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Original article

Low threshold to Vestibular and Oral Sensory stimuli might affect quality of sleep among children with autism spectrum disorder

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Abstract

Aims: The current study aimed to validate the relationship between sensory characteristics and sleep dynamics among children with autism spectrum disorder (ASD) using an actigraph, which is an objective assessment device used for sleep monitoring.

Methods: A total of 40 children (age range, 3–6 years) participated in this study (n = 20, with ASD and n = 20, age-matched children with typical development [TD]). We examined sleep dynamics using actigraph for 7 consecutive days, and the relationship between sleep parameters and sensory characteristics was analyzed using the Japanese Version of Sensory Profile (SP-J).

Results: Significant differences were observed in terms of activities per minute during sleep (p = 0.02), sleep efficiency (SE) (p = 0.005), and wake after sleep onset (WASO) (p = 0.02) between the two groups. In the ASD group, significant positive correlations were observed between activities per minute during sleep and low thresholds for Vestibular Sensory stimuli (p = 0.046) and Oral Sensory stimuli (p = 0.006) using the SP-J. Based on a multiple regression analysis, the activities per minute during sleep were associated with low thresholds for Oral Sensory stimuli ($\beta = 0.51$, t = 2.29, p = 0.03), but not with other factors, in the ASD group.

Conclusions: The current study showed that atypical Vestibular and Oral Sensory modulation may be a risk indicator for high activities during sleep among preschool children with ASD. Thus, whether the interventions for these sensory characteristics are effective in improving sleep quality, daytime activities, behaviors, and cognitive functions in this group of children must be considered.

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Keywords: Sleep disorder; Autism spectrum disorder; Actigraph; Sensory processing profile; Sensory sensitivity

1. Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by a persistent impairment in reciprocal social communication and interaction and restricted, repetitive patterns of

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behavior, interests, or activities [1]. Sleeping disorders are more common in children with ASD than in those with typical development (TD). Recent studies showed that the prevalence rate of sleep disturbance among children with ASD was 32%-71.5% [2-4]. Sleep deprivation affects the development of cognitive and academic abilities during childhood. Moreover, sleep duration within 24 months after birth had an inverted U-shape relationship with cognitive function at 6 years, and frequent awaking is associated with lower nonverbal intelligence [5]. In addition, children who have a short duration of sleep during infancy had high hyperactivity scores at preschool [6]. Thus, the quality of sleep, particularly sleep duration, might be correlated to problems with daytime cognitive and adaptive functions in children with ASD [7].

There are various hypotheses about the causes of sleep disorders among children with ASD, which include brain organizational and maturational differences, circadian-relevant genes, abnormal melatonin synthesis, and arousal and sensory dysregulation [8]. The current study focused on sensory abnormality, which is considered a cause of sleep disorders among children with ASD. Recently, sensory abnormalities were added to the diagnostic criteria of the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) and were considered important symptoms of ASD [1]. Tzischinsky et al. revealed that the severity of sleep disturbance was moderately associated with touch and oral sensitivities based on the Caregiver Sensory Profile and the Children's Sleep Habits Questionnaire [9].

Interventions for sleep problems may improve daytime activities and cognitive functions among children with ASD. Hence, the sleep characteristics of children with ASD must be identified. Sleep and wake rhythm at home during early childhood is solely subjective based on the evaluation of parents using a sleep log or questionnaire, and an accurate assessment is often challenging to perform particularly after falling asleep. Therefore, we used actigraph, which is an objective evaluation device. In recent years, this device has been used in various studies of the pediatric population [10,11]. Actigraph is a wristwatch-type accelerometer. Thus, it can be used at home and can record daily activity rhythms not only at night. Moreover, it is an easy-to-use and noninvasive device. Hence, it can be utilized to assess sleep among children.

There are no studies available which have investigated the sleep characteristics of children with ASD using Sensory Profile and actigraph. Therefore, the current study aimed to validate the relationship between sensory characteristics and sleep dynamics among children with ASD using Sensory Profile and actigraph.

2. Participants and methods

The study protocol and procedures were approved by the ethics committee of the University of Fukui, Japan (assurance #: 20170034), and the study was conducted in accordance with the Declaration of Helsinki and the Ethical Guidelines for Clinical Studies of the Ministry of Health, Labour and Welfare of Japan. The purpose and significance of the study were explained to all the parents of the participants, and subsequently, a written informed consent was obtained.

