant during REM sleep than during NREM sleep (83.9% vs. 16.1%, $P < 0.05$). Of the 18 NM patients, 15 (83.3%) were under 45 years old. The index of NM was higher in REM sleep than in NREM sleep ($P < 0.05$). Of 576 NM events, 40 events (6.9%) were accompanied by cortical arousal. No awakening was found with NM events. No PLMS were associated with NM events. For other types of leg movements which did not satisfy the PLMS criteria (isolated LM or short duration of LM), 2/94 of neck myoclonus in NREM sleep (2.1%) and 31/482 of neck myoclonus in REM sleep (6.4%) accompanied with these other types of leg movements. These other types of leg movements accompanying with neck myoclonus were observed in three patients and occurred in arousal response following neck myoclonus.

3.2. Clinical findings and patient diagnoses

Table 2 presents clinical information of the patients. Thirteen patients (72.2%) complained of excessive daytime sleepiness at the first visit. Of all 18 patients with NM, final diagnoses were the following: sleep talking, $n = 2$; REM sleep behavior disorder, $n = 1$; narcolepsy type 2, $n = 1$; idiopathic hypersomnia, $n = 2$; insufficient sleep syndrome, $n = 3$; mild obstructive sleep apnea, $n = 2$; sleep-related eating disorder, $n = 1$; hypersomnolence not otherwise specified, $n = 6$. Six patients who complained of excessive daytime sleepiness but who exhibited no clear sleep disorder

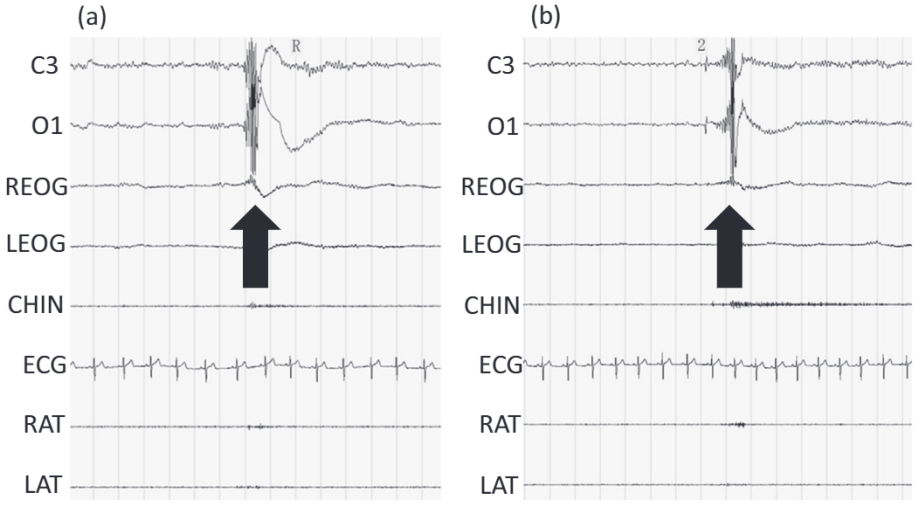

Fig. 1. Examples of neck myoclonus. Arrows indicate neck myclonus in REM sleep (a) and in NREM sleep (b). REM, rapid eye movement sleep; NREM, non-REM sleep, REOG, right electrooculogram; LEOG, left electrooculogram; ECG, electrocardiogram; RAT, right anterior tibialis; LAT, left anterior tibialis.

Table 1
Demographic and polysomnographic characteristics of the subject NM cases.

