Research Article / Araştırma Makalesi

Doi: 10.4274/npa.y6477
Archives of Neuropsychiatry 2013; 50: 252-255
Nöropsikiyatri Arşivi 2013; 50: 252-255

Comparison of Polysomnography and Multiple Sleep Latency Test Findings in Subjects with Narcolepsy and İdiopathic Hypersomnia

Narkolepsi ve İdiopatik Hipersomnia Olgularının Polisomnografi ve Çoklu Uyku Latans Testi Bulgularının Karşılaştırılması

Murat ERDEM, Abdullah BOLU, A. Gazi ÜNLÜ, Mustafa ALPER, Sinan YETKİN

Gülhane Military Medical Academy, Division of Psychiatry, Ankara, Turkey

ABSTRACT

Introduction: Both narcolepsy and idiopathic hypersomnia are the main causes of excessive daytime sleepiness. In this study, we aimed to compare polysomnography (PSG) and multiple sleep latency test (MSLT) findings in narcolepsy and idiopathic hypersomnia patients.

Methods: The files of patients with narcolepsy and hypersomnia who were admitted between 1995 and 2009 were reviewed. We evaluated data from 94 patients with narcolepsy with cataplexy, 49 with narcolepsy without cataplexy and 140 patients with idiopathic hypersomnia.

Result: Sleep latency and REM latency were longer in idiopathic hypersomnia group than in narcolepsy with and without cataplexy group. Mean sleep latency in MSLT was the shortest in narcolepsy with cataplexy group. There was no difference in sleep efficiency, percentage of sleep stage and number of awakenings in PSG between three groups.

Conclusion: The findings of the study indicated that narcolepsy patients differ from idiopathic hypersomnia patients in terms of sleep latency and REM latency in PSG. *(Archives of Neuropsychiatry 2013; 50: 252-255)*

Key words: Narcolepsy, idiopathic hypersomnia, polysomnography, multiple sleep latency test

Conflict of interest: The authors reported no conflict of interest related to this article.

ÖZET

Giriş: Narkolepsi ve İdiopatik hipersomni gündüz aşırı uykululuk yakınmasının başlıca sebepleridir. Bu çalışmada narkolepsi ve idiopatik hipersomni olgularının Polisomnografi ve Çoklu Uyku Latans Testi bulgularının karşılaştırılması amaçlandı.

Yöntemler: GATA Uyku Araştırma Merkezine 1995-2009 yılları arasında başvuran, narkolepsi ve idiopatik hipersomni tanısı alan hastaların dosyaları retrospektif olarak incelendi. 94 katapleksili narkolepsi, 49 katapleksi bulunmayan narkolepsi ve 140 uzun uyku süreli olmayan idiopatik hipersomni hastasının verileri değerlendirildi.

Bulgular: İdiopatik hipersomni grubunda uyku latansı ve REM latansının her iki narkolepsi grubundan uzun olduğu, Çoklu Uyku Latans Testi ortalama uyku latansının katapleksili narkolepsi grubunda en kısa olduğu, üç grup arasında uyku etkinliği, uyku evrelerinin oranı, uyanıklık sayısı yönünden fark olmadığı saptandı.

Sonuç: Bulgular narkolepsi olgularının uyku latansı ve REM latansı yönünden idiopatik hipersomni olgularından ayrıştığını göstermektedir. *(Nöropsikiyatri Arşivi 2013; 50: 252-255)*

Anahtar kelimeler: Narkolepsi, idiopatik hipersomni, polisomnografi, çoklu uyku latans testi

Çıkar çatışması: Yazarlar bu makale ile ilgili olarak herhangi bir çıkar çatışması bildirmemişlerdir.