2.1. Participants

In total, 40 children (n = 20, with ASD and n = 20, age-matched children with TD) (age range, 3-6 years) who were recruited from February 2018 to June 2019 participated in this study. 20 subjects recruited from two hospitals (Department of Child and Adolescent Psychological Medicine, University of Fukui Hospital, and Department of Pediatrics, Fukui Prefectural Center for Children with Developmental Disabilities) were diagnosed with ASD by two pediatric neurologists according to the DSM-5 criteria. In the ASD group, developmental quotient (DQ) was assessed using the Kyoto Scale of Psychological Development [12] or intelligence quotient (IO) with the Wechsler Intelligence Scale for Children Fourth Edition [13]. Children with TD were recruited from the general population through advertising. We confirmed that the children with TD did not have ASD or comorbid disorders (e.g., attention deficit/hyperactivity disorder), were evaluated by a semi-structured diagnostic interview, via the shortened version of the Parent-Interview ASD Rating Scale, text revision [14], which was administered by an experienced clinician. However, children with atopic dermatitis or bronchial asthma and those taking drugs, such as antihistamines, antiepileptic drugs, and ramelteon, that may affect sleep were excluded.

2.2. Actigraph

The participants were instructed to wear an actigraph (Actiwatch Spectrum Plus, Philips Respironics, Inc., the USA) at home on their wrist for 7 consecutive days. The data were only recorded during nighttime in two patients in the ASD group and one in the TD group and for 6 days in one patient in the ASD group. The sampling frequency was 32 Hz, and the epoch length was 1 min. The active-rest threshold per epoch was set at 4, and the wake–sleep threshold per epoch was set at 40. The data obtained using the actigraph were translated into sleep parameters using a software (Actiware 6.0, Philips Respironics, Inc.). Bedtime was automatically calculated using the peak and bottom of the activ-

ity values. Sleep onset time was defined as the first epoch in which all but one epoch was scored as rest for 10 consecutive minutes. Using the same method, wake up time was defined as the first epoch in which all but one epoch was scored as active for 10 consecutive minutes. Time in bed (TIB) was defined as time from bedtime to wake up time. Total sleep time (TST) was the sum of all sleep epochs. Sleep latency (SL) was defined as time from bedtime to sleep onset time. Wake after sleep onset (WASO) was defined as the sum of all wake epochs during the sleep period. Sleep efficiency (SE) was defined as the TST-to-TIB ratio.

2.3. Sensory processing traits

The Japanese Version of the Sensory Profile (SP-J) [15] was administered to evaluate the tendency to probe sensory behaviors and characteristics. This questionnaire contains 125 questions that quantify the frequency of abnormal behavioral responses to various sensory experiences. Caregivers check the most applicable item regarding the frequency of actions described in the question item, which are divided into five stages [1 = never(0%), 2 = seldom (25%), 3 = occasionally (50%), 4 = frequently (75%), and 5 = always (100%)]. We focused on the six sensory subscales of the SP-J, which include questions about Auditory, Visual, Vestibular, Touch, Multisensory, and Oral Sensory processing. These question items are divided based on high or low threshold. High threshold means habituation. Children whose thresholds are too high tend to be under-responsive (i.e., it takes a lot of stimuli to reach the threshold, as observed when children do not respond to cues around them). Low threshold means sensitization. Children whose thresholds are too low tend to be over responsive (i.e., very little stimuli cause a reaction, as observed when children are distracted by every stimulus) [16]. We summed the scores of questions about the high and low thresholds for each item. We compared the differences between the ASD and TD scores, and the relationship between the SP-J score and the actigraph parameters.

2.4. Japanese Sleep Questionnaire for Preschoolers

The Children's Sleep Habit Questionnaire is often used. However, the current study utilized the Japanese Sleep Questionnaire for Preschoolers (JSQP) to compare sleep problems among children with ASD and TD. Sleep habits considerably differ between Japan and Western countries [17]. In fact, almost all younger children sleep with their parents in Japan, as opposed to those in Western countries who sleep alone in their own room. Therefore, we believed that JSQP was appropriate to analyze Japanese sleep habits. In 2014, Shimizu *et al.* introduced the JSQP, which was established according to Japanese sleeping habits [17]. The JSQP is a questionnaire that covers common sleeping problems among preschool children. Caregivers rate each question on a six-point scale. When the score is higher, the problem is worse. The questionnaire comprises 39 items that are classified into 10 subscales.

