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Sleep Terrors in Children: A Prospective Study of Twins

Bich Hong Nguyen, MD*, Daniel Pérusse, PhD*, Jean Paquet, PhD*, Dominique Petit, PhD*, Michel Boivin, PhD*, Richard E. Tremblay, PhD*, Jacques Montplaisir, MD, PhD*

*Sleep Disorders Center, Sacre-Coeur Hospital, †Research Center, Sainte-Justine Hospital, and ‡Research Unit on Children’s Psychosocial Maladjustment, University of Montreal, Montreal, Quebec, Canada; §Research Unit on Children’s Psychosocial Maladjustment, Laval University, Quebec City, Quebec, Canada

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**What’s Known on This Subject**

There is growing evidence that genetic factors are involved in the occurrence of sleep terrors. Three studies have examined the prevalence and concordance of sleep terrors in twins, and only 1 dealt with children specifically.

**What This Study Adds**

We determined the precise contribution of genetic and environmental factors to the manifestation of sleep terrors in a large cohort of early-childhood twins followed prospectively. In addition, we determined the prevalence of sleep terrors at the age at which they emerge.

**ABSTRACT**

OBJECTIVE. There is growing evidence that genetic factors are involved in the occurrence of sleep terrors. Twin studies provide invaluable information regarding genetic and environmental factors that can affect the manifestation of the disease; however, most previous twin studies on sleep terrors were performed retrospectively or with a sample that was too small to yield conclusive results. The aim of this large prospective study was to clarify the genetic and environmental contributions to sleep terrors in childhood.

METHODS. In all, 390 pairs of monozygotic and dizygotic twins were recruited at birth for a longitudinal study. The prevalence and frequency of sleep terrors were assessed at 18 and 30 months of age with a questionnaire administered to the biological mother of the twins. Zygosity was determined by a questionnaire and genotyping. The prevalence and polychoric correlation for each type of twins were calculated. Structural-equation modeling was used to determine the proportion of variance attributable to additive genetic, shared, and nonshared environmental factors.

RESULTS. The prevalence of sleep terrors was 36.9% at 18 months and 19.7% at 30 months; 49% of affected children were boys, and 51% were girls. At 18 months, the polychoric correlations were 0.63 for monozygotic and 0.36 for the dizygotic twins. These were 0.68 (monozygotic) and 0.24 (dizygotic) at 30 months. Our model-fitting analysis showed that sleep terrors were explained by a 2-component model at 18 months (43.7% additive genetic effects and 56.3% nonshared environment) and at 30 months (41.5% additive genetic effects and 58.5% nonshared environment).

CONCLUSIONS. These results strongly support the heritability of sleep terrors. There also seems to be continuity in genetic effects with the persistence of sleep-terror symptoms. *Pediatrics* 2008;122:e1164–e1167

Sleep terrors (also referred to as night terrors, pavor nocturnus [in children], and incubus attacks [in adults]) are a common childhood parasomnia. They represent the most dramatic disorder of arousal. The onset of sleep terrors is abrupt and frightening, usually sudden arousal with screaming. Sleep terrors are associated with intense autonomic discharge: tachycardia, tachypnea, flushing, diaphoresis, and mydriasis. The universal feature of this disorder is inconsolability. During these events, children seem confused and disoriented. Any attempt to awaken them may increase their agitation and prolong their episode. These events are brief and cease abruptly, and the child returns to a deep sleep and is amnesic about the episode. These events usually occur within 2 hours of sleep onset and are a result of a partial arousal from deep slow-wave sleep.