Introduction

Narcolepsy (NC) and idiopathic hypersomnia (IH) constitute the two main causes of the complaint of excessive daytime sleepiness. NC is a well known clinical picture with its symptoms including hypnagogic or hypnopompic hallucinations, sleep paralysis and disrupted night sleep as well as excessive daytime sleepiness and cataplexy. In different populations, the prevalence of narcolepsy with cataplexy has been found to be 2-5/10.000 (1,2,3). Higher prevalence rates have been found in Japan (16-18/10.000) and lower prevalence rates have been found in Israel (2/100.000) (4,5,6). Although narcolepsy frequently begins at the age of mid-twenties, cases of narcolepsy with an onset in the childhood have also been reported (7). Cataplexy is not present in 20%-40% of narcolepsy cases (8). IH is a disease with unknown etiology characterized with difficulty in wakening in the morning and unrestful sleep. Although its prevalence is not known exactly, it has been proposed that it ranges between 1/10 and 1/2. According to American

Correspondence Address/Yazışma Adresi
Murat Erdem MD, Gülhane Military Medical Academy, Division of Psychiatry, Ankara, Turkey
Gsm: +90 536 13 51 E-mail: drmerdem@yahoo.com **Received/Geliş tarihi:** 02.06.2012 **Accepted/Kabul tarihi:** 06.12.2012

Classification of Sleep Disorders (ICSD-2), it is divided into two groups as IH with long sleep period and IH with normal sleep period (9).

According to ICSD-2 criteria, to make a diagnosis of narcolepsy, cataplexy should be present in addition to presence of daytime sleep attacks for at least 3 months or sleep for at least 6 hours should be demonstrated by PSG in individuals without cataplexy and the mean sleep latency should be at least 8 minutes in multiple sleep latency test (MSLT) performed afterwards and presence of SOREM (sleep onset REM) should be demonstrated in at least two studies. According to the same diagnostic criteria, the diagnosis of IH is made with increased sleepiness at daytime for at least three months, a mean sleep latency shorter than 8 minutes in MSLT and presence of SOREM in at least one study. IH cases with a night sleep longer than 10 hours are called IH with long sleep time and IH cases with a night sleep of 6-10 hours are called IH without long sleep time (10). According to these criteria MSLT has an important role in the differential diagnosis of these two diseases. There are limited number of studies which have investigated the clinical and polysomnographic differences of narcolepsy and IH. Takei et al. found that the subjects with cataplexy (+) narcolepsy (C(+) NC) wakened more frequently at night compared to the subjects with cataplexy (-) narcolepsy (C(-) NC) in polysomnogrpahic examination and sleep latency in MSLT was longer in the subjects with IH compared to both narcolepsy groups (11). Anderson et al. found that sleep efficiency was lower, deep stage sleep was shorter and mean MSLT latency was shorter in the subjects with NC compared to the subjects with IH (12). In this study, it was aimed to compare polysomnography and MSLT findings between patients with C(+) and (-) NC and patients with IH with normal sleep time.

Method

The files of the patients who presented to Gülhane Military Medical Faculty Sleep Investigation Center between January 1995 and December 2009 with the complaint of excessive sleepiness were examined. Anamnesis, PSG and MSLT recordings were evaluated. A total of 283 patients who met the ICSD-2 diagnostic criteria were included in the study (94 C(+) NC, 49 C(-) NC and 140 patients with IH with normal sleep time). MSLT recordings in all patients were done after a single night PSG. The data of the patients who used any psychotropic drug and who were found to have slept shorter than 6 hours in PSG study were not included in the assessment. In addition, patients with sleep apnea syndrome, circadian rhythm disorder and movement disorder during sleep were also excluded. PSG and MSLT were performed using standard sytems (GRASS Model 78-tip analog system and Somnostar Alpha Sleep Polysomnography). In PSG examination, 4-channel electroencephalogram (EEG) (C3-A2, C4-A1, O1-A2 and O2-A1), 2-channel electrooculogram (EOG), mandibular muscle electromyography (EMG), electrocardiography (ECG), oral and nasal airflow, thoracic and abdominal respiratory movements, tibialis anterior muscle EMG, percutaneous O2 saturation were measured. Scoring of sleep stages was done according to the criteria of the American Academy of Sleep Medicine (13). PSG examinations performed before the publication of these criteria were scored according to the Rechtschaffen and Kales criteria (14). MSLT was performed with 2-hour intervals throughout the day (10.00, 12.00, 14.00, 16.00) and for 20 minutes according to the standard protocol (15). SOREM (Sleep onset REM) was defined as detection of the REM period in the first 20 minutes after the onset of sleep in PSG scoring and in the first 15 minutes after onset of sleep in MSLT.

