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Regular Article

Clinical utility of the Chinese version of the Pediatric Daytime Sleepiness Scale in children with obstructive sleep apnea syndrome and narcolepsy

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Aim: The present study examined the psychometric properties of the Chinese version of the Pediatric Daytime Sleepiness Scale (PDSS) and the utility of the PDSS as a screening tool for pathological daytime sleepiness in teenagers with obstructive sleep apnea (OSA) and narcolepsy.

Methods: The PDSS was first administered to 238 middle and high school students to assess the reliability of the scale, and then administered to 28 teenagers with OSA, 31 teenagers with narcolepsy, and 34 normal controls to evaluate its clinical utility.

Results: Test–retest reliability and internal consistency were acceptable. The PDSS scores were significantly higher in narcoleptic subjects than in subjects with OSA, and higher in OSA syndrome (OSAS) subjects than normal controls. Furthermore, the scores decreased in narcoleptic subjects after medical treatment. Both reliability and validity were proven to be good. As a screening tool for narcolepsy, receiver operator characteristic (ROC) curve analysis showed that the PDSS, with a cut-off score of 16/17, had good sensitivity (87.1%) and fair specificity (74.3%) for identifying individuals with narcolepsy. When used for screening OSA, however, the differentiating power was not as good.

Conclusion: The PDSS is a reliable and valid tool for the measurement of sleepiness in clinical youth populations. When used as a screening tool, it is useful for sleep disorders involving more severe pathological sleepiness, as in narcolepsy.

Key words: children and adolescents, measurement, narcolepsy, obstructive sleep apnea, sleepiness.

EXCESSIVE DAYTIME SLEEPINESS (EDS) among children and adolescents has become more and more a concern of health professionals and a focus of research in educational fields durings the past decade. One reason for the increased concern is the recognition that this population tends to have insufficient amounts of sleep in general. The restricted amount of sleep was found to be associated with increased level of sleepiness, as well as various behavioral and emotional disturbances and academic impairment.[1–12] In clinical practice it is also recognized that many sleep disorders characterized by sleepiness are more common than previously thought. For example, some sleep disorders that disrupt sleep, such as sleep-related breathing disorders and periodic limb movement disorder, may lead to sleepiness and other related behavioral or cognitive symptoms.[13–15] Also, various sleep disorders with excessive sleepiness as their primary symptom, such as narcolepsy and Kleine–Levin syndrome, typically begin during late childhood and adolescence.[16] Therefore, it is important to evaluate the level of sleepiness in

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the assessment of sleep disorders in children and adolescents.

Standardized procedures, such as the multiple sleep latency test (MSLT) and the maintenance of wakefulness test (MWT), have been developed for the assessment of sleepiness in both research and clinical settings. The actual practice of these procedures, however, is limited due to the need for equipment, time and labor. In contrast, several self-rating scales were developed for the evaluation of sleepiness. For example, the Epworth Sleepiness Scale (ESS) is one of the most popular self-rating scales for adult sleepiness in both clinical and research settings. The ESS has been validated in different clinical populations,[17–23] and in different languages.[24–30] It measures the level of sleepiness by listing eight daily life situations for which raters judge the likelihood of them falling asleep.[31] The ESS, however, may not be appropriate for children and adolescents because some of the situations included in it, such as driving and sitting in a meeting, may not be applicable to children and adolescents. Furthermore, some situations that may elicit sleepiness in adults, such as watching TV, may actually cause arousal in children. Therefore, sleepiness rating scales should be developed specifically for children and/or adolescents.

Studies on subjective measures of sleepiness in children and adolescents are relatively limited. Although the ESS has been recently modified for children and adolescents, the sensitivity in detecting obstructive sleep apnea (OSA) in children is low.[32] Among the few self-rating scales available, the Pediatric Daytime Sleepiness Scale (PDSS) has been shown to have good reliability and validity in middle-school children from 11 to 15 years old.[33] The PDSS consists of eight questions regarding sleep-related behaviors for which the frequency of these behaviors is rated on a 5-point Likert scale. The PDSS was shown to have good internal consistency, with Cronbach alphas of 0.80 and 0.81 for the split-half samples. Also, higher scores on the PDSS were associated with reduced total sleep time, poorer school achievement, poorer anger control, and frequent illness.[33] In addition, frequent snorers were found to score higher on a Spanish version of the PDSS in comparison to occasional snorers or non-snorers. Furthermore, PDSS scores were shown to predict failure in mathematics and language grades.[34] Therefore, the PDSS has been found to be a useful tool in measuring the level of sleepiness in school settings.

