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Discrepancy between subjective and objective sleepiness in adolescents

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ABSTRACT

Background: Lack of correlation between subjective and objective measurements of daytime sleepiness is common. Here, the frequency of discrepancy between subjective and objective sleepiness, as well as possible predictors, were examined for an adolescent cohort.
Methods: This study included pediatric patients (aged 10–18 years, n = 211) with various sleep disorder symptoms were evaluated between August 2011 and February 2021. Subjective and objective sleepiness were assessed based on eleven or more scores of the Japanese version of Epworth Sleepiness Scale and a mean sleep latency of 8.0 min or less on the Multiple Sleep Latency Test, respectively. Patients were then classified as both subjectively and objectively sleepy, objectively sleepy, subjectively sleepy, and non-sleepy. Discrepancy-related factors were identified with multivariable logistic regression analysis.
Results: The frequency of discrepancy between subjective and objective sleepiness was 46.4%, with 35.5% (75/211) of the patients exhibiting subjective sleepiness without objective sleepiness and 10.9% (23/211) of the patients exhibiting objective sleepiness without subjective sleepiness. Co-existence of neurodevelopmental disorders was associated more often with subjective sleepiness compared to non-sleepiness (odds ratio (OR), 4.12; 95% confidence interval (CI), 1.30 to 12.99) or concordant sleepiness (OR, 7.54; 95% CI, 2.43 to 23.38).
Conclusions: Nearly half of the patients exhibited discrepancy between subjective and objective sleepiness, and it more often involved subjective sleepiness. Furthermore, age, bedtime, and neurodevelopmental disorders were identified as significant factors related to subjective sleepiness without objective sleepiness.

1. Introduction

Discrepancy between subjective and objective sleepiness is paradoxical in clinical practice. For example, a person can be assessed as excessively sleepy by a self-reported screening tool, and not sleepy according to an objective test. Conversely, pathological sleepiness measured by an objective test may not be confirmed by a self-reported screening tool. Typically, when an individual feels sleepy, they recognize the condition and take corresponding measures (e.g., take a nap or seek stimulus). However, under the condition of objective sleepiness without subjective sleepiness, it is not likely that an individual will seek help or take precautionary measures because they are unaware that they are sleepy. Moreover, the latter condition represents a safety issue. On the other hand, when excessive subjective sleepiness without objective sleepiness is observed in clinical practice, patient management is an issue. For example, self-perception of excessive daytime sleepiness cannot be an adequate feature to fulfill the diagnostic criteria for some of the central disorders of hypersomnolence (narcolepsy and idiopathic hypersomnia) when pathological sleepiness is not confirmed on objective measures even in patients with obvious clinical presentation of the disease.

Excessive daytime sleepiness (EDS) is one of the most common complaints among patients who visit sleep centers and the cardinal symptom of central disorder of hypersomnolence. But other sleep-related conditions and neurodevelopmental disorders, are also

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associated with EDS in children and adolescents [1,2]. Furthermore, adolescents are more vulnerable to EDS due to the physiological changes they undergo and environmental factors [3]. Unfortunately, sleepy adolescents are prone to being regarded as lethargic or disinterested, while prepubertal children often manifest EDS as hyperactivity and irritability [1], and EDS is associated with declines in school performance [4,5]. Due to these consequences of EDS, adolescents often experience difficulties in maintaining their daily life. Consequently, proper assessment of sleepiness and making an earlier diagnosis of a disease is crucial and can prevent further complications in this population.

In sleep practice, the most widely used subjective tool to define sleepiness is the Epworth Sleepiness Scale (ESS). This self-administered questionnaire includes eight questions which address the chance of dozing off in various situations [6]. Meanwhile, the Multiple Sleep Latency Test (MSLT) is considered the gold standard for assessing objective sleepiness. It includes a series of five nap opportunities which are scheduled at 2-h intervals in a clinical setting [7]. It has been reported that the results of the ESS and MSLT do not exhibit a strong enough correlation, with poor to moderate correlations observed in adult studies [8,9].

There are several factors which may contribute to a lack of strong correlation. In fact, subjective sleepiness and objective sleepiness are not exactly parallel [10]. Thus, the ESS and MSLT evaluate different aspects of daytime sleepiness [11]. Other factors to consider regarding the performance of these assessments include: limited sample size, inappropriate methods of study, underestimated cut-off values [12,13], and co-existing psychiatric disorders [14,15]. Moreover, a comparison of correlation and discrepancy between these two measurements in adolescents has not been made.

Therefore, the aims of the present study are to identify: a) how often discrepancy between subjective and objective sleepiness occurs, and b) what patient factors may be related to this discrepancy in adolescent populations.