Statistical Analysis

Continuous variables were presented as mean ± standard deviation, categorical variables were presented as numbers and percentages. The ages, body mass indexes (BMI), Epworth Sleepiness Scale scores and PSG and MSLT values of the cataplexy (+) and (-) NC groups and the IH group were compared using multi-factor analysis of variance where parametric conditions were met and using Kruskall-Wallis test where parametric conditions were not met. Post hoc evaluation was made using Tukey test when differences were found between the groups using ANOVA. The comparison of the three groups in terms of gender was done using chi-square test. A p value of ≤0.05 was considered statistically significant.

Results

Table 1. shows the comparison of the three groups in terms of age, gender, education level, Epworth sleepiness Scale score and BMI.

It was found that there was no difference between the three groups in terms of age, gender, Epworth sleepiness Scale score and BMI.

Table 2. shows the comparison of the three groups in terms of PSG values and MSLT SOREM number.

Table 1. Comparison of the NC and IH groups in terms of age, gender, Epworth Sleepiness Scale score and BMI

Variable	C(+) NC (I) (n=94)	C(-) NC (II) (n=49)	IH (III) (n=140)	Statistics
Age	25.10±7.46	24.92±7.50	26.90±6.84	2.20a
Gender (Male,%)	88.3	91.8	80.0	4.99b
Epworth score	18.44±2.93	17.74±3.65	18.20±3.33	0.75a
BMI (kg/m²)	25.24±3.89	25.30±4.20	24.21±3.66	2.30a

a: ANOVA test value (F), b:Chsquare test value, BMI: Body Mass Index, *: p>0.05

It was found that sleep latency and REM latency were longer in the IH group compared to the C(+)NC and C(-)NC groups and the MSLT SOREM number was higher in the C(+) NC group compared to the C(-)NC group.

Figure 1. shows mean sleep latency values of the three groups detected by MSLT.

The shortest mean MSLT sleep latency was found in the C(+)NC group and the longest mean MSLT sleep latency was found in the IH group (f=45.15).

Discussion

In this study, it was aimed to compare the PSG and MSLT values of C(+) NC, K(-)NC and IH subjects. It was found that sleep latency and REM latency were longer in the IH group compared to the NC groups and the shortest MSLT sleep latency was found in the C(+) NC group.

The fact that sleep latency was longer in the IH group (mean 39.85 min) compared to the C(+) and (-)NC groups (4.80 and 6.45 min, respectively) shows that these patients began sleeping later. This finding is not compatible with the study of Takei et al.. In this study in which 52 C(+), 62 C(-) NC patients were compared with 50 IH patients, no difference was found in sleep latency between the three groups (6.5, 8.3 and 7.7 min, respectively) (11). In the study of Anderson et al. in which 77 IH patients were evaluated as a single group without dividing the patients according to the sleep period at night, no difference was found in sleep latency between the IH and NC groups (11.5, 10.2 min, respectively) (12). In the study performed by Vernet et al. which included 75 IH patients, no difference was found in sleep latency between IH patients with long and normal sleep time and the control group (35, 26 and 32 min, respectively) (16). These findings suggest that sleep latency which is consistently short in NC patients shows variance individually or during the course of the disease in IH patients.

The fact that REM latency was shorter and the rate of SOREM was higher in the NC groups compared to the IH group showed that REM period started early in NC patients and no such change occurred in IH patients. However, no difference was found between the three groups in terms of REM percentage. This finding is compatible with the findings of Takei et al. (11). In the study performed by Vernet et al., REM latency was not found to be different in IH patients with normal sleep period (82.8±53.2 min) from the control group (16). These findings showed that REM latency was not different in IH patients compared to healthy controls and was shorter in NC patients.

