Genetic and Environmental Influences on Daytime and Nighttime Sleep Duration in Early Childhood

WHAT’S KNOWN ON THIS SUBJECT: Sleep patterns of adult monozygotic twins are more similar than those of dizygotic twins, showing moderate heritability and little effects of environmental influences. There have been very few genetically informative studies of sleep in preschool children and results appear inconsistent.

WHAT THIS STUDY ADDS: From previous studies, we investigated daytime and nighttime continuous sleep duration longitudinally. This is the first time that the etiologies of daytime and nighttime continuous sleep duration trajectories were studied in early childhood.

abstract

OBJECTIVES: To determine the relative contributions of genetic and environmental factors on daytime and nighttime continuous sleep duration at 6, 18, 30, and 48 months of age, and to identify different subgroups of children who followed different daytime and nighttime sleep duration trajectories and to investigate their etiology.

METHODS: The current study included 995 twins (405 monozygotic and 586 dizygotic) of the Quebec Newborn Twin Study recruited from the birth records of the Quebec Statistics Institute. Daytime and nighttime sleep was assessed through maternal reports at 6, 18, 30, and 48 months of age. A semiparametric modeling strategy was used to estimate daytime and nighttime sleep duration trajectories. Quantitative genetic models were used to examine to what extent genetic and environmental factors influenced daytime and nighttime continuous sleep duration.

RESULTS: Genetic modeling analyses revealed environmental influences for all daytime sleep duration trajectories. In contrast, strong genetic influences were found for consolidated nighttime sleep duration (except at 18 months and for the short-increasing sleep duration trajectory).

CONCLUSIONS: This is the first indication that early childhood daytime sleep duration may be driven by environmental settings, whereas the variance in consolidated nighttime sleep duration is largely influenced by genetic factors with a critical environmental time-window influence at ~18 months. Pediatrics 2013;131:e1874–e1880

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KEY WORDS
sleep duration, genetic and environmental interplay, early childhood, twin study

ABBREVIATIONS
BIC—Bayesian information criterion
DZ—dizygotic
MZ—monozygotic

Dr Touchette conceptualized the paper, analyzed/interpreted the data, and drafted the initial manuscript; Dr Dionne conceptualized and designed the study, interpreted the data, and drafted the initial manuscript; Dr Forget-Dubois carried out the genetic analyses and critically reviewed the manuscript; Drs Petit, Falissard, and Tremblay critically reviewed the manuscript; Dr Pérusse conceptualized and designed the study; Dr Boivin coordinated and supervised data collection and critically reviewed the manuscript; Dr Montplaisir elaborated the sleep measures and critically reviewed the manuscript; and all authors approved the final manuscript as submitted.

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Although environmental factors can influence sleep duration and quality, it is now known that differences in sleep behaviors depend largely on genetic variance among individuals. First, the findings of strong genetic contributions to many sleep-related disorders (eg, narcolepsy, idiopathic hypersomnia, insomnia, restless legs syndrome, sleepwalking, sleep terrors, sleep bruxism, enuresis, sleep apnea, delayed sleep phase syndrome) (for review, see ref 1) provided convincing indications. Then, the discovery of clock genes that regulate the circadian timing of sleep and wakefulness (for review, see ref 2) and of genes that account for the homeostatic process of sleep (for reviews, see refs 3 and 4) dramatically changed the backdrop for the study of sleep.

A review of the relative contributions of genetics and environmental factors to sleep patterns and sleep behaviors in adults, obtained through twin studies, demonstrated a moderate heritability (∼30% to 44%) for sleep duration and little effect of shared environmental influences (for review, see ref 1). In infants, however, our group found that heritability explained the greater proportion of the variance on a day/night sleep ratio at 6 months (64%), whereas the variance was mainly explained by shared environment influences at 18 months (58%). Another study of 18-month-old twins found a strong contribution of shared environmental factors for both nighttime sleep duration (64%) and daytime sleep (61%), with a moderate contribution of additive genetic effects (31% and 36%, respectively). This was supported by a recent and very large study of 15-month-old twins; the shared environment influence was predominant for both nocturnal (66%) and diurnal (57%) sleep and the genetic effect was modest (26% and 37%, respectively). Finally, 2 twin studies concluded that the genetic contributions to sleep patterns and sleep problems of preadolescents and adolescents were close to those found in adults.