The utility of the PDSS in clinical settings remains to be evaluated. One study showed that children and adolescents (8–18 years old) with epilepsy scored significantly higher on the PDSS than matched controls. Moreover, PDSS scores correlated significantly with daytime sleepiness reported by the parents.[35] As far as we know, however, there has been no study reporting on the psychometric properties of the PDSS when used in the assessment of sleep disorders that are characterized by EDS. Moreover, parent reports and teacher reports are used widely in surveying children, but it may sometimes be better for children themselves to report on their sleep problems so as to reduce parental oversight during questionnaire administration and allow for more straightforward responses from teenagers.

The purposes of the present study were therefore (i) to evaluate the reliability and validity of the Chinese version of the PDSS; and (ii) to examine the clinical utility of the PDSS in the screening for sleep disorders characterized by EDS in children, specifically narcolepsy and obstructive sleep apnea syndrome (OSAS). To achieve the first study goal, the PDSS was first administered to a larger normal population to establish the reliability of the scale. It was then administered to two clinical samples, OSAS and narcolepsy patients, to examine its validity. For the second purpose, receiver operator characteristic (ROC) analyses were conducted separately for the OSAS and the narcolepsy samples to exam the clinical utilities of the PDSS for the screening of OSAS and narcolepsy.

METHODS

Measures

Pediatric Daytime Sleepiness Scale–Chinese version

The PDSS, developed by Drake *et al.*, is a self-rating scale that is designed to measure daytime sleepiness in children and adolescents.[33] It consists of eight items describing sleep-related behaviors. The child rates the frequency of these behaviors on a 5-point Likert scale: never, 0; seldom, 1; sometimes, 2; frequently, 3; and always, 4. The ratings on all the items were summed to calculate the total score, which ranged from 0 to 32. The PDSS was shown to have a good internal consistency, with Cronbach alphas of 0.80 and 0.81 for the split-half samples. Also, higher score on the PDSS was shown to be associated with

reduced total sleep time, school achievement, anger control, and frequent illness.

Permission to translate the PDSS into Chinese was obtained before the start of the study. The Chinese version of the PDSS was initially translated from the Drake *et al.* English version into Mandarin Chinese by one of the investigators.[33] Following this, it was translated back to English simultaneously by two bilingual individuals until the versions were considered completely interchangeable conceptually and linguistically.

Participants and procedures

Study 1

Participants for study 1 were recruited from school settings. The Institutional Review Board (IRB) approved the study, and informed consent was obtained from all subjects prior to implementation. We contacted the school counseling centers and explained to teachers the purpose of the study and procedures that were necessary. All eligible students and their respective parents were informed that participation in the survey was completely voluntary and thereafter parents' approval and signed informed consents were obtained. Three hundred and twelve students (175 boys and 137 girls) were recruited from schools to participate in the study. A package of questionnaires, including the PDSS and basic demographic questions, was administered during class by trained research assistants. The PDSS was given again 4 weeks later to obtain re-test data. Participants with missing data or who did not complete the questionnaire twice were excluded from the study. Two hundred and thirty-eight students (129 boys and 109 girls), including 170 middle school students and 68 high school students, completed the study.

Study 2

The participants for study 2 included 28 teenagers with OSA, 31 with narcolepsy, and 35 normal controls. The participants and their respective parents were informed that participation was completely voluntary and thereafter parents' approval and signed informed consents were obtained. Their age and gender distribution is presented in Table 1. One-way ANOVA showed no significant difference in their ages. Also, χ^2 test was not significantly different with regard to gender distribution among the three groups.