In our study, no difference was found between the three groups in terms of deep stage sleep and awakening number. However, Takei et al. found the excitation index and

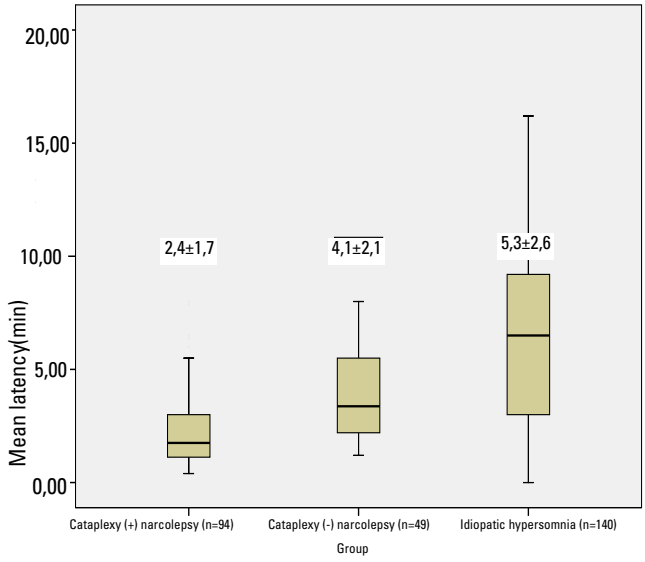

Figure 1. Comparison of the C(+) NC, C(-) NC and IH groups in terms of MSLT mean sleep latency

Table 2. Comparison of the NC and IH groups in terms of PSG and MSLT values

Variable	C(+) NC (I) (n= 94)	C(-) NC (II) (n=49)	IH (III) (n=140)	Statistics	Comparison
TST (min	570.7±150.4	566.1±165.2	608.9±137.0	2.1a. p=0.35	------
Sleep efficiency (%)	91.6±6.3	91.1±5.4	89.5±9.2	4.6b. p=0.01	-----
Sleep latency (min)	4.8±5.8	6.5±6.1	39.9±84.0	76.6a. p<0.001	I=II<III
REM latency (min)	29.6±55.4	26.9±46.6	99.0±27.6	154.1a. p<0.001	I=II<III
Stage 1 (%)	2.0±2.2	2.0±2.9	2.4±2.7	1.8a. p=0.41	-----
Stage 2 (%)	64.0±10.4	61.9±10.3	62.3±11.8	0.6b. p=0.58	-----
Stage 3+4 (%)	17.1±9.0	16.5±9.4	18.7±9.7	2.4a. p=0.22	-----
REM (%)	16.9±5.6	18.7±4.2	16.6±6.6	2.5a. p=0.22	-----
Number of awakenings	16.7±8.7	17.0±7.3	15.8±8.9	1.5a. p=0.47	-----
SOREM(+) (PSG) (n,%)	(73. 77.7)	(38. 77.6)	(2. 1.7)	182.1c. p<0.001	I=II>III
SOREM (MSLT)(mean±s.d)	3.3±0.8	3.0±0.9	0.3±0.5	641.a. p<0.001	I>II>III

a: Kruskall-Wallis test value, b: ANOVA test value (F), c: Chi-square, TST: Total sleep time

the percentage of stage I to be higher in C (+) NC group compared to the other two groups. Although this showed that night sleep in C(+) NC patients was split frequently, no difference could be found between the three groups in terms of deep stage sleep in this study similar to our findings (11). Anderson et al. found the percentage of deep sleep to be lower in the NC group compared to the IH group (12). This difference between the findings may be related with factors including different sample sizes and presence or absence of administration of treatment.

The finding that mean MSLT sleep latency was shorter in the C(+) NC group compared to the C(-) NC and IH groups was compatible with some studies (11). In the study in which the mean MSLT sleep latencies of the NC, IH and healthy control groups were compared, the shortest sleep latency was found in the NC group (17). In another study performed by Martinez-Rodriguez et al., it was found that the mean MSLT sleep latency was shorter only in the C(+) CN group compared to the IH group and there was no difference between the C(+) NC and C(-) NC groups and between the C(-) NC and the IH groups (18). It was interpreted such that this result was related with the small size of the study sample.