2. Methods

2.1. Participants

This retrospective study included 211 pediatric patients who were aged 10–18 years at their first evaluation of both subjective and objective measurements. These patients underwent the MSLT and completed a Japanese version of the Epworth Sleepiness Scale (JESS) at the Center for Sleep Medicine of Ehime University Hospital between August 2011 and February 2021. In addition, since this sleep center receives referrals from the other community hospitals and clinics in surrounding regions, as well as from other departments within the hospital, children who did not initially complain about daytime sleepiness also underwent polysomnography (PSG) at the center and completed the MSLT, with the concurrence of their parent. Thus, data from children who were not subjectively sleepy but objectively sleepy was available. Participants were categorized into the following four groups according to established clinical cutoff values for JESS scores and mean sleep latency (MSL) on the MSLT. These groups included: 1) concordantly sleepy (CS) - sleepy according to both the JESS (score $\geq$11) and the MSLT (MSL $\leq$8.0 min), 2) objectively sleepy (OS) - objective sleepiness without subjective sleepiness (JESS score <11, MSL $\leq$8.0 min), 3) subjectively sleepy (SS) - subjective sleepiness without objective sleepiness (JESS score $\geq$11, MSL >8.0 min), and 4) non-sleepy (NS) - not sleepy according to the JESS and MSLT (JESS score <11, MSL >8.0 min). This stratified group approach allowed patient-related factors leading to discrepancy between subjective and objective sleepiness to be identified.

Sleep disorders were diagnosed according to the International Classification of Sleep Disorders (ICSD), 2nd or 3rd edition depending on the timing of evaluation. We reviewed the medical record and confirmed the diagnosis to fit with the current diagnostic criteria. Regarding comorbid disorders, 19.9% (42/211) of the children exhibited neurodevelopmental disorders. These included: autism spectrum disorders (ASD) (11.8%, 25/211), attention deficit hyperactive disorders (ADHD) (3.3%, 7/211), ASD and ADHD (3.3%, 7/211), and other developmental disorders (1.4%, 3/211). Some of the patients (16.1%) also had ear-nose-throat (ENT) problems (e.g., adenoid hypertrophy, nasal allergies, and small jaw). Of total, 15.6% (33/211) of the patients were taking medications at the first visit to our practice, including stimulants (7 patients), H-1 antihistamines (5 patients), modafinil (4 patients), Chinese medicine (4 patients), antidepressants (2 patients). The protocol for this study was approved by the Institutional Review Board at Ehime University Hospital.

2.2. Subjective assessment

ESS is one of the most widely used scales for evaluating subjective sleepiness. It is a simple self-rating questionnaire which requires patients to estimate their chance of dozing off during eight different activities. In the present study, subjective sleepiness was assessed with the JESS. Japanese translation of ESS initially included questions regarding alcohol usage and driving-related questions, official translation by the Japanese investigators avoided these questions so that the JESS fit for wider range of population and has been broadly used to assess sleepiness in adults and adolescents in clinical practices in Japan [16]. Prior to conducting sleep study, all of our patients completed the JESS. In order to enhance more accurate response to the questions of JESS, we advised caregivers to assist and help children to answer the questions in JESS as necessary. Patients also answered the following three sleep habit-related questions obtained from the Pittsburg Sleep Quality Index: *1.During the past month, what time have you usually gone to bed at night?; 2.During the past month, what time have you usually gotten up in the morning?; 3.During the past month, how many hours of actual sleep did you get at night?* Bedtime and wake time on weekdays and weekends were extracted from sleep diary.

2.3. Objective assessment

Prior to the MSLT, all of our patients underwent a nocturnal PSG sleep study. PSG was performed in our sleep laboratory according to guidelines of the American Academy of Sleep Medicine (AASM) [17,18]. A trained sleep technologist was present and recorded: six electroencephalograms (F4-M1, F3-M2, C4-M1, C3-M2, O2-M1, O1-M2), right and left electrooculograms, submental and bilateral anterior tibialis electromyograms, electrocardiogram, pulse oximeter and respiratory channels (oronasal thermistor, nasal air pressure transducer, thoracic and abdominal strain gauges, and snoring microphone). The montage of the MSLT was similar to that of the PSG, except that the patients did not wear the respiratory channels. MSLT was performed immediately following PSG according to the recommended protocol described in the Practice Parameter defined by the AASM (2005) [19]. The initial nap opportunity was started 1.5–3 h after termination of PSG recording. Patients were instructed to keep a sleep diary at least a week prior to PSG/MSLT to evaluate sleep-wake schedule. Medications that may affect the study result such as stimulants, stimulant like medications, and REM suppressing medications were discontinued prior to the study. Instructions were given to avoid vigorous physical activity, stimulating activities, caffeinated beverages, and unusual sunlight exposure during the studies. The MSLT consists of five naps which are consecutively administered 2-h apart on the same bed in a quiet, darkened room. If sleep does not occur, the nap session is terminated within 20 min and a 20-min sleep onset latency is recorded for the session. If during this time frame the patient does fall asleep, they are allowed to maintain the nap for 15 min (from onset of sleep). MSL was averaged from five naps, and values $\leq$ 8.0 min were defined as EDS according to the International Classification of Sleep Disorders (3rd edition) [14]. Procedure of the MSLT was performed by experienced technician. Adolescent patients, especially patients with neurodevelopmental disorders were carefully observed if the patient followed instructions during the study. All events potentially affected the study result was carefully reviewed. Recording with at least four successful nap opportunities was considered as a completed MSLT result.