Diagnosis is based on the identification of these symptoms and exclusion of organic pathologies. The exact prevalence of sleep terrors is unknown. There are major discrepancies in the literature (as a result of different sampling methods, sample age, and definitions of sleep terrors). One study reported an overall prevalence of 19.2% for children between 4 and 9 years of age. In a population-based study of parasomnias in children, the overall prevalence of sleep terrors between the ages of 3 and 10 years (measured retrospectively) was estimated at 14.7% for children. Prevalence was estimated prospectively thereafter to be 3.8% at 11 years, 2.3% at 12 years, and 1.2% at 13 years. The peak of prevalence occurs during childhood, and the resolution of sleep terrors is typically before adolescence; however, few studies, if any, studied prospectively the prevalence of sleep terrors in very early childhood.
Precipitating factors of sleep terrors are physical stress such as fever, nocturnal asthma, gastroesophageal reflux, sleep deprivation, and central nervous system medications. There is, however, growing evidence that genetic factors are involved in the occurrence of sleep terrors. Familial aggregation is found, and these studies suggest an autosomal dominant mode of inheritance. Kales et al reported that the prevalence of sleepwalking and sleep terrors was ~10 times greater in first-degree relatives of affected patients than in the general population. Twins studies can provide invaluable information regarding the relative contribution of genetic and environmental factors. Three studies looked specifically at the prevalence and concordance of sleep terrors in twins, and only 1 studied children directly. Hublin et al conducted a retrospective study by using an adult Finnish twin population and reported a higher polygenic correlation for childhood sleep terrors in monozygotic twins (males: 0.38; females: 0.35) than in dizygotic twins (males: 0.17; females: 0.18). In a retrospective study of 881 pairs of junior high school Japanese monozygotic and dizygotic twins, results showed a moderate to strong contribution of genetic factors for sleep talking, sleepwalking, and sleep terrors. Finally, a higher concordance rate was found for sleep terrors among 47 pairs of monozygotic twins than for the 14 pairs of dizygotic twins (15% vs 0%; P = .05) in a sample of 3- to 8-year-old children.

The objective of this study was to determine the relative contribution of genetic and environmental factors to the manifestation of sleep terrors in a large cohort of preschool twins who were followed prospectively. This study will also allow determination of the prevalence of sleep terrors at the age at which they are more likely to emerge (~1.5–3.0 years).

Methods

Subjects

Subjects for this study were monozygotic and dizygotic twins who were born between November 1995 and July 1998 in Montreal, Canada. These participants were part of an ongoing longitudinal study, the Quebec Newborn Twin Study. The descriptive statistics on prevalence were based on 887 twins at 18 months and 796 twins at 30 months. Our genetic–environment model-fitting analyses were based on results that were obtained from 161 pairs of monozygotic and 229 pairs of dizygotic twins at 18 months of age and 140 pairs of monozygotic and 207 pairs of dizygotic twins at 30 months of age (pairs for whom we had no missing data on the main variable). For same-gender twin pairs, zygosity was evaluated at 5 and 18 months of age by using a physical resemblance questionnaire and genotyping of 8 to 10 highly polymorphic genetic markers. Eighty-four percent of the families were of European descent, 3% were of African descent, 2% were of Asian descent, 2% were Native North Americans, and the remaining families (9%) did not provide ethnic information; 49% of the subjects are boys. All twin pairs were reared together.

Before participating in the study, all families had received detailed information by mail on the aims and procedures of the research program and had signed a consent form. The study was approved by the ethics committee of the Sacre-Coeur Hospital of Montreal (affiliated with the University of Montreal). This research was conducted in accordance with the ethical standards of International Conference on Harmonisation and with the Declaration of Helsinki.

Measures

The sleep portion of the Quebec Newborn Twin Study contained 18 sleep-related questions. Sleep terrors were assessed by asking the biological mother to determine the frequency of sleep terrors experienced by the twins prospectively at 18 and 30 months of age: “Does your child have sleep terrors (this means sudden arousal with screams, sometimes with confusion and sweating)? Please circle one of the following answers: 1-never, 2-sometimes, 3-often, or 4-always.”