The patient groups were recruited from a sleep disorder center in a general hospital, and were diagnosed by a pediatric psychiatrist specializing in pediatric sleep medicine. OSAS was diagnosed by administering a clinical interview followed by one night of polysomnographic (PSG) recording. Based on the diagnostic criteria in the International Classification of Sleep Disorders Second Edition (ICSD-II),[16] OSAS patients should have an apnea–hypopnea index (AHI) >1/h and have breathing-related arousals or oxygen desaturation. Narcolepsy with cataplexy was diagnosed using clinical interviews, and then confirmed by MSLT and PSG scores and human leukocyte antigen (HLA) typing of DQB1*0602 positive. Normal controls were recruited from the community. A clinical interview was conducted to rule out medical and psychiatric disorders, and one night of PSG recording showed no indication of any sleep disorders.

After the diagnoses were confirmed, a research assistant administered a package of questionnaires consisting of the PDSS and demographic questions to the participants individually, with help from their parents. For the narcoleptic group, the PDSS was given again after they received medication treatment.

Table 1. Subject characteristics and PDSS score

	Group				
	Narcolepsy	OSAS	Control	*F*	χ^2
n	31	28	34		
Gender (boy/girl)	21/10	22/6	23/11		0.91
Age (years) (mean ± SD)	12.55 ± 3.40	13.07 ± 1.98	13.97 ± 1.71	2.76	
PDSS score (mean ± SD)	22.03 ± 4.83	17.61±4.57	14.51 ± 4.28	22.53*	

*$P < 0.001$.
OSAS, obstructive sleep apnea syndrome; PDSS, Pediatric Daytime Sleepiness Scale.

Statistic analysis

The reliability and validity of the PDSS–Chinese version was evaluated first. Test–retest reliability was evaluated by correlating the scores of the two administrations of PDSS in school subjects. The internal consistency of the PDSS was examined by calculating the Cronbach alpha. The discriminative validity was then evaluated by comparing the PDSS scores across the three groups (normal controls, OSA, and narcolepsy), as well as between the ratings of narcoleptic subjects before and after pharmacological treatment. Significance was set at $P < 0.05$ in two-tailed tests.

ROC analyses were also conducted to examine the clinical utility of the PDSS in distinguishing narcolepsy subjects from normal controls, and OSAS subjects from normal controls.

RESULTS

The participants in the school setting obtained a mean PDSS score of 14.77 ± 4.44. The test–retest correlation was 0.78 ($P < 0.001$) between the scores for the two administrations at 4 weeks apart. Test–retest reliability was acceptable considering the fluctuating nature of sleepiness in teenagers. In terms of internal consistency, an acceptable Cronbach alpha of 0.66 was obtained for middle and high school students and 0.61 for OSA patients, and a good Cronbach alpha of 0.81 for the narcoleptic group.

Table 1 lists the mean scores on the PDSS across different groups in study 2. One-way ANOVA comparing the PDSS scores across the three groups (normal controls, OSA, and narcolepsy) showed a significant main effect ($F(2,91) = 22.53$, $P < 0.001$). Post-hoc analysis using the Scheffe method showed that the narcolepsy group scored significantly higher on the PDSS than both the OSAS group and the control group ($P < 0.005$ and $P < 0.001$, respectively). The OSAS group also scored significantly higher than the normal control group ($P < 0.05$).

ROC curves were generated separately for narcolepsy and OSAS groups to test the clinical utility of using PDSS to differentiate participants with sleep pathologies from normal controls. The area under the curve (AUC) for differentiating narcolepsy from normal controls was 0.877 (Fig. 1). The value of 0.50 was not included within a 95% confidence interval of the AUC (0.80–0.96), suggesting that the discriminating capability of the PDSS score is statistically sound. Detailed output from this ROC analysis is listed in Table 2, which includes sensitivity and specificity, positive predictive value (PPV) and negative predictive value (NPV), with cut-off scores ranging from 13/14 to 18/19. For the discrimination of OSAS patients from normal controls, the AUC was 0.681 (Fig. 2). The value of 0.50 was also not included within a 95% confidence interval of the AUC (0.55–0.84). Details of the ROC analysis for differentiating OSAS patients from controls are also presented in Table 2.