2.4. Statistical analysis

Both the CS and NS groups were treated as reference groups in our analyses. Both groups exhibited no discrepancy of sleepiness and results of the tests were matched perfectly. However, the groups were not completely interchangeable since they included very different features of pathological sleepiness or non-sleepiness. Correlations between sleep measures were assessed according to Spearman correlation coefficients and Point-Biserial correlation (r_{pb}). Categorical variables were compared by using Chi-square test. Age-adjusted *p* values for self-reported measures (bed time, wake time, and sleep duration) and PSG measures (total sleep time, sleep latency, sleep efficiency, wake after sleep onset (WASO), sleep stage percentages, rapid eye movement (REM) sleep onset latency, apnea-hypopnea index (AHI), and arousal index) were calculated and compared across the four groups with analysis of variance. Pairwise multiple comparisons between groups were calculated with post-hoc tests. The Kruskal-Wallis test was applied to continuous data exhibiting a non-normal distribution. Multicollinearity was tested through variance inflation factors and tolerance. Multivariable logistic regression analysis was conducted as a generalized logit model to define associations between predictors (e.g., age, gender, self-reported bed time, self-reported wake time, self-reported sleep duration, bedtime difference, wake time difference, sleep efficiency, arousal index, AHI, and neurodevelopmental disorders) and a response variable (sleepiness status). In this model, 16 observations were deleted due to missing values for the explanatory variables. Results with P values < 0.05 were considered significant. Analyses were performed with SAS OnDemand for Academics software (SAS Institute, Cary, NC, USA).

3. Results

The mean age of the participants (n = 211) in this study was 14.8 y (SD 1.9), and 63.03% were male. The MSL was 9.54 min (SD 5.13) and the mean JESS score was 12.76 (SD 5.23). Spearman's correlation between JESS scores and MSL was −0.23 ($p = 0.0006$). There was a positive correlation between JESS score and EDS complaint, $r_{pb} = 0.47$ ($p < 0.0001$).

Discrepancy between subjective and objective sleepiness was identified in 35.5% (75/211) of the patients with subjective sleepiness without objective sleepiness (SS group) and in 10.9% (23/211) of the patients with objective sleepiness without subjective sleepiness (OS group) in Table 1. Nearly half of the patients in this study exhibited discordant sleepiness (a discrepancy between subjective and objective sleepiness). The frequencies of concordant sleepiness (CS group) and non-sleepiness (NS group) were 33.2% (70/211) and 20.4% (43/211), respectively.

Characteristics of the four groups according to their chief complaints are summarized in Table 2. The predominant complaint was EDS in this population. Central disorders of hypersomnolence were common in the CS and OS groups compared to the SS and NS groups (Table 3). In contrast, insufficient sleep syndrome and unclassified EDS were more common in the SS group compared to the other groups. Delayed sleep phase disorder and sleep-related breathing disorders were also more common in the SS group compared to the CS group. Furthermore, the patients with insufficient sleep syndrome, unclassified EDS, long sleeper, sleep-related movement disorder, and neurodevelopmental disorders had subjective sleepiness according to the JESS score, while MSL remained within the normal range (Table 4).

Patient demographics and clinical characteristics of the four groups according to their sleepiness status are presented in Table 5. Most of the differences were observed between the CS and NS groups. However, for this study, we primarily focused on the differences between the OS and SS groups compared to the CS and NS groups as references, respectively. The age of the participants increased monotonically from the NS group to the CS group ($p < 0.0001$). In a pairwise comparison analysis, the SS group was characterized by younger participants ($p = 0.010$), longer duration of self-reported sleep ($p = 0.049$), lower sleep efficiency ($p = 0.013$), and higher WASO ($p = 0.025$) compared to the CS group. When

Table 1
Frequency of the discrepancy between subjective and objective sleepiness.