Statistical Analyses

The prevalence of sleep terrors was first calculated at 18 and 30 months of age. The categories “often” and “always” were combined together as “often-always” for statistical reporting (Table 1). Age and gender effects were assessed with $\chi^2$ tests. Then, the correlation in disorder tendency between the 2 members of each type of twin pairs (monozygotic and dizygotic) was computed by using SAS 9.1 (SAS Institute, Inc, Cary, NC) and represents the polygenic correlation. The polygenic correlation estimates the bivariate normal distribution that most closely approximates the cell probabilities from a contingency table. Finally, a structural equation modeling by using a maximum likelihood fit function applied to the twin variances-covariances matrices was performed to obtain a formal estimation of the genetic and environmental parameters. Under the current design of twins reared together, it is possible to model 3 different parameters: an additive genetic component (A), shared or common environmental components (family, living conditions, physical environment) (C) and nonshared (or unique to a single twin) environmental components (eg, hospitalization, illness) (E) that could explain the variation in the tendency to express the disorder (phenotypic variance). One can, thus, test different models that represent different combinations of factors (genetic, shared environmental, and nonshared environmental; genetic and nonshared environmental; shared environmental and nonshared environmental; and nonshared environmental). In the first series of analyses, such univariate estimates of variance decomposition were calculated by using the Mx statistical package. To determine the best-fitting and most parsimo-

| Table 1 Frequency of Sleep Terrors According to Age |
|-----------------|-----------------|-----------------|-----------------|
| Age, mo         | Never, n (%)    | Sometimes, n (%)| Often-Always, n (%) |
| 18 (N = 887)    | 560 (63.1)      | 314 (35.4)      | 13 (1.5)        |
| 30 (N = 796)    | 639 (80.3)      | 146 (18.3)      | 11 (1.4)        |
nious model given the pattern of intercorrelations observed within twin pairs, the model fit was assessed on the basis of the \( \chi^2 \) statistics and the Akaike information criterion (AIC). The AIC was based on the likelihood and not on the \( \chi^2 \) statistics. The lowest AIC value indicates the best combination of model fitting and parsimony.

**RESULTS**

The prevalence of sleep terrors (ie, the mother responded as either sometimes, often, or always) was 36.9% at 18 months of age and 19.7% at 30 months of age. The number and percentage of children in each frequency of occurrence category are reported in Table 1 for both 18 and 30 months. There was a significant age effect (\( \chi^2 = 60.2, P < .0001 \)) on the reported prevalence of sleep terrors; this parasomnia was almost twice as frequent at 18 months than at 30 months. Among the 140 cases that presented sleep terrors at 30 months (and for whom the data were available within twin pairs, the model fit was assessed on the basis of the \( \chi^2 \) statistics and the Akaike information criterion (AIC). The AIC was based on the likelihood and not on the \( \chi^2 \) statistics. The lowest AIC value indicates the best combination of model fitting and parsimony.

The polychoric correlation was 0.63 for the monozygotic and 0.36 for the dizygotic twins at 18 months of age. At 30 months of age, the polychoric correlation was almost twice as frequent at 18 months than at 30 months. Among the 140 cases that presented sleep terrors at 30 months (and for whom the data were available for 18 months), 52 (37.1%) were new cases and 88 (62.9%) were persistent cases; however, no gender difference was found at either 18 (\( \chi^2 = 0.07, P = .80 \)) or 30 (\( \chi^2 = 2.51, P = .11 \)) months of age.

The polychoric correlation was 0.63 for the monozygotic and 0.36 for the dizygotic twins at 18 months of age. At 30 months of age, the polychoric correlation was 0.68 for the monozygotic and 0.24 for the dizygotic twins. Results for the model-fitting analysis are shown in Table 2. Sleep terrors were best explained by a 2-component model (genetic and nonshared environmental). At 18 months, genetic accounted for 43.7% and nonshared environmental accounted for 56.3% of the phenotypic variance. At 30 months, genetic and nonshared environmental similarly accounted for 41.5% and 58.5% of the variance, respectively.