Number of patients	18
Number of female patients (%)	3 (16.7)
Age (yr)	32.7 ± 15.1 (19–76)
BMI (kg/m2)	23.1 ± 3.4 (16.6–31.7)
ESS	14.3 ± 5.8 (4–22)
Heart rate (/min)	72.6 ± 7.3 (60–86)
Systolic blood pressure (mmHg)	116 ± 16 (88–146)
Dyastolic blood pressure (mmHg)	68 ± 13 (38–92)
TST (min)	457.4 ± 55.6 (335–514.5)
WASO (%SPT)	9.3 ± 7.8 (2.3–31.5)
REM (%SPT)	21.8 ± 5.6 (10.4–32)
N1 (%SPT)	10.6 ± 5.2 (4.1–25.5)
N2 (%SPT)	50.8 ± 8.1 (33.3–67.7)
N3 (%SPT)	7.5 ± 4.5 (0–14.7)
AHI (/hr)	3.2 ± 2.3 (1.0–9.0)
AI (/hr)	14.1 ± 6.6 (6.3–35.3)
RAI (/hr)	1.5 ± 1.5 (0–4.8)
PLMI (/hr)	2.7 ± 7.4 (0–29.8)
PLMAI (/hr)	0.2 ± 0.6 (0–2.5)
SE (%)	88.9 ± 8.7 (66.5–97)
Phasic EMG activity during REM sleep (%)	1.9 ± 2.6 (0–8.5)
Tonic EMG activity during REM sleep (%)	0.3 ± 0.4 (0–1.2)
Total number of NM	576
Number of NM during NREM (%/total)	93 (16.1)
Number of NM during REM (%/total)	463 (83.9)
NM/NREM (n/hr)	1.0 ± 1.4 (0–5.9)
NM/REM (n/hr)	15.5 ± 10.6 (1.8–43.3)

Values are expressed as average ± standard deviation (range).
BMI, body mass index; ESS, Epworth Sleepiness Scale; TST, total sleep time; SPT, sleep period time; WASO, wake after sleep onset; REM, rapid eye movement sleep; AHI, apnea hypopnea index; AI, arousal index; RAI, respiratory arousal index; PLMI, periodic limb movement index; PLMAI, periodic limb movement related arousal index; SE, sleep efficiency; EMG, electromyogram; NM, neck myoclonus; NREM, non-REM sleep

or difficulty other than NM were found. They were diagnosed as having hypersomnolence not otherwise specified. From v-PSG results, two patients showed symptoms such as sleep terrors (shouting) during REM sleep; one showed dream enactment behavior during REM. Five patients (27.8%) showed mild bruxism during NREM sleep.

3.3. Heart rate change associated with neck myoclonus

Fig. 2 presents heart rates before and after neck myoclonus. During REM sleep, the heart rate increased 20 s before NM without cortical arousal and 50 s before NM with cortical arousal ($p < 0.05$, respectively). The maximum heart rate increase reached 13% in NM without cortical arousal. In contrast, the rate reached 18% in NM with cortical arousal. During NREM sleep, no NM with cortical arousal was detected. For NM without cortical arousal in NREM sleep, the heart rate increased 20 s before NM onset ($p < 0.05$). The rate of increase was as high as 13%, roughly equivalent to that in REM sleep.

4. Discussion

This report is the first to describe a study eliciting a heart rate increase accompanied with NM and clinical symptoms, with detailed diagnosis of coexisting sleep disorders. Results showed a 13% heart rate increase from the average heart rate of 20 s before the onset of NM without cortical arousal, and showed an 18% heart rate increase 50 s before the onset of NM with cortical arousal. Results show that NM was predominant during REM sleep (REM sleep vs. NREM sleep: 83.9 vs. 16.1%). Results of v-PSG revealed two patients who showed symptoms such as sleep terrors; one patient showed dream enactment behavior during REM sleep. Six patients who complained of excessive daytime sleepiness but who did not have sleep disorders or difficulties other than NM showed excessive daytime sleepiness.