JESS Score	MSLT	
	Normal (MSL >8 min)	Objectively Sleepy (MSL ≤8 min)
Normal (<11)	43 (20.4%)	23 (10.9%)
Subjectively Sleepy (≥11)	75 (35.5%)	70 (33.2%)

JESS, Japanese Version of Epworth Sleepiness Scale; MSLT, Multiple Sleep Latency Test; MSL, Mean Sleep Latency.

Table 2
Chief complaints of the four groups, n (%).

	CS (N = 70)	OS (N = 23)	SS (N = 75)	NS (N = 43)
EDS	66 (94.3)	12 (52.2)	49 (65.3)	14 (32.6)
SDB symptoms	2 (2.9)	4 (17.4)	8 (10.7)	12 (27.9)
Unable to wake up	1 (1.4)	2 (8.7)	7 (9.3)	7 (16.3)
Sleep-wake rhythm issue	0	1 (4.3)	3 (4.0)	4 (9.3)
Insomnia	0	1 (4.3)	1 (1.3)	2 (4.7)
Long sleep	1 (1.4)	1 (4.3)	2 (2.7)	1 (2.3)
Sleep terror	0	0	0	3 (7.0)
Nightmare	0	0	1 (1.3)	0
Sleep walking	0	1 (4.3)	1 (1.3)	0
Leg discomfort	0	1 (4.3)	2 (2.7)	0
Movement sensation	0	0	1 (1.3)	0

CS, Concordantly sleepy; OS, Objectively sleepy; SS, Subjectively sleepy; NS, Non-sleepy; EDS, Excessive daytime sleepiness; SDB, Sleep-disordered breathing.

Table 3
Clinical diagnosis and other conditions according to group, n (%).

	CS (N = 70)	OS (N = 23)	SS (N = 75)	NS (N = 43)
Narcolepsy type 1	16 (22.9)	0	1 (1.3)	0
Narcolepsy type 2	32 (45.7)	5 (21.7)	1 (1.3)	0
Idiopathic hypersomnia	17 (24.3)	4 (17.4)	0	0
Insufficient sleep syndrome	2 (2.9)	4 (17.4)	21 (28.0)	3 (7.0)
Unclassified EDS	0	1 (4.3)	13 (17.3)	2 (4.7)
Long sleeper	0	1 (4.3)	5 (6.7)	3 (7.0)
Hypersomnia due to a medical disorder	0	0	1 (1.3)	0
Hypersomnia associated with a psychiatric disorder	0	1 (4.3)	2 (2.7)	0
Delayed sleep phase disorder	1 (1.4)	1 (4.3)	13 (17.3)	11 (25.6)
Sleep related breathing disorders	1 (1.4)	2 (8.7)	10 (13.3)	13 (30.2)
NREM parasomnia	0	1 (4.3)	1 (1.3)	2 (4.7)
Sleep related movement disorders	1 (1.4)	2 (8.7)	3 (4.0)	2 (4.7)
Inadequate sleep hygiene	0	0	3 (4.0)	2 (4.7)
Anxiety	0	0	1 (1.3)	0
School refusal	0	1 (4.3)	0	1 (2.3)
Within normal limit	0	0	0	4 (9.3)

CS, Concordantly sleepy; OS, Objectively sleepy; SS, Subjectively sleepy; NS, Non-sleepy; EDS, Excessive daytime sleepiness; NREM, Non-rapid eye movement.

Table 4
JESS score and MSL according to clinical diagnosis, mean ± SD.

	N	JESS score	MSL
Narcolepsy type 1	17	17.75 ± 3.17	3.32 ± 2.54
Narcolepsy type 2	38	14.74 ± 4.4	4.56 ± 1.89
Idiopathic hypersomnia	21	13.33 ± 4.03	4.78 ± 1.92
Insufficient sleep syndrome	30	13.77 ± 4.71	10.86 ± 3.56
Unclassified EDS	16	13.94 ± 4.34	11.84 ± 2.95
Delayed sleep phase disorder	26	9.92 ± 4.68	14.29 ± 3.99
Sleep-related breathing disorders	26	10.12 ± 5.14	13.37 ± 3.55
Long sleeper	9	12.44 ± 5.59	13.51 ± 4.42
Sleep-related movement disorders	8	11.29 ± 5.12	11.21 ± 5.01
Neurodevelopmental disorders	42	12.76 ± 4.95	11.35 ± 4.64

JESS, Japanese Version of Epworth Sleepiness Scale; MSL, Mean sleep latency; EDS, Excessive daytime sleepiness.