The etiology of daytime and nighttime sleep duration in early childhood differ from that of adults for many reasons. First, sleep consolidation goes through major changes in childhood before reaching adult sleep patterns. Newborns follow a mostly ultradian cycle, with periods of 3 to 4 hours of sleep spread out across day and night. By 2 to 4 months, a circadian rhythm starts to emerge, with most sleep occurring during the night. Daytime sleep becomes well-defined naps: 2 to 3 naps per day until age 6 months (3.5 hours total), followed by 2 naps per day at ∼9 to 12 months, and finally, 1 nap in the afternoon after 18 months (2.5 hours) up to age 3. Approximately 68% of children have stopped taking a nap by age 4. Consolidated nighttime sleep duration increases considerably from birth to adolescence, when it reaches adult sleep patterns, but substantial individual variability remains at all ages. Second, sleep-wake periods are regulated by 2 processes that mature from birth: (1) sleep homeostasis (Process S), by which sleep pressure increases during wakefulness and dissipates during sleep and (2) circadian rhythmicity (Process C), which shows a wakefulness propensity as a sinus function over a 24-hour period. The progressive development of these sleep processes could explain why childhood sleep patterns differ from adult ones. Third, there are indications that parental practices around the sleep periods of young children affect the latter. Putting children to bed while they are dozy but still awake was found conducive to developing appropriate sleep-onset associations. Conversely, the need for parental presence until sleep onset is considered as a protodyssomnia, a potential precursor of later sleep problems. Variations in sleep consolidation during early childhood could thus be the result of family-based routines.

Because the maturation of daytime and nighttime sleep changes throughout early childhood, we first aimed to determine the relative contributions of genetic and environmental factors on these sleep phenotypes at 6, 18, 30, and 48 months of age in a sample of twins. Rather than assume that all children follow the same developmental daytime and nighttime patterns of sleep over time, the second objective of this study was to identify different subgroups of children who followed different daytime and nighttime sleep duration trajectories and to investigate their etiology.

**METHODS**

**Study Design**

Data were collected as part of the Quebec Newborn Twin Study on a population-based sample of 1392 twins (696 families) born between November 1995 and July 1998 in the greater Montreal Area (Canada). Infants had to be born without major medical conditions, had to have available birth records at the Quebec Statistics Institute, and their mother had to be fluent in either French or English. A 2-week interval separated the twins’ assessments to minimize the homogenization of answers as much as possible. Families received detailed information by mail on the aims and procedures of the research program and signed a consent form before each data collection.

**Participants**

The analyses presented here are based on data gathered during 4 times of assessments (at 6, 18, 30, and 48 months of age) of daytime and nighttime continuous sleep duration. Data
were available for 983 individual children (397 monozygotic [MZ], 582 dizygotic [DZ], and 4 children whose zygosity was missing; boys = 478, girls = 505) for both daytime and nighttime continuous sleep trajectories. Corrected age (for gestational age) at the targeted 6-, 18-, 30-, and 48-month assessments were as follows: 5.41 months (SD = 0.52), 18.64 months (SD = 0.60), 30.96 months (SD = 0.79), and 49.31 months (SD = 1.76). The mean gestation duration of the present sample (n = 983) was 36.14 weeks (SD = 2.52). Based on a full-term criterion of 37 weeks, 484 twins (49.2%) were born premature. Mean birth weight was 2459 g (SD = 543 g), mean 1 minute Apgar scores were 8/10 (SD = 1.6), and mean number of days spent in the hospital after birth was 9.4 (SD = 12.9); 47.2% of twins were born by cesarean delivery. Mean age of mothers at delivery was 30.7 years (SD = 4.7), and 15.4% had no high school diploma, average family income was between CAD $40 000 and CAD $50 000 per year, and ∼5% of the twins were born to single mothers. The first language of the mother was French for 78.6% of infants, English for 10.0%, both for 4.9%, and other than French or English for 6.5%.

### Measures

#### Zygosity

Zygosity was determined on physical resemblance using the Zygosity Questionnaire for Young Twins. At 18 months, zygosity was confirmed with DNA analysis of 8 to 10 highly polymorphic genetic markers on half the sample (n = 237 pairs) for same-sex twin pairs. This concordance rate (94%) was obtained after a case-by-case assessment of each pair’s physical similarity using a shortened version of the Zygosity Questionnaire for Young Twins, which is similar to rates in other twin samples. Uncertain zygosity diagnoses were periodically reevaluated based on the assessment of physical similarity by interviewers and genetic markers whenever possible.

### Measures of Infant Sleep Characteristics

The Self-Administered Questionnaire for the Mother, which took about 20 minutes to complete, provided information on the infant’s sleep characteristics, such as the number of consecutive hours slept in general per day and per night. As previously described in the methodology of another study, the number of consecutive hours of sleep during daytime or nighttime was obtained through maternal reports, with the question: “In general, how many uninterrupted hours does your baby/twin sleep during the day or at night?” The duration of continuous sleep during nighttime was assessed in rounded hours ranging from <4 hours to ≥8 hours at 6 months and up to ≥10 hours (which we considered to be 11 hours) at other ages. The duration of the longest continuous sleep period during daytime was assessed in rounded hours ranging from the following: does not nap (0), ≤1, >1 hour to <2 hours, ≥2 hours to <3 hours, ≥3 hours to <4 hours, and ≥4 consecutive hours. The “less than” and “more than” categories were rounded to the next whole value, as previously done in the same twin cohort. The terms “consolidated” or “continuous” do not imply the absence of brief awakening episodes. It simply means that the child did not signal any awakening to the parents, and thus was able to self-soothe and rapidly go back to sleep. Indeed, it was shown using video recordings that even the “good sleepers/nonsignalers” woke up on average 3 times per night between the ages of 1 and 5. However, it was also revealed that signalers were losing on average 1 hour and 22 minutes of sleep per night compared with nonsignalers.