For these consecutive patients, NM was quite prevalent in patients younger than 45 years old (83.3%) and was predominant during REM sleep (83.9%). The NM index was 1.0 ± 1.4/hr during NREM sleep and 15.5 ± 10.6/hr during REM sleep. This finding is consistent with those of an earlier study, which comprehensively assessed NM characteristics for 112 patients: the NM index was higher in patients younger than 45 years old than in patients older than 45 years old. Also, NM was common in REM sleep (Frauscher et al., 2010). These characteristics were quite different from those of PLMS or fragmentary myoclonus; these two diseases increase with age and PLMS is common in NREM sleep (Frauscher et al., 2011; American Academy of Sleep Medicine, 2014).

In this study, NM with cortical arousal was observed in 6.9% of all NM events (only during REM sleep). None of the NM events were accompanied with awakening. A recent Swiss study found NM events associated with arousals to be 62.1% in total sleep, 60.7% in REM sleep, and 74.3% in NREM sleep (Wolfensberger et al., 2019). In their study, 19.2% and 52.2% of NM events were accompanied respectively with awakening and leg movements. Compared with this study, NM events in our cases were less frequently associated with arousals and awakenings. The Swiss study selected patients with at least 5 video- confirmed NMs. In contrast, we selected patients with at least one v-PSG confirmed NM. Therefore, the present study might include more mildly affected patients than the Swiss study. Other possible reason for this discrepancy might be the younger subjects examined for the present study: 32.7 ± 15.1 vs. 36.0 ± 15.1 years of mean age; 5.6% vs. 27.3% of subjects over 45 years of age. Additionally, the difference in comorbid sleep disorders should also be considered. Of 11 patients examined in the Swiss study, 5 were suspected of OSA and were ultimately diagnosed as sleep-related breathing disorders including OSA, snoring, and flow limitation. Sleep-disordered breathing of these types can trigger sleep disruption, which might cause high arousability because of NM. In contrast, of 18 patients examined for the present study, 13 were suspected of narcolepsy; 12 were finally diagnosed as having central disorders of hypersomnolence. The effect of NM on sleep stability or daytime function can be inferred as depending on patients' underlying disease associated with sleep propensity or sleep depth. Future studies of many more patients can be conducted to clarify this issue.

The main finding in this study is that the heart rate increased in association with NM events. The heart rate increased several tens of seconds before NM events. The increase rate was higher in NMs with cortical arousal than those without. To date, PLMS, isolated LM or short-interval LM have been reported to be associated with heart rate increase: the heart rate increased several seconds before PLMS events (Ferri et al., 2017; Ferrillo et al., 2004). This finding was also reportedly observed for isolated leg movements (Ferri et al., 2007). Moreover, sympathetic hyperactivity and changes in cortical electroencephalographic hyperactivity have been associated with PLMS (Allena et al., 2009). These findings suggest that autonomic nervous activity and relevant cortical activity are associated with the pathophysiology of PLMS; the activity might be linked to PLMS formation. In this study, NM events also accompanied with heart rate increase although they were not associated with PLMS at all. Given these findings, the heart rate increase proceeding to NM might be associated with the pathophysiology of NM. As NM is a more dynamic axial movement than PLMS, the heralding autonomic activation is considered to be stronger and occur earlier than that heralding PLMS. Among patients with NM examined for the current study, some patients showed 30 or more NM events. Moreover, the heart rate increases accompanying NM events apparently occurred earlier and continued longer than those accompanying PLMS events. The pathologi-

Table 2
Clinical information of the subject NM cases.