2.5. Statistical analysis

Statistical analysis was performed using the Statistical Package for the Social Sciences software (version 25, IBM), and data about bed time, sleep onset time, and wake up time were analyzed using Excel (Microsoft Office Home and Business 2016). A p value <0.05 was considered statistically significant. Data were expressed as mean \pm standard deviation or median (min-max). The differences between the two groups (demographic characteristics and sleep measurement using actigraph) were analyzed using unpaired t-test. The group differences based on the SP-J were assessed using the Mann-Whitney U test because the scores had a nonnormal distribution. The relationships between the scores for each SP-J item and the activities per minute during sleep were examined using Spearman's correlation. To investigate the substantial contribution of activities during sleep to sensory characteristics and other factors correlated to sleep, a standard multiple regression with the forced entry method was used.

3. Results

3.1. Clinical characteristics of the participants

The study included 20 children with ASD (age: 3–6 [mean: 5.1 ± 1.3] years) and 20 age-matched children with TD (age: 3–6 [mean: 5.1 ± 0.9] years). No significant difference was found in terms of sex, gestational week, and birth weight between children with ASD and those with TD. All participants attended elementary school or nursery school and had siblings. No

Table I	
Demogra	phics

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	ASD	TD	<i>p</i> -value
N	20	20	_
Male	17	12	0.08
Age (years)	5.2 ± 1.3	5.1 ± 0.9	0.65
Gestational weeks (weeks)	38.6 ± 2.7	38.1 ± 1.1	0.46
Birth weight (g)	3070 ± 550	2960 ± 290	0.42
DQ (n = 17)	86.1 ± 27.7	_	-
IQ (n = 3)	94.7 ± 7.6	_	-
Attending in school	20	20	-
Sleeping with parents	18	18	-
Presence of siblings	20	20	-

Data are shown as mean \pm SD, ASD; Autism spectrum disorders, TD; Typical development, DQ; Developmental quotient, IQ; Intelligence quotient.

differences were observed between the two groups in terms of parents sleeping with their children (Table 1).

3.2. Sleep problems among children with ASD according to the JSQP

Children with ASD had significantly higher scores than those with TD based on the total JSQP score and subscale scores for restless legs syndrome (RLS) sensory, obstructive sleep apnea syndrome (OSAS), parasomnia, insomnia or circadian rhythm disorders, daytime excessive sleepiness, and daytime behaviors. The scores were above the cutoff, except for OSAS. A significant difference was observed in terms of the total score for OSAS, insomnia or circadian rhythm disorders, daytime excessive sleepiness, and daytime behaviors (p < 0.01) (Table 2).

3.3. Sensitivity to sensory stimuli among children with ASD

All subscales significantly differed in both the high and low thresholds between the ASD and TD groups, except for low threshold to Multisensory processing in the SP-J (Table 3).

3.4. Various indicators for sleep using actigraph

Bed time, sleep onset time, wake up time, time in bed, total sleep time, and sleep latency did not significantly differ between the ASD and TD groups. Moreover, there was no difference during daytime activity. However, a significant difference was observed in activities during sleep (9790 \pm 3420/7710 \pm 1840, p = 0.023) and activities per minute during sleep (18.5 \pm 6.5/14.3 \pm 3.3, p = 0.017). In addition, SE (82.8 \pm 4.8%/86.6 \pm 3.1%, p = 0.005) and WASO (70.2 \pm 17.9 min/57.1 \pm 16.1 mi n, p = 0.019) significantly differed between the two groups (Table 4).

3.5. Relationship between activities per minute during sleep and SP-J scores

The relationship between the activities per minute during sleep and SP-J scores was examined. In the ASD group, a significant positive correlation was observed between the activities per minute during sleep and low threshold to Vestibular Sensory stimuli (p = 0.046) and Oral Sensory stimuli (p = 0.006) using Spearman's correlation. By contrast, no significant correlation was observed between the activities per minute during sleep and SP-J scores in the TD group (Table 5).