compared to the NS group, the SS group was only distinguished by an older age (p = 0.002). The OS group was characterized by an older age (p = 0.006), shorter duration of self-reported sleep (p = 0.024), higher sleep efficiency (p = 0.0003), and lower WASO (p = 0.008) compared to the NS group. In contrast, there were no significant differences between the OS group and the CS group. Overall, there was a tendency to wake up later on weekends than on weekdays. Difference between weekday and weekend for both bedtime and wake time did not differ among the groups. The MSLT measured five sleep latencies that reflected similar trends across the four groups, including a decrease in sleep latency from the NS group to the CS group. Sleep architecture was also relatively normal in all of the four groups. Meanwhile, there were no significant differences in stage N2, N3 and REM sleep between the SS and OS groups compared to the reference groups.

Multivariable logistic regression analysis was conducted using a smaller dataset (N = 195) to further investigate the observed discrepancies. The OS and SS groups were compared to the CS and NS groups, respectively (Table 6). When the CS group was set as the reference, younger age (odds ratio (OR) = 0.75, 95% confidence interval (CI): 0.58 to 0.97), lower sleep efficiency (OR = 0.93, 95% CI: 0.87 to 0.99), and neurodevelopmental disorders (OR = 7.54, 95% CI: 2.43 to 23.38) were significantly associated with subjective sleepiness. When the NS group was set as the reference, later bedtime (OR = 2.56, 95% CI: 1.28 to 5.15) and neurodevelopmental disorders (OR = 4.12, 95% CI: 1.30 to 12.99) were associated with subjective sleepiness. For objective sleepiness, only higher sleep efficiency (OR = 1.21, 95% CI: 1.04 to 1.40) was associated with objective sleepiness compared to non-sleepiness. In subgroup analyses, ASD and ADHD were added separately. Only ASD (SS vs. CS: OR = 17.23, 95% CI: 3.46 to 85.9; SS vs. NS: OR = 5.36, 95% CI: 1.43 to 20.03) exhibited statistical significance in the multivariable regression model. In contrast, ADHD was not related with discrepancy (SS vs. CS: OR = 0.44, 95% CI: 0.07 to 2.66; SS vs. NS: OR = 1.94, 95% CI: 0.32 to 11.74).

4. Discussion

Evaluation of excessive daytime sleepiness is of clinical importance in the management of sleep disorders. Despite the number of studies which have investigated correlations between subjective and objective sleepiness in adults [11–13], limited research has addressed the discrepancy between subjective and objective sleepiness in adolescents.

Initially, we observed that nearly half of the patients examined in this study exhibited a discrepancy between subjective and objective sleepiness. Thus, almost one in two adolescents which visit the sleep center are likely to have a discrepancy in their evaluation of sleepiness when only the MSLT or JESS is used. Moreover, one-third of our participating patients had discrepancy of subjective sleepiness, while only 10.9% of the participating patients had discrepancy of objective sleepiness. While the latter appears less likely to occur, it still requires investigation. In patients with OSA, objective but not subjective EDS is associated with inflammation, suggesting that objectively and subjectively measured EDS most likely reflect different central nervous system processes [20]. Systemic inflammation was associated with objective sleepiness, findings not observed with subjective sleepiness in patients with heart failure [21]. These findings suggest that underlying mechanistic pathways of inflammation may provide the explanation for this objective sleepiness, but not for the subjective sleepiness.

A second finding of the present study involves potential factors related to discrepancy between subjective and objective sleepiness. These factors include: age, bedtime, sleep efficiency, and neurodevelopmental disorders. Patient age increased from the NS to the CS groups, and a significant association between younger age and subjective sleepiness without objective sleepiness was identified with multivariable regression analysis. It is reported that sleepiness is affected by age [22]. Age has also been identified as a significant predictor for certain screening instruments, including the ESS, when assessing subjective sleepiness [23]. Carskadon et al. demonstrated that older adolescents exhibit a significantly greater tendency to sleep on the MSLT than their younger peers [24]. However, it is also possible that pubertal maturation is another

Table 5
Patient demographics and clinical characteristics according to group.