No.	Sex	Age	ESS	Chief complaint at the first visit	Suspected diagnosis / Reason for v-PSG	Symptoms confirmed on v-PSG	Medication	Diagnosis	Number of NM during NREM	Number of NM during REM
1	F	26	4	nocturnal body movements, sleep talking/shouting	NREM parasomnia	shouting during REM	Varenicline	sleep talking	0	52
2	M	22	11	daytime sleepiness	narcolepsy	shouting during REM		narcolepsy type 2	10	79
3	M	32	6	sleep talking, elementary movements during sleep, insomnia	insomnia			sleep talking	8	34
4	M	26	15	daytime sleepiness, difficulty awakening	narcolepsy, CRSWD			hypersomnia not otherwise specified	0	29
5	M	24	13	daytime sleepiness	narcolepsy	bruxism during NREM (0.35/hr)		hypersomnia not otherwise specified	1	16
6	M	40	10	nocturnal eating	SRED			SRED	0	9
7	M	57	13	nocturnal abnormal behavior, insufficient sleep	RBD			ISS	1	9
8	M	26	17	daytime sleepiness	narcolepsy	bruxism during NREM (0.12/hr)		hypersomnia not otherwise specified	10	35
9	M	22	21	daytime sleepiness, , difficulty awakening	narcolepsy, CRSWD			IHS with LST	0	40
10	M	21	21	daytime sleepiness	narcolepsy	bruxism during NREM (0.61/hr)		ISS	5	22
11	M	76	4	nocturnal abnormal behavior	RBD, NREM parasomnia	talking during REM, DEB		RBD	29	30
12	M	24	13	daytime sleepiness	narcolepsy	bruxism during NREM (0.17/hr)		hypersomnia not otherwise specified	13	17
13	M	27	18	daytime sleepiness	narcolepsy	AHI = 7.0/hr		mild OSA	7	23
14	M	22	21	daytime sleepiness, difficulty maintaining sleep	narcolepsy	bruxism during NREM (2.58/hr)		ISS	3	19
15	M	55	10	daytime sleepiness	narcolepsy	AHI = 9.0/hr		mild OSA	0	3
16	F	19	22	daytime sleepiness	narcolepsy			hypersomnia not otherwise specified	3	47
17	M	28	22	daytime sleepiness	narcolepsy			hypersomnia not otherwise specified	0	13
18	F	42	17	daytime sleepiness	narcolepsy			IHS w/o LST	3	6

ESS, Epworth Sleepiness Scale; v-PSG, video polysomnography; REM, rapid eye movement sleep; NREM, non-REM sleep; CRSWD, circadian rhythm sleep wake disorder; SRED, sleep related eating disorder; RBD, REM sleep behavior disorder; DEB, dream enactment behavior; AHI, apnea hypopnea index; OSA, obstructive sleep apnea; ISS, insufficient sleep syndrome; IHS, idiopathic hypersomnia, LST, long sleep time.

cal level of NM cannot be ascertained from these findings. However, frequent NMs might disrupt restorative sleep because of their dynamic autonomic arousal.

From results of the present study, six patients who had no sleep disorder or difficulty other than 20 or more NM events, in whom all the NM events with cortical arousal were detected, were reported to have excessive daytime sleepiness (ESS $\geq$ 11). Although further analyses comparing their daytime sleepiness with control's should be conducted, this result may possibly suggest that NM can be associated with sleep disruption and with subsequent subjective un-restorative sleep or daytime sleepiness. Moreover, other than an elderly patient diagnosed with RBD, two young subjects had sleep talking or sleep terror-like symptoms (shouting) during REM sleep. Comorbid conditions such as obstructive sleep apnea, PLMS, and chronic pain are known to induce or exacerbate sleep fragmentation and subsequently to promote or trigger NREM parasomnia episodes (Castelnovo et al., 2018). In some cases, NM events may possibly provoke incomplete awakening and trigger sleep talking, shouting, and dream enactment behaviors during REM sleep, albeit that this small retrospective study cannot clarify causal relationship. To clarify this issue, further large study is needed. Regarding bruxism, the NM episodes were not associated with bruxism: all bruxism events occurred during NREM sleep.