### Statistical Analyses

#### Analyses of Transversal Twin Data

Analyses of twin data are based on the comparison of the covariance between MZ twins and DZ twins, knowing the coefficient of genetic relatedness, which is 1.0 for MZ twins and on average 0.5 for DZ pairs. It is hypothesized that a higher within-pair correlation in MZ reflects a genetic influence on the variance of the phenotype under study, assuming that the environment of both types of twins is equally similar. The phenotypic variance that is not explained by genetic variance is attributed to the environmental variance, either common (the environmental influences that make twins more alike) or unique (the environmental influences that make twins more different; this portion includes the measurement error). We analyzed the within-pair covariance at 6, 18, 30, and 48 months of age to estimate genetic and environmental contributions to the variance in daytime and nighttime continuous sleep duration. We used the Full Information Maximum Likelihood estimation procedure, which fits the tested model to all nonmissing data for all participants, and is the default estimator in the statistical package Mx. Full Information Maximum Likelihood with missing data at random has been shown to produce accurate parameter estimates.
methodology uses all available developmental data points and assigns individuals to trajectories based on a posterior probability rule. We chose the model that permitted the inclusion of the greater number of subjects (up to 2 missing data points out of 4 are allowed). The best model, with the optimal number of groups and slopes that best fit the longitudinal data, was identified based on the Bayesian information criterion (BIC).

To estimate the genetic and environmental etiology of both daytime and nighttime continuous sleep duration trajectories, we analyzed the concordance of twin pairs in trajectory membership, represented by the within-pair tetrachoric correlation coefficient. For each model fitted, membership to a given trajectory was dichotomized into (0) not a member of this trajectory and (1) a member of this trajectory. The genetic analysis of ordinal data assumes an underlying normal distribution and estimates the thresholds on the hypothesized normal distribution corresponding to the proportions of participants in the 2 categories (in the trajectory or not). Equality of thresholds is assumed between MZ and DZ twins. All analyses were conducted by using raw data to maximize power and treat missing data appropriately.$^{23}$

**RESULTS**

**Genetic and Environmental Influences of Daytime and Nighttime Sleep Duration**

Because of the small N and resulting low power, we reported only the full genetic ACE (A, additive genetic component of variance; C, shared environmental component of variance; E, unique environmental component of variance) models and did not attempt to test more parsimonious nested models. All models showed adequate fit when compared with their respective saturated models, except for the 30-month nighttime continuous sleep duration. Table 1 shows that as the child becomes a toddler, shared environmental influences explain a larger proportion of variance in daytime sleep duration (18 months = 33%, 30 months = 48%, and 48 months = 79%). In contrast, strong genetic influences were found for nighttime sleep duration at 6 months (47%), 30 months (58%), and 48 months (54%); however, we observed a strong shared environmental effect (on nighttime sleep duration at 18 months (48%).

**Sleep Duration Trajectories**

Daytime continuous sleep duration was moderately stable across ages ($r$ = 0.20–0.25; all correlations significant at $P < .001$). The best-fitting and most parsimonious model was a 3-trajectory model (BIC = −3262.20) in which 2 trajectories had a quadratic shape and 1

<table>
<thead>
<tr>
<th>TABLE 1</th>
<th>Genetic and Environmental Influence on Infant Daytime and Nighttime Sleep in the Quebec Newborn Twin Study</th>
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<tr>
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<td>Within-Pair Correlations</td>
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<td>6 mo*</td>
<td>0.67</td>
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<tr>
<td>18 mo*</td>
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<td>30 mo*</td>
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<td><strong>Trajectories</strong>$^{a,c}$</td>
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<tr>
<td>Rapidly decreasing daytime sleep$^d$</td>
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<td>Slowly decreasing daytime sleep</td>
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<td><strong>Nighttime continuous sleep duration</strong></td>
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<td>Short-increasing nighttime sleep$^d$</td>
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<td>10-h nighttime sleep</td>
<td>0.84</td>
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<td>11-h nighttime sleep</td>
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$^a$ Intraclass correlations with 95% CIs.

$^b$ Concordance rates.

$^c$ Tetrachoric correlations.