3.6. Factors contributing to activities during sleep

We performed a multiple regression analysis to determine the impact of sensory characteristics on the activities per minute during sleep in the ASD group. We considered low threshold to Oral Sensory stimuli as an independent variable, which was scored using the SP-J. Low threshold to Vestibular Sensory stimuli was not considered as an independent variable because of its significant correlation to low threshold to Oral Sensory stimuli. The scores of the RLS sensory domain and OSAS domain in the JSQP significantly differed between the two groups, and they were considered as independent variables. Our analysis revealed that low threshold to Oral Sensory stimuli was a significant predictor of the activities per minute during sleep in all participants $(\beta = 0.60, t = 3.92, p = 0.00038)$, and it explained approximately 25% of variance (adjusted $R^2 = 0.25$, p = 0.004). In the ASD group, low threshold to Oral Sensory stimuli was a significant predictor of the activities per minute during sleep ($\beta = 0.51$, t = 2.29, p = 0.03), and it explained approximately 14% of variance (adjusted $R^2 = 0.14$, p = 0.15). However, the activities per minute during sleep were not associated with other independent factors. In the TD group, there was no association between the independent factors (Table 6).

	ASD $(n = 20)$	TD $(n = 20)$	<i>p</i> -value
Total score	106.2 ± 23.8	75.4 ± 14.0	<0.001**
RLS sensory	6.1 ± 4.0	4.0 ± 1.8	0.04*
RLS motor	3.6 ± 2.4	2.5 ± 1.1	0.07
OSAS	19.5 ± 5.6	13.3 ± 4.3	< 0.001**
Parasomnia	12.7 ± 7.2	8.2 ± 4.1	0.02*
Insomnia or circadian rhythm disorders	22.6 ± 6.7	16.3 ± 4.4	0.001**
Morning symptom	11.3 ± 4.7	9.5 ± 3.3	0.18
Daytime excessive sleepiness	7.9 ± 2.7	4.8 ± 1.5	< 0.001**
Daytime behaviors	8.1 ± 1.7	3.3 ± 1.6	< 0.001**
Sleep habit	8.2 ± 1.7	7.7 ± 2.9	0.51
Insufficient sleep	6.5 ± 2.8	6.0 ± 2.7	0.61
*			

Table 2 The score of Japanese Sleep Questionnaire for Preschoolers.

Data are shown as mean \pm SD, RLS; restless legs syndrome, OSAS; obstructive sleep apnea syndrome, group differences were tested by unpaired *t*-test, **p < 0.01, *p < 0.05.

Table 3				
The score of Japanese	Version	of the	Sensorv	Profile.

Sensory processing		ASD $(n = 20)$	TD (n = 20)	<i>p</i> -value
Auditory	High threshold	7.5 (3.0–13.0)	3.0 (3.0–14.0)	<0.001**
	Low threshold	15.0 (6.0–21.0)	5.5 (5.0–15.0)	<0.001**
Visual	High threshold	6.0 (2.0–9.0)	2.0 (2.0–6.0)	0.001^{**}
	Low threshold	11.5 (7.0–27.0)	7.0 (7.0–11.0)	< 0.001^{**}
Vestibular	High threshold	13.0 (5.0–23.0)	5.0 (5.0–19.0)	0.0014*
	Low threshold	6.0 (6.0–16.0)	6.0 (6.0–8.0)	0.026*
Touch	High threshold Low threshold	14.0 (7.0–28.0) 18.5 (11.0–37.0)	7.0 (7.0–12.0) 11.0 (11.0–19.0)	$<\!\!0.001^{**}$
Multisensory	High threshold	6.0 (4.0–11.0)	4.0 (4.0–7.0)	<0.001**
	Low threshold	1.0 (4.0–1.0)	1.0 (1.0–1.0)	0.06
Oral Sensory	High threshold	13.0 (7.0–30.0)	7.0 (7.0–13.0)	$<\!\!0.001^{**}$
	Low threshold	10.5 (5.0–21.0)	5.0 (5.0–7.0)	$<\!\!0.001^{**}$

Data are shown as median (min-max), group differences were tested by Mann-Whitney U test, **p < 0.01, *p < 0.05.