Several limitations should be explained along with these study results. This small retrospective study was unable to clarify a causal relation between NM and daytime function or nocturnal symptoms. In addition, a lack of normal controls limits to generalize of the current results, heart rate and clinical characteristics, for NM patients. Additionally, we selected the patients from consecutive patients who showed neck myoclonus in v-PSG recordings accumulated during April 2016 through April 2018; however, all the selected patients had a complaints of sleep disorders e.g., excessive daytime sleepiness or nocturnal behaviors, and those who had medications possibly influencing motor symptoms were excluded. The small and specifically selected patient group from a single center limits to conclude that the present findings are related to neck myoclonus per se. A further investigation for more patients with NMs using a control group is warranted. Furthermore, in this study no NM events accompanied by awakenings. This is possibly because NM events accompanied by awakenings were excluded as the 11 un-scorable epochs. Finally, this study was unable to compare the impact of leg movements accompanied with NM on heart rate, because quite small number of leg movements were found in this small study (2.1% of NMs in NREM sleep and 6.4% of NMs in REM sleep of total NM events); all the NMs with leg movements occurred in arousal response following NM. Therefore, we unfortunately could not analyze, standardize, and compare the heart rate change with NM with and without leg movements separating from arousal response. In future large study, comparison among various types of neck myoclonus (with arousal, with leg movements or with PLMS) is warranted.

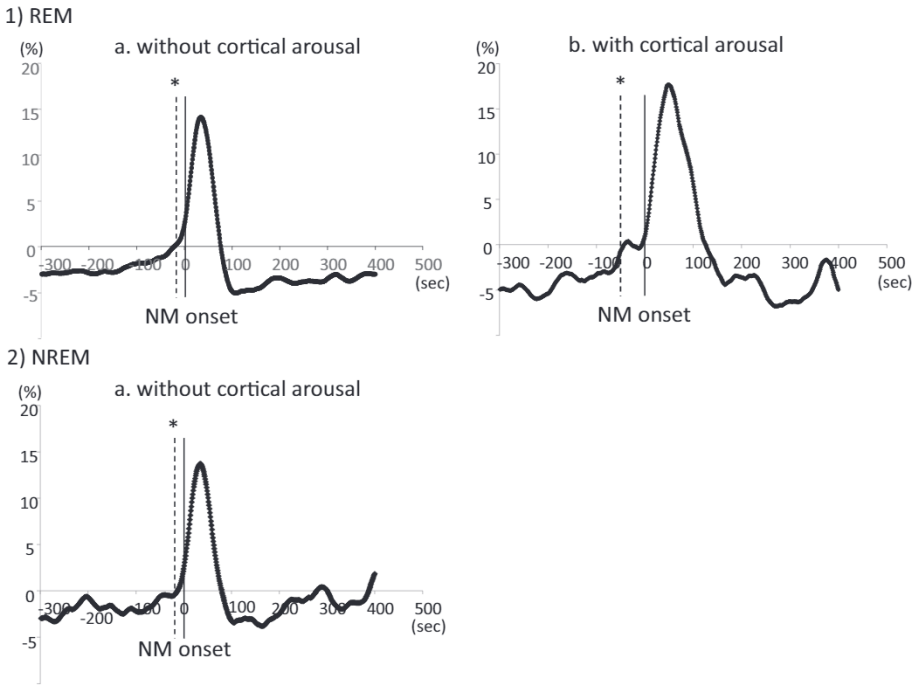

Fig. 2. Heart rate change before and after NM onset. REM, rapid eye movement sleep; NREM, non-REM sleep; NM, neck myoclonus. Y-axis represents the increase rate of heart rate and x-axis represents seconds. Vertical solid line represents the onset of NM; vertical dotted line represents the point of increased heart rate (*$p < 0.05$, repeated measured ANOVA).

5. Conclusions

Results showed that heart rate increased with NM events. The rate of increase was higher in NM with cortical arousal than in those without: NM could be associated with excessive daytime sleepiness or parasomnia-like symptoms, possibly because of incomplete arousal during REM sleep. Clarification of detailed information about nocturnal symptoms observed in patients with NM should be accumulated. Further investigation of causal relations between daytime function and NM is warranted.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

This study was supported by a MEXT/JSPS KAKENHI Grant-in-Aid for Scientific Research (C) (No. 18K07512) to T. S. We appreciate the cooperation of participants in this study and medical staff members of the Yoyogi Sleep Disorder Center.