Demographics	CS (N = 70)	OS (N = 23)	SS (N = 75)	NS (N = 43)	p value
Age, y	15.7 ± 1.4	15.1 ± 2.1	14.8 ± 1.9	13.6 ± 1.9	<0.0001
Sex – male, n (%)	46 (65.7)	14 (60.9)	42 (56.0)	31 (72.1)	0.338
Sleep habit measures					
Bedtime					
Self-reported bedtime (PSQI)	23.7 ± 1.1	23.4 ± 1.3	23.3 ± 1.4	22.6 ± 0.9	<0.0001
Weekday	23.4 ± 1.0	23.4 ± 1.3	22.9 ± 1.1	22.5 ± 0.8	<0.0001
Weekend	23.6 ± 1.0	23.6 ± 1.1	23.0 ± 1.1	22.6 ± 1.0	<0.0001
Weekday-Weekend difference	0.23 ± 0.7	0.23 ± 0.55	0.09 ± 0.51	0.14 ± 0.49	0.162
Wake time					
Self-reported wake time (PSQI)	7.2 ± 1.6	7.1 ± 1.8	7.7 ± 2.2	7.7 ± 1.8	0.001
Weekday	7.0 ± 0.7	6.7 ± 0.6	6.9 ± 0.8	7.2 ± 1.1	0.010
Weekend	8.7 ± 1.6	8.3 ± 1.5	8.3 ± 1.5	8.4 ± 1.7	0.073
Weekday-Weekend difference	1.67 ± 1.45	1.66 ± 1.50	1.41 ± 1.26	1.25 ± 1.20	0.295
Sleep duration, hr (PSQI)	7.4 ± 1.8	7.3 ± 1.6	8.3 ± 2.4	8.8 ± 2.0	0.003
MSLT measures					
1st sleep latency, min	3.5 (1.5–5.0)	4.0 (3.0–6.5)	9.0 (6.5–14.5)	10.5 (7.5–20.0)	<0.0001
2nd sleep latency, min	3.0 (1.5–4.0)	3.0 (1.5–5.0)	9.0 (5.0–16.0)	13.0 (7.0–20.0)	<0.0001
3rd sleep latency, min	4.3 (2.5–6.0)	5.0 (3.0–7.5)	11.5 (7.5–17.0)	14.5 (8.5–20.0)	<0.0001
4th sleep latency, min	4.0 (2.0–6.5)	4.0 (2.0–7.0)	13.0 (10.0–20.0)	19.8 (13.5–20.0)	<0.0001
5th sleep latency, min	5.5 (2.5–8.3)	6.5 (3.0–9.0)	18.5 (12.5–20.0)	20.0 (16.5–20.0)	<0.0001
PSG measures					
Total sleep time, min	497.2 ± 61.5	496.9 ± 56.3	487.2 ± 74.4	460.6 ± 74.0	0.018
Sleep latency, min	7.9 ± 21.3	5.6 ± 5.1	11.5 ± 15.7	15.2 ± 18.8	0.083
Sleep efficiency, %	93.2 ± 6.1	94.5 ± 3.5	88.4 ± 10.9	84.5 ± 11.9	<0.0001
WASO (min)	30.3 ± 28.4	23.2 ± 17.0	53.8 ± 63.2	64.7 ± 55.6	0.0006
REM sleep onset latency, min	100.8 ± 60.5	96.7 ± 57.7	131.8 ± 68.4	139.2 ± 77.8	0.003
Stage N1, %	6.7 ± 4.7	4.8 ± 3.1	6.5 ± 3.7	6.6 ± 4.1	0.165
Stage N2, %	50.2 ± 7.8	48.4 ± 8.9	50.7 ± 7.9	47.4 ± 9.9	0.007
Stage N3, %	21.8 ± 7.8	26.4 ± 9.6	23.5 ± 8.1	28.2 ± 10.3	<0.0001
Stage R, %	21.1 ± 4.8	20.3 ± 3.7	19.3 ± 4.3	17.8 ± 5.1	0.006
Arousal index	11.2 ± 4.3	10.9 ± 4.7	11.0 ± 5.1	11.5 ± 6.1	0.148
AHI	3.0 ± 2.5	3.1 ± 2.6	3.0 ± 3.4	3.2 ± 3.8	0.927
Comorbid disorders, n (%)					
Neurodevelopmental disorders	5 (7.1)	3 (13.0)	26 (34.7)	8 (18.6)	0.0004
ENT problems	7 (10.0)	5 (21.7)	12 (16.0)	10 (23.3)	0.251

CS, Concordantly sleepy; OS, Objectively sleepy; SS, Subjectively sleepy; NS, Non-sleepy; PSQI, Pittsburg Sleep Quality Index; MSLT, Multiple sleep latency test; PSG, Polysomnography; WASO, Wake after sleep onset; REM, Rapid eye movement.
AHI, Apnea-hypopnea index; ENT, Ear, nose, throat.
ANOVA and Kruskal-Wallis tests applied to continuous data; Chi-square test applied to categorical data.
Results are presented as mean ± SD and median (Q1-Q3). P values reflect analysis adjusted for age effects.

Table 6
Predictors of discrepancy between subjective and objective sleepiness.