Table 4

Sleep measurement using actigraph.

	ASD $(n = 20)$	TD $(n = 20)$	<i>p</i> -value
Bed time	21:56 ± 0:42	$21:52 \pm 0:42$	0.80
Sleep onset time	$22:00 \pm 0:41$	$22{:}02\pm0{:}40$	0.83
Wake up time	$6:46\pm0:49$	$6:52\pm0:33$	0.79
Time in bed (min)	530.9 ± 27.3	536.2 ± 35.3	0.60
Total sleep time (min)	439.0 ± 37.8	460.3 ± 28.4	0.05
Sleep latency (min)	4.5 ± 4.7	4.0 ± 2.7	0.63
Activities during daytime	$513,000 \pm 160,000$	$535,000 \pm 116,000$	0.63
Activities during sleep	9790 ± 3420	7710 ± 1840	0.023*
Activities per minute during sleep	18.5 ± 6.5	14.3 ± 3.3	0.017*
Sleep efficiency (%)	82.8 ± 4.8	86.6 ± 3.1	0.005**
WASO (min)	70.2 ± 17.9	57.1 ± 16.1	0.019*

Data are shown as mean \pm SD, WASO; wake after sleep onset, the group differences were tested using unpaired *t*-test, **p < 0.01, *p < 0.05.

Table 5

Relationship between activities per minute during sleep and the score of SP-J.

Sensory processing ASD (n = 20)TD (n = 20)r r р -0.59Auditory High threshold 0.02 0.80 Low threshold -0.120.08 0.62 Visual High threshold -0.010.99 -0.15Low threshold 0.15 0.52 -0.10Vestibular High threshold 0.21 0.37 -0.18Low threshold 0.45 0.046* -0.18Touch High threshold 0.28 0.23 0.29 Low threshold 0.18 0.46 -0.24-0.010.99 Multisensory High threshold -0.39Low threshold 0.05 0.83 . a Oral Sensory High threshold 0.35 0.13 0.08 0.006** Low threshold 0.59 -0.43

This table shows correlation between activities per minute during sleep measured using actigraph and the score of SP-J using Spearman's correlation, *p < 0.01, *p < 0.05, ^acorrelation coefficient was not calculated because all scores of low thresholds of Multisensory processing were equal in the TD group.

р

0.95

0.73

0.52

0.67

0.44

0.45

0.21

0.31

0.09

0.73

0.06

. ^a

regression insuch of statistics and estimated for addition per initiate daring steep.							
	All $(n = 40)$		ASD $(n = 20)$		TD (n = 20)		
Independent parameters	β	<i>t</i> -value	β	<i>t</i> -value	β	<i>t</i> -value	
The score of low threshold of Oral Sensory	0.60**	3.92	0.51*	2.29	-0.39	-1.74	
RLS sensory	-0.11	-0.70	-0.13	-0.56	-0.07	-0.30	
OSAS	-0.09	-0.62	-0.11	-0.48	-0.30	-1.34	
Adjusted R^2	0.25**		0.14		0.09		

Table 6 Regression model of statistics and coefficient of variables for activities per minute during sleep.

Standardized regression coefficient (β), *t*-values, and adjusted R^2 are reported. The corrected statistical threshold was set at **p < 0.01, *p < 0.05. Low threshold of Oral Sensory was scored in SP-J, RLS sensory and OSAS were scored using JSQP.

4. Discussion

We analyzed the relationship between sensory characteristics and sleep dynamics among children with ASD using actigraph, which is an objective assessment device used for sleep monitoring. The JSQP was used to assess the sleep status of children with ASD and those with TD. Similar to the study of Hirata [18], children with ASD had a significantly higher total JSQP score and score for each domain (OSAS, parasomnia, insomnia or circadian rhythm disorders, daytime excessive sleepiness, and daytime behaviors), and this result indicated that sleeping problems were more common in children with ASD than in those with TD, particularly in terms of symptoms before and after falling asleep or symptoms during daytime according to the parent's evaluation.