**DISCUSSION**

To our knowledge, this is the largest prospective twin cohort (390 pairs of twins) ever studied on the genetic–environmental etiology of sleep terrors. In addition, no study of twins has ever reported the prevalence of sleep terrors so early in life. The prevalence of sleep terrors is high in infants (36.9% at 18 months) but decreases to 19.7% by 30 months of age. Our prevalence at 30 months is similar to the prevalence found in a smaller sample of twins aged 3 to 8 years (19.1%). It is also similar to that of a recent study, which found a prevalence of 19.9% in a population of 2.5-year-old single-birth children also from the province of Quebec, Canada. Thus, twins do not show a different prevalence of sleep terrors than single-birth children.

The results of this study also confirm that sleep terrors are a partially hereditary parasomnia. Genetic factors play an important role in the manifestation of sleep terror at a very young age. Of the total phenotypic variance in sleep terrors, the proportions that were attributable genetic influences were >40% for both 18- and 30-month-old twins. The polychoric correlations found for our cohort are higher than those published by Hublin et al, (monozygotic male: 0.38, female: 0.35; dizygotic male: 0.17, female: 0.18); however the latter study was a retrospective study, and information bias may have accounted for the difference in the polychoric correlations observed. In addition, the childhood period in that study covers up to ~15 years of age, whereas our results are for preschool children. Our polychoric correlations were also higher than those found in the study by Abe et al. Again, the children in the latter study were older than those of this study, and the results were based on small samples of monozygotic and dizygotic twins. On the basis of heritability, this study suggests that there also seems to be continuity in genetic effects with the persistence of sleep-terror symptoms (at least to the age of 30 months).

The role of nonshared environmental factors was also significant in our study (>55% of the variance), although this percentage also includes measurement error. In our cohort of twins, all pairs were reared together; therefore, they mostly shared the same postnatal environment. One of the twins may still have undergone a prolonged hospitalization, may have had a comorbid disease, may have received medication, may have been a poorer sleeper, or may have been of smaller birth weight than the other twin (nonshared environmental). It must be kept in mind that the way that an individual reacts to a given common environmental event (eg, both twins may have been hospitalized) may also be a factor in the equation of why 1 twin only would

<table>
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<th>Age, mo</th>
<th>Model</th>
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<th>df</th>
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\( df \) indicates degrees of freedom; \( a^2 \), variance attributed to additive genetic influence (heritability); \( c^2 \), variance attributed to common environment; \( e^2 \), variance attributed to unique environment; ACE, the phenotype is attributed to both genetic influence and common environment, and unique environment; AE, the phenotype is attributed to both genetic influence and unique environment; CE, the phenotype is attributed to both common environment and unique environment (no genetic influence). E, the phenotype is attributed to the unique environment only.
develop sleep terrors and not the other twin. To that effect, Laberge et al.\textsuperscript{10} found an association between anxiety and the occurrence of sleep terrors.

This study has some limitations. Our data were obtained from parental reports, and children with a history of sleep terrors did not have objective sleep laboratory evaluations to validate their diagnosis. Although our questionnaire contained an operational definition for sleep terrors, it is nevertheless possible that some parents mistook nightmares for sleep terrors, and vice versa. It is also possible that some sudden awakenings with crying were mistaken for sleep terrors. The results therefore should be interpreted with caution, and the prevalence of sleep terrors in early childhood should be reconfirmed in large prospective studies.

**CONCLUSIONS**

Despite these limitations, the large sample size allowed the testing of models that assessed the role of genetic and environmental factors. Our results show that there is a substantial effect of genetics factors in sleep terrors. The findings also demonstrate that half of the children who experience sleep terrors at the age of 18 months will not experience this problem at 30 months, although the gene–environment etiology was found to be essentially the same at both times. To date, no specific genes have been identified for sleep terrors. Additional studies are needed; however, this large prospective twin study is the first step toward identifying susceptibility genes for sleep terrors.

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