	Reference: CS (N = 67)				Reference: NS (N = 39)			
	OS (N = 19)		SS (N = 70)		OS (N = 19)		SS (N = 70)	
Parameter[†]	OR	(95% CI)	OR	(95% CI)	OR	(95% CI)	OR	(95% CI)
Age	0.78	(0.55–1.12)	0.75	(0.58–0.97)*	1.33	(0.89–1.97)	1.27	(0.94–1.70)
Sex, male	0.64	(0.20–2.11)	0.71	(0.30–1.66)	0.48	(0.12–1.92)	0.53	(0.19–1.46)
Bedtime (PSQI)	0.49	(0.21–1.10)	0.79	(0.47–1.34)	1.57	(0.60–4.07)	2.56	(1.28–5.15)**
Wake time (PSQI)	1.47	(0.74–2.90)	1.44	(0.94–2.18)	0.58	(0.28–1.20)	0.56	(0.35–1.91)
Sleep duration (PSQI)	0.64	(0.35–1.14)	0.97	(0.69–1.36)	0.93	(0.49–1.78)	1.41	(0.91–2.20)
Bedtime difference	0.65	(0.26–1.62)	0.47	(0.20–1.10)	1.83	(0.54–6.20)	1.34	(0.50–3.57)
Wake time difference	1.19	(0.80–1.76)	0.89	(0.64–1.22)	1.52	(0.91–2.54)	1.13	(0.77–1.67)
Sleep efficiency	1.09	(0.94–1.26)	0.93	(0.87–0.99)*	1.21	(1.04–1.40)*	1.03	(0.99–1.08)
Arousal index	0.97	(0.84–1.12)	0.96	(0.87–1.06)	1.00	(0.85–1.19)	0.99	(0.88–1.11)
AHI	1.07	(0.88–1.31)	0.98	(0.84–1.15)	1.11	(0.87–1.41)	1.02	(0.87–1.20)
Neurodevelopmental disorders, yes	1.36	(0.23–8.08)	7.54	(2.43–23.38)***	0.74	(0.11–4.85)	4.12	(1.30–12.99)*

CS, Concordantly sleepy; OS, Objectively sleepy; SS, Subjectively sleepy; NS, Non-sleepy; AHI, Apnea-hypopnea index;
OR, Odds ratio; CI, Confidence interval. PSQI, Pittsburg Sleep Quality Index. Bedtime difference represents the difference between weekday and weekend for bedtime. Wake time difference represents the difference between weekday and weekend for wake time. Sixteen observations were deleted due to missing values for the predictor variables.
[†]Generalized Logit Type 3 Analysis of Effects; Degrees of freedom = 3; *p < 0.05, **p < 0.01, ***p < 0.001.

internal factor to consider for adolescents [25]. The question also arises whether a child first becomes subjectively sleepy, objectively sleepy, or the two features appear simultaneously. Another possibility is that a discrepancy of sleepiness may occur temporarily while an individual transitions from non-sleepiness to pathological sleepiness. It is reported that narcolepsy, a chronic disorder mainly characterized by EDS, takes about ten years to be diagnosed and pediatric onset of symptom was one of the predictors of this delayed diagnosis [26]. We suggest that discrepancy between subjective and objective sleepiness may be one of the factors related to the delayed diagnosis, which needs to be studied in further research. From a clinical perspective, it is important to observe how children with discordant sleepiness are affected over time. Further longitudinal studies are also needed to clarify the evolution of subjective and objective sleepiness during an individual's progression to maturity.

Adolescents are more likely to report delayed sleep and wake times than younger peers due to many intrinsic and extrinsic factors [27]. A major consequence of delayed sleep time is daytime sleepiness. In the present study, later bedtime was associated with subjective sleepiness. One study suggested that sleepiness is related to self-reported bedtime because a circadian phase shift produces both later bedtime and daytime sleepiness, and later bedtime accurately reflect the slower accumulation of sleep need [22]. We hypothesize that later bedtime contributes to subjective sleepiness in children; yet does not significantly influence MSL.

Higher sleep efficiency was associated with objective sleepiness compared to non-sleepiness, while lower sleep efficiency was associated with subjective sleepiness compared to concordant sleepiness. Notably, sleep efficiency was higher in both the CS and OS groups than in the SS and NS groups. Most of the patients diagnosed with central disorders of hypersomnolence were included in the CS and OS groups (Table 3). This result is consistent with the results of other studies where patients with central disorders of hypersomnolence (e.g., idiopathic hypersomnia) exhibited increased sleep efficiency [28,29] compared with subjectively sleepy controls [30]. Therefore, we speculate that the present results are more related to the pathophysiology of hypersomnolence disorders rather than the factor associated with discrepancy of objective sleepiness.