This study was conducted at the Japan Somnology Center. All authors have seen and approved the manuscript.

References

Allena, M., Campus, C., Morrone, E., De Carli, F., Garbarino, S., Manfredi, C., et al., 2009. Periodic limb movements both in non-REM and REM sleep: relationships between cerebral and autonomic activities. Clin. Neurophysiol. 120 (7), 1282–1290.

American Academy of Sleep Medicine. International Classification of Sleep Disorders, 3rd ed. Darien, IL: American Academy of Sleep Medicine, 2014.

Berry, R.B., Brooks, R., Gamaldo, C.E., Harding, S.M., Marcus, C.L., Vaughn, B.V., 2012. The AASM Manuasl for the Scoring of Sleep and Associated Events: Rules, Terminology and Technical Specifications. American Academy of Sleep Medicine, Darien, Illinois.

Castelnovo, A., Lopez, R., Proserpio, P., Nobili, L., Dauvilliers, Y., 2018. NREM sleep parasomnias as disorders of sleep-state dissociation. Nat. Rev. Neurol. 14 (8), 470–481.

Ferri, R., Zucconi, M., Rundo, F., Spruyt, K., Manconi, M., Ferini-Strambi, L., 2007. Heart rate and spectral EEG changes accompanying periodic and non-periodic leg movements during sleep. Clin. Neurophysiol. 118 (2), 438–448.

Ferri, R., Rundo, F., Silvani, A., Zucconi, M., Aricò, D., Bruni, O., Lanuzza, B., Ferini-Strambi, L., Manconi, M., 2017. Short-interval leg movements during sleep entail greater cardiac activation than periodic leg movements during sleep in restless legs syndrome patients. J. Sleep Res. 26 (5), 602–605.

Ferrillo, F., Beelke, M., Canovaro, P., Watanabe, T., Aricò, D., Rizzo, P., et al., 2004. Changes in cerebral and autonomic activity heralding periodic limb movements in sleep. Sleep Med. 5 (4), 407–412.

Frauscher, B., Brandauer, E., Gschliesser, V., Falkenstetter, T., Furtner, M.T., Ulmer, H., et al., 2010. A descriptive analysis of neck myoclonus during routine polysomnography. Sleep 33 (8), 1091–1096.

Frauscher, B., Kunz, A., Brandauer, E., Ulmer, H., Poewe, W., Högl, B., 2011. Fragmentary myoclonus in sleep revisited: a polysomnographic study in 62 patients. Sleep Med. 12 (4), 410–415.

Johns, M.W., 1991. A new method for measuring daytime sleepiness: the Epworth sleepiness scale. Sleep 14 (6), 540–545.

Pennestri, M.-H., Montplaisir, J., Fradette, L., Lavigne, G., Colombo, R., Lanfranchi, P. A., 2013. Blood pressure changes associated with periodic leg movements during sleep in healthy subjects. Sleep Med. 14 (6), 555–561.

Sasai, T., Matsuura, M., Inoue, Y., 2013. Change in heart rate variability precedes the occurrence of periodic leg movements during sleep: an observational study. BMC Neurol. 13, 139.

Sforza, E., Pichot, V., Barthelemy, J.C., Haba-Rubio, J., Roche, F., 2005. Cardiovascular variability during periodic leg movements: a spectral analysis approach. Clin. Neurophysiol. 116 (5), 1096–1104.

Wolfensberger, B., Ferri, R., Bianco, G., Abbafati, M., Miano, S., Kaelin-Lang, A., Bassetti, C.L., Manconi, M., 2019. From physiological neck myoclonus to sleep related head jerk. J. Sleep Res. 28 (5). https://doi.org/10.1111/jsr.v28.510.1111/jsr.12831.