Some studies using polysomnography or actigraph showed that the most consistent features of the sleeping pattern of children with ASD were shorter TST [10,19], prolonged SL [10,11], increased WASO [20,21], and lower SE [20,21]. Significant differences were observed in activities per minute during sleep, WASO, and SE. However, TST, SL, bedtime, sleep onset time, and wake up time did not significantly differ between the two groups. In children with ASD, bedtime and SL can change before falling asleep based on the environment [22]. Bedtime and wake up time are relatively easy to confirm according to the caregiver's observation. Meanwhile, caregivers find it difficult to assess the activities per minute during sleep, WASO, and SE. Thus, the sleep parameters after falling asleep are more important in differentiating ASD from TD.

In the ASD group, a significant positive correlation was observed between activities per minute during sleep and low threshold to Vestibular and Oral Sensory stimuli. We hypothesized that in addition to these sensory characteristics, RLS and OSAS might affect activities per minute during sleep. However, the multivariate analysis revealed that a low threshold to Oral Sensory stimuli had a more significant effect on activities per minute during sleep than RLS and OSAS in all participants and particularly in the ASD group. The higher nighttime activities among children with ASD might be explained by the low threshold to Vestibular or Oral Sensory stimuli. The question items that are often applicable to the ASD group in terms of low threshold to Vestibular Sensory stimuli are "Becomes anxious or distressed when feet leave the ground" and "Dislikes activities where head is upside down (for example, somersaults, roughhousing)." Meanwhile, those that are commonly applicable in terms of low threshold to Oral Sensory stimuli are "Avoids certain tastes or food smells that are typically part of children's diets" and "Picky eater, especially regarding food textures." The higher sleeping activity among children with ASD might be attributed to the higher sensitivity or avoidance of Vestibular or Oral Sensory stimuli.

Vestibular Sensory stimuli affect sleep. The vestibular nuclei are connected via the inter-geniculate lateral region with the suprachiasmatic hypothalamic nucleus, which regulates biological rhythmicity [23,24] and controls the alternation of waking and sleeping state [25]. In addition, neuroanatomical connections from the orexin neurons to the vestibular nuclei have been observed [23]. Sugaya et al. showed that vestibular rehabilitation improved sleep disturbance in patients with chronic dizziness [26]. A previous study revealed the efficacy of weighted blanket when used as an intervention in ASD children with sleeping disorders. However, in a randomized controlled study, the use of a weighted blanket was not effective in increasing the duration of sleep among children with ASD. Moreover, it was not effective in helping these children fall asleep significantly faster or wake up less often [27]. Considering the characteristics of low threshold to Vestibular sensation (e.g., dislike in posture changes, losing stability when changing posture), the use of a weighted blanket may be useful for sleeping among children with ASD who have a low threshold to Vestibular sensation.

Sleep restriction might affect somatosensory perception in the orofacial area [28], and sleep bruxism is correlated to daytime fatigue and sleepiness [29]. There are few studies about approaches for Oral hypersensitivity that can be used to improve sleep, and interventions for Oral hypersensitivity before bedtime, including desensitization [30], might be effective in improving sleep quality among children with feeding difficulties. Sleep disorders, which affect daytime behaviors and cognitive functions, are highly prevalent among children with ASD. Hence, sleep disorders among children with ASD are a major problem that must be addressed. In children with ASD, a more active sensory evaluation by an occupational therapist or assessment using SP and interventions for sensory characteristics may be effective in improving activities during sleep. Whether interventions for sensory characteristics can improve sleep quality, daytime behaviors, and cognitive functions should be considered.

The current study had several limitations. First, the sample size was small. Second, the DQ and IQ of children with TD could not be measured. Hence, the relationship between DQ/IQ and the actigraph parameters was not evaluated. Third, as participants with ASD had different DQ/IQ scores, it cannot be denied that their intelligence level might have influenced the results. Fourth, because scoring tools for ASD such as Parent-Interview ASD Rating Scale were not used, we could not analyze the relationship between the severity of autistic tendency and the differences in sleep and sensory characteristics of children with ASD.

5. Conclusion

Low threshold to Vestibular and Oral Sensory stimuli may contribute to high activities during sleep among preschool children with ASD. Based on our data about the overall associations, whether interventions for sensory characteristics can improve sleep quality, daytime activities, behaviors, and cognitive functions must be considered.

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Conflict of interest disclosures

The authors declare no competing interests.

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