$^d$ Less than 5% of the sample in this category

$^e$ Assumption of equality of variance between MZ and DZ twins not respected.

$^f$ Assumption of equality of variance between MZ and DZ twins not respected.

$^g$ Assumption of equality of variance between MZ and DZ twins not respected.

$^h$ Assumption of equality of variance between MZ and DZ twins not respected.
had a linear shape. A small proportion of children (4.3%) were in the “rapidly decreasing daytime sleep duration” trajectory. Most children (75.7%) were in the “normally decreasing daytime sleep duration” trajectory. The remaining 20% of the children were in the “slowly decreasing daytime sleep duration” trajectory (see Fig 1A).

Nighttime continuous sleep duration was moderately stable across ages ($r = 0.31–0.40; P < .001$). The best-fitting and most parsimonious model was a 4-trajectory model (BIC = -4539.1) in which each trajectory had a quadratic shape. Small proportions of children were in the “short-persistent nighttime sleep duration” trajectory (4.9%) and in the “short-increasing nighttime sleep duration” trajectory (4.6%). Most children were in the “10-hour nighttime sleep duration” trajectory (47.6%) and the “11-hour nighttime sleep duration” trajectory (42.8%) of the sample (see Fig 1B).

**Genetic and Environmental Influences of Sleep Duration Trajectories**

All genetic-environmental models of daytime and nighttime sleep trajectories showed adequate fit when compared with their respective saturated model. The full genetic-environmental models showed strong common and unique environment influences for all daytime sleep duration trajectories, and no variance was attributed to genetic factors. In contrast, the models showed strong heritability for the variance in the nighttime short-persistent sleep and 10-hour sleep trajectories. A combination of genetic and common environment influences was found for the 11-hour sleep trajectory. Finally, only common and unique environment accounted for the variance in the short-increasing sleep trajectory.

**DISCUSSION**

To our knowledge, this is the first article to investigate daytime sleep duration longitudinally. These results illustrate a decrease of daytime sleep duration when the child grows up. However, we found a lower prevalence of children who had stopped taking a nap by age 4 years (4.3%) compared with that reported by an Italian study (68.0%)$^{10}$; this could be explained by different cultural influences. Indeed, we found that all patterns of daytime sleep duration are strongly influenced by shared environmental factors, as shown by Brescianini and colleagues$^6$ and by Fisher and colleagues.$^7$

The consolidated nighttime sleep duration trajectories of the present twin study replicated the number and shapes of other sleep trajectories found in a sample of singletons from the province of Quebec.$^{21}$ We found that consolidated nighttime sleep duration is largely influenced by genetic factors (at 6, 30, and 48 months) with a critical environmental time-window influence at $\sim 18$ months. To our knowledge, 2 other infant twin studies have demonstrated the strongest shared environmental influence on nighttime sleep duration at 18 months (64%$^6$ and 66%$^7$ compared with 48% in our study). The current study corroborates this important consistency. At 18 months, it could suggest a good timing for parental interventions on nighttime sleep.

A strong heritability (71%) was observed for the short-persistent nighttime sleep duration trajectory. A study investigating specific genes observed...
REFERENCES


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This study has a number of strengths, including the use of a relatively large sample of twins and of longitudinal assessments of daytime and nighttime continuous sleep in early childhood. One limitation important to mention is that the twin similarity may have been inflated by the recourse to a single informant for both twins of each pair, leading, in turn, to inflated heritability and shared environment estimates, even though the questionnaire for the second twin was filled out 2 weeks after that for the first twin. Another important limitation is the use of ordinal categories for daytime and nighttime sleep duration. This study needs to be replicated with quantitative sleep measures (actigraphy or polysomnography) to minimize informant bias and reduce the limitations induced by ordinal categories of daytime and nighttime sleep duration. In addition, one has to keep in mind that sleep is more fragmented than parents are aware of; however, brief awakenings are normal and, thus, the analysis of pure sleep consolidation is not what was targeted here. Finally, one of the genetic models, the 30-month nighttime continuous sleep duration ACE model, did not meet the assumption of equality of variance between MZ and DZ twins and should be interpreted with caution.

CONCLUSIONS

This study is the first to show that daytime sleep duration in early childhood is strongly influenced by environmental factors. In contrast, consolidated nighttime sleep duration is largely influenced by genetic factors with a critical environmental time-window influence at ~18 months. The role of genetic factors was especially evident for the short-persistent sleep duration trajectory. Results need to be replicated with objective sleep measures and more studies are needed to identify the biological mechanisms contributing to short-persistent nighttime sleep duration in early childhood. Future studies will be needed to understand which family settings seem to play a role in daytime sleep duration during early childhood. Future investigations will also be needed to identify the genetic mechanisms contributing to short-persistent nighttime sleep duration in early childhood and beyond to promote the development of adequate treatment modalities.
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