Excessive daytime sleepiness is one of the most pervasive and common sleep problems that present in individuals with neurodevelopmental disorders [31,32]. During childhood, ASD and ADHD are the most commonly diagnosed neurodevelopmental disorders [33]. Previously, a meta-analysis observed discrepancies between subjective and objective sleep measures in both children with ASD and children with ADHD [34]. In the present study, neurodevelopmental disorders was associated with subjective sleepiness without objective sleepiness compared to both the CS and NS reference groups. Furthermore, when analyses were performed for ASD and ADHD cases separately, the ASD cases only maintained a statistically significant association with subjective sleepiness. Two other studies have reported that older children and adolescents with ASD exhibit subjective sleepiness during the daytime [35,36]. A cohort study of 1859 children with ASD and typical development (TD) identified a higher frequency of problems associated with subjective daytime sleepiness [35]. Children with ASD also exhibit markedly greater subjective daytime sleepiness with age than children without ASD [36]. Although both studies used a Children's Sleep Habits Questionnaire to assess daytime sleepiness rather than the ESS, adolescents with ASD exhibited greater subjective sleepiness than their peers with TD. To date, the MSLT and subjective sleepiness scale have not been compared for adolescents with ASD. However, we did find one study which included data from both ESS and MSLT assessments to evaluate daytime sleepiness in children with ADHD [37]. A correlation between ESS scores and MSLT results was reported to be similar in both their ADHD and TD groups, yet did not significantly correlate in children with ADHD ($r = -0.31$, $p > 0.05$), and did significantly correlate in TD children ($r = -0.31$, $p < 0.05$) [36]. Cortese et al. has emphasized that even though a limited number of studies have provided data regarding daytime sleepiness, high heterogeneity in both objective studies with MSLT and subjective studies included children with ADHD is observed [38]. On the other hand, there is another possibility that patients showing the discrepancy of sleepiness could have an undiagnosed neurodevelopmental disorders. We suggest that clinicians may employ the discrepancy between subjective and objective sleepiness to identify children at risk for neurodevelopmental disorders.

Currently, there is no questionnaire which specifically assesses sleepiness for children with neurodevelopmental disorders [32]. Moreover, it is possible that adolescents with neurodevelopmental disorders may have some difficulty understanding the questionnaire without parents' assistance. Therefore, in clinical practice, it is necessary to evaluate subjective sleepiness with a specific questionnaire, while also considering the complaints and symptoms of patients with neurodevelopmental disorders.

There were several limitations associated with the present study. The main limitation is that the OS group included a small number of patients. It is possible that this limitation prevented significant factors for discrepancy of objective sleepiness to be identified in our multivariable regression analysis. Therefore, further studies employing a large number of participants should be conducted to examine potentially significant factors. Another limitation of the present study is the absence of Tanner scale results for the participants due to the retrospective nature of the study. It has been reported that maturation stage is associated with sleepiness [24,25]. In the present study, patient age was found to be related to subjective sleepiness. However, given the adolescent age range of our cohort, it is possible that pubertal maturation may also mediate an effect on the observed discrepancy of sleepiness. Finally, a diagnosis for all of the sleep disorders affecting our cohort were not included in our multivariable regression model for several reasons. First, there was not a sufficient number of samples to examine a greater number of predictors in our model. Second, some of the clinical diagnoses were based on MSLT and JESS results, and we divided our participants into four groups based on these results. Because of these conditions, a statistical program may produce questionable validity of model fit. Consequently, we only included neurodevelopmental disorders as a clinical diagnosis.

5. Conclusions

This study identified the frequency of discrepancy between subjective and objective sleepiness in a cohort of adolescent patients. Nearly half of the patients exhibited discrepancy between subjective and objective sleepiness which need to be taken into consideration in the management of hypersomnia. In addition, we observed that discrepancy of subjective sleepiness was more likely to occur compared to objective sleepiness. Significant factors related to subjective discrepancy included: age, delayed bedtimes, and neurodevelopmental disorders in the adolescent population examined.

CRediT authorship contribution statement

Oyunsuren Munkhjargal: Conceptualization, Data curation, Writing – original draft, preparation. **Yasunori Oka:** Conceptualization, Data curation, Writing – review & editing. **Sakurako Tanno:** Data curation. **Hiroshi Shimizu:** Data curation. **Yoko Fujino:** Data curation. **Tomoko Kira:** Data curation. **Akiko Ooe:** Data curation. **Mariko Eguchi:** Supervision. **Takashi Higaki:** Supervision.

Declaration of competing interest

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