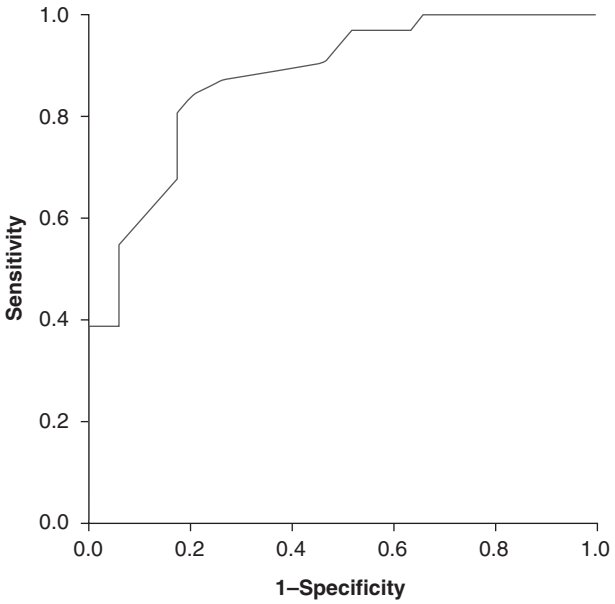

Figure 1. Receiver operator characteristic curve of the Pediatric Daytime Sleepiness Scale for the differentiation of narcolepsy and normal controls (area under curve = 0.877).

DISCUSSION

The goal of the present study was to examine the psychometric properties of the PDSS–Chinese version, and to evaluate the clinical utility of the PDSS in screening for children and adolescents with sleep disorders associated with pathological daytime sleepiness.

First of all, the Chinese version of the PDSS demonstrated good validity in clinical settings. The Chinese version of the PDSS is able to reflect different levels of sleepiness in different clinical populations. Children and young adolescents with OSAS had higher scores on the PDSS in comparison to children and adolescents with no sleep complaints; children

Table 2. ROC analysis for discrimination of narcolepsy, OSAS and normal control subjects on PDSS

Groups	Cut-off score	Sensitivity	Specificity	PPV	NPV
Narcolepsy vs control	13/14	0.968	0.371	0.577	0.929
	14/15	0.968	0.486	0.625	0.944
	15/16	0.903	0.543	0.636	0.864
	16/17	0.871	0.743	0.750	0.867
	17/18	0.839	0.800	0.788	0.848
	18/19	0.806	0.829	0.806	0.829
OSAS vs control	14/15	0.714	0.486	0.526	0.680
	15/16	0.679	0.543	0.543	0.679
	16/17	0.571	0.743	0.640	0.684
	17/18	0.500	0.800	0.667	0.667
	18/19	0.429	0.829	0.667	0.644

NPV, negative predictive value; OSAS, obstructive sleep apnea syndrome; PPV, positive predictive value; ROC, receiver operator characteristic.

and adolescents with narcolepsy scored higher than the OSAS group. Moreover, narcoleptic patients had a significant reduction in PDSS scores after medication, indicating that the PDSS can be applied as an outcome measure for the treatment of sleepiness in children. In terms of the test–retest reliability, although the correlation between the two administrations was not optimal, the correlation coefficient of 0.78 is acceptable, considering the fluctuating nature of the levels of sleepiness in young adolescents. The internal consistency of the PDSS was also found to be acceptable–good among different populations. Overall, the PDSS was proven to be a reliable and valid tool in clinical patients. Consistent with previous studies on the English and Spanish versions of the PDSS,[33–34] the current results suggest that the PDSS could be a useful measure across different cultures and nationalities.

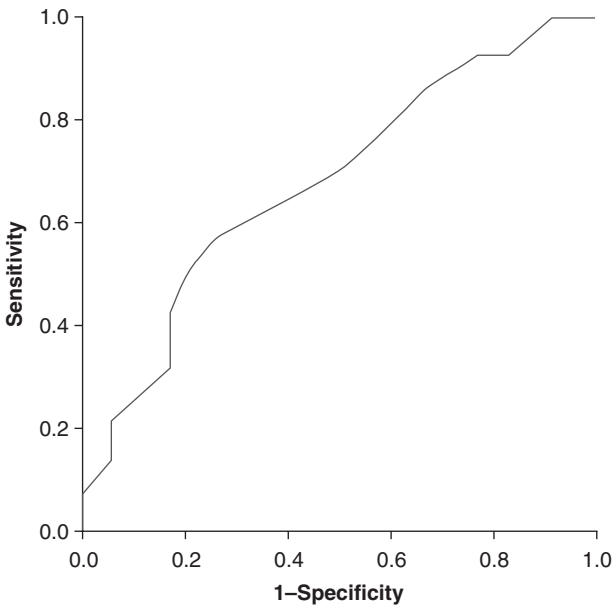

Figure 2. Receiver operator characteristic curve of the Pediatric Daytime Sleepiness Scale for the differentiation of obstructive sleep apnea and normal controls (area under curve = 0.681).