The absence of PSG and MSLT results of the healthy control group was interpreted as the limitation of the study. In addition, the fact that cyclic alternating pattern evaluation which is another indicator of excitation in PSG examination was not performed is another limitation. We think that future studies should investigate if there is a difference between the responses to psychostimulant therapies including modafinil in NC and IH patients.

References

1. Ohayon MM, Priest RG, Zulley J, Smirne S, Paiva T. Prevalence of narcolepsy symptomatology and diagnosis in the European general population. Neurology 2002; 58:1826-1833.
2. Silber MH, Krahn LE, Olson EJ, Pankratz VS. The epidemiology of narcolepsy in Olmsted County, Minnesota: a population-based study. Sleep 2002; 25:197-202.
3. Wing YK, Li RH, Lam CW, Ho CK, Fong SY, Leung T. The prevalence of narcolepsy among Chinese in Hong Kong. Ann Neurol 2002; 51:578-584.
4. Honda Y. Census of narcolepsy, cataplexy and sleep life among teenagers in Fujisawa city. Sleep Res 1979; 8:191.
5. Tashiro T, Kanbayashi T, Lijima S. An epidemiological study on prevalence of narcolepsy in Japanese. J Sleep Res 1992; (Suppl 1):228.
6. Lavie P, Peled R. Letter to the editor: narcolepsy is a rare disease in Israel. Sleep 1987; 10:608-609.
7. Durukan İ, Erdem M, Öz O, Karaman D, Özgen F. Çocukluk çağında narkolepsi: olgu sunumu. Nöropsikiyatri Arşivi 2011; 48:158-161.
8. Billiard M. Diagnosis of narcolepsy and idiopathic hypersomnia. An update based on the International Classification of Sleep Disorders, 2nd edition. Sleep Med Rev 2007; 11:377-388.
9. Frenette E. Kushida CA. Primary Hypersomnias of Central Origin. Semin Neurol 2009; 29:354-367.
10. Sateia MJ, editor. American Academy Sleep Medicine. The International Classification of Sleep Disorders, 2nd ed. Diagnostic and Coding Manual. Westchester, IL: American: Academy Sleep Medicine, 2005.
11. Takei Y, Komada Y, Namba K, Sasai T, Nakamura M, Suqiura T, Hayashida K, Inoue Y. Differences in findings of nocturnal polysomnography and multiple sleep latency test between narcolepsy and idiopathic hypersomnia. Clin Neurophysiol 2012; 123:137-141.
12. Anderson KN, Pilsworth S, Sharples LD, Smith IE, Shneerson JM. Idiopathic Hypersomnia: A Study of 77 Cases. Sleep 2007; 30:1274-1281.
13. Iber C, Ancoli-Israel S, Chesson A, Quan S. The AASM manual for the scoring of sleep and associated events: rules, terminology and technical speciŞcations for the American Academy of Sleep Medicine. 1st ed. Westchester, IL: American Academy of Sleep Medicine; 2007.
14. Rechtschaffen A, Kales A. A manual of standardized terminology, techniques and scoring system for sleep stages in human subjects. Washington, DC: US Government Printing Office; 1968.
15. Carskadon MA, Dement WC, Mitler MM, Roth T, Westbrook PR, Keenan S. Guidelines for the multiple sleep latency test (MSLT): a standard measure of sleepiness. Sleep 1986; 9:519-524.
16. Vernet C, Arnulf I. Idiopathic hypersomnia with and without long sleep time: a controlled series of 75 patients. Sleep 2009; 32:753-759.
17. Komada Y, Inoue Y, Mukai J, Shirakawa S, Takahashi K, Honda Y. Difference in the characteristics of subjective and objective sleepiness between narcolepsy and essential hypersomnia. Psychiatry Clin Neurosci 2005; 59:194-199.
18. Martínez-Rodríguez JE, Iranzo A, Casamitjana R, Graus F, Santamaria J. Análisis comparativo de un grupo de pacientes con narcolepsia-cataplejía, narcolepsia sin cataplejía e hipersomnia idiopática. Med Clin (Barc) 2007; 128:361-365.