When considering the use of the PDSS as a screening tool for narcolepsy, ROC analysis showed that with a cut-off score of 16/17, the PDSS had good sensitivity (0.87) in detecting narcoleptic children from children with no sleep disorders. As well, it had fair specificity (0.74), which meant that it was able to avoid selecting too many false positive cases. It also demonstrated good PPV (0.75) and NPV (0.87). If needed, the specificity could be raised by using cut-off scores of 17/18 or 18/19, but the sensitivity would be decreased slightly in doing so. In sum, the PDSS could be a good screening tool for severe level of sleepiness as seen in narcoleptic children. Of course, the diagnosis of narcolepsy does not rely merely on the level of sleepiness. Further diagnostic procedures, such as mean sleep onset latency and sleep onset rapid eye movement period in the MSLT, and HLA typing, are needed to confirm the diagnosis.

In contrast, when the PDSS is used for the screening of OSAS, ROC analysis shows a sensitivity of 0.68 and a specificity of 0.54, with a cut-off score of 15/16. Although specificity can be increased by raising the

cut-off score, the sensitivity would drop to a level that is not acceptable for screening purposes. A recent study using a modified ESS for children reported good specificity (0.91) but poor sensitivity (0.29) with the recommended cut-off score. It was concluded that increased sleepiness is a specific but not a sensitive symptom in children with significant OSAS.[32] Although sleepiness is commonly considered to be a primary feature of OSAS in adults, the increased level of activity associated with sleep disruption in children may mask the sleepiness in some cases. In an early study by Guilleminault *et al.*, 50 children with OSAS were diagnosed on PSG. Among these children, 84% had EDS, 76% had some behavior disturbance, 42% were hyperactive, and 16% had decreased school performance.[36] Another study, however, found that fewer children with OSA reported EDS when compared to adults with OSA, with the notable exception of obese children.[37] Thus, although subjective EDS may be present in OSAS, especially for older children and adolescents,[16] it may not be a major symptom that could be relied on to detect OSAS in children and adolescents. In other words, OSAS children may show increased sleepiness as a group, but sleepiness may not have good discriminability when used to identify individual children with OSAS. Consistent with this finding, ICSD-II[16] did not include EDS as an essential symptom to diagnose OSAS. EDS was listed with hyperactivity, and aggressive behavior as one of the associated symptoms of OSAS.

In sum, the PDSS is a clinically useful tool to measure sleepiness in teenagers with pathological daytime sleepiness. When used as a screening tool for narcolepsy, it could successfully differentiate a substantial percentage of patients from normal children. Although children with OSAS, however, as a group, had higher level of sleepiness than normal control children, the PDSS tends to generate too many false-positive cases for a low cut-off score, or too many false-negative cases when the cut-off score is increased. It may therefore not be a suitable screening tool for OSAS in children, but it might be used as a measure for treatment outcome because it could reflect the difference in sleepiness between pediatric OSA and control groups.

There are some limitations of the present study. The study examined only the clinical utility of the PDSS with regards to two diagnoses associated with EDS. Future studies are needed to evaluate the scale for use in different clinical populations. In addition, the age range of the clinical subjects used in the present study was broad. Level of sleepiness in children has been shown to increase with age.[11,33] Due to the limited sample size for each diagnostic group in the present study, it was not feasible to divide the subjects into different age groups. Future studies may be conducted to compare the discriminability of the PDSS in identifying OSAS in children, young adolescents, and older adolescents. Moreover, comparisons between the PDSS and objective measures of sleepiness, such as MSLT and MWT, can be conducted in future studies to further confirm the validity of the PDSS.

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