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Hereditary Factors in Sleepwalking and Night Terrors

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SUMMARY The families of 25 probands with sleepwalking and 27 probands with night terrors were studied. Eighty per cent of the sleepwalking pedigrees and 96 per cent of the night terror pedigrees included one or more individuals, other than the proband, who were affected by sleepwalking, night terrors, or both. Our data appear to fit a 'two threshold' multifactorial mode of inheritance. This finding supports the hypothesis that sleepwalking and night terrors share a common genetic predisposition, with sleepwalking being a more prevalent and less severe manifestation of the same substrate that underlies night terrors. Heritable factors predispose an individual to develop sleepwalking and/or night terrors, but expression of the trait may be influenced by environmental factors.

Sleepwalking and night terrors are associated with impaired arousal (Broughton, 1968) and usually occur early in the night when slow wave sleep (stages 3 and 4) predominates (Gastaut and Broughton, 1965; Kales *et al*, 1966; Fisher *et al*, 1973). Sleepwalking, or somnambulism, is an episodic state in which the phenomena of sleeping and waking are combined and the individual wanders about, appearing dazed and uncoordinated. Night terrors are episodic events of extreme vocalization, motility and autonomic discharge that occur suddenly out of sleep. Both disorders occur more frequently in childhood than in adulthood and appear to be related to a delay in the maturation of the central nervous system (Jacobson and Kales, 1967). Although most children outgrow these disorders, in some cases the disorder continues into adulthood; in these individuals there is often increased evidence of psychopathology (Kales *et al*, 1978a, b).

It has been estimated that sleepwalking occurs in 1–6 per cent of different patient populations (Kleitman, 1963), 2.2 per cent in healthy young adults (Thomas and Pederson, 1963), and in 2.5 per cent of the general population (Bixler *et al*, 1979). Males and females seem to be equally affected. The prevalence of night terrors in the general population is not known, but in a group of patients referred to a child psychiatry clinic, about 1.5 per cent of the children had night terrors (Shirley and Kahn, 1958).

Several authors have noted a familial predisposition to sleepwalking and night terrors. Abe and Shimakawa (1966) found an increased incidence of somnambulism in children of parents who had histories of sleepwalking and suggested "a recessive mode of inheritance with incomplete penetrance". Bakwin (1970) noted that monozygotic twins were concordant for the disorder six times as often as dizygotic twins. He observed that near relatives of sleepwalkers had a greater likelihood of having a history of the disorder than did more distant relatives or non-family members, and concluded that sleepwalking may have a heritable component.

Night terrors also may occur in multiple family members. Hallstrom (1972) reported three affected individuals in three consecutive generations, and suggested an autosomal dominant pattern of heritability.

The hypothesis that sleepwalking and night terrors may be variable expressions of the same clinical entity is supported by the facts that (1) they are both disorders of impaired arousal

occurring out of slow wave sleep, (2) they share common clinical manifestations, and (3) they both appear to be related to CNS immaturity in children and psychopathology in adults. This paper reports family studies in which we have attempted to determine if sleepwalking and night terrors have a predictable mode of inheritance.

Subjects and Methods

The families of 52 patients referred to a Sleep Research and Treatment Center because of sleepwalking (N = 25) or night terrors (N = 27) were studied. A detailed family history was obtained from each proband and the resultant pedigree was confirmed and expanded by interviewing two or more additional family members from generations other than the proband's. The ascertainment was single and incomplete (Fisher, 1934). The families studied were classified as sleepwalking (SW) or night terror (NT) pedigrees based on the presenting complaint of the proband. In cases where the proband presented with both complaints, he/she was classified as having sleepwalking or night terrors based on the predominant current symptomatology. Each pedigree was then assessed for the presence of SW, NT, or both.

An individual was determined to be a sleepwalker, either currently or in the past, according to the following criteria: (1) he was repeatedly observed to get up at night and wander about, apparently asleep; (2) during the episode, he appeared dazed and poorly co-ordinated and was difficult to arouse, and (3) if aroused, he was confused and disoriented and had very little or no recall of the event.

The presence of night terrors, either currently or in the past, was ascertained as follows: (1) the individual was repeatedly observed to suddenly sit up, or get up while asleep, and have intense vocalization (usually screaming or crying) and motility (thrashing about, behaving as if frightened, etc.) and, (2) if aroused, he was confused and disoriented and had very little or no recall of the event. The diagnostic criteria were applied independently by two investigators and both investigators had to agree on the diagnosis for an individual to be considered to have sleepwalking or night terrors.

Segregation analysis was applied to the pedigree data to test for autosomal dominant and recessive modes of inheritance. The pedigree data were also evaluated from the point of view of the Falconer (1965) model for multifactorial inheritance using methods described by Reich *et al* (1972, 1979).

In the segregation analysis, it was assumed that sleepwalking and night terrors were independent. Thus, a given individual could be considered twice, once for sleepwalking and once for night terrors, if both sleepwalking and night terrors were present. Within the analysis using the multifactorial models, each individual was assumed to have only the most severe symptom. Thus, in this case, each subject could be counted only once.

Results

Fifty-two families were evaluated; 25 pedigrees with sleepwalking (SW) and 27 pedigrees with night terrors (NT). None of the 52 families was related. Two of the 25 probands with sleepwalking also had a history of night terrors, whereas 12 of the 27 probands with night terrors also had a history of sleepwalking. Representative pedigrees are shown in Fig 1.

In five of the 25 SW pedigrees, the proband was the only affected individual. In the remaining 20 families, one or more family members other than the proband were affected by sleepwalking, night terrors, or both. In 14 of these 20 families, the relatives were affected only with SW, and in five families, several individuals had either sleepwalking or night terrors. In the remaining family, individuals were affected by sleepwalking, night terrors, or both.

Of the 27 NT pedigrees, 26 (96 per cent) had one or more family members other than the proband with either sleepwalking, night terrors, or both. In 11 of these 26 pedigrees, the affected relatives had only sleepwalking, and in one family, one relative had only night terrors. In three families, affected relatives had both sleepwalking and night terrors. In four families, some relatives had sleepwalking alone while others had night terrors alone, and in seven families, there were various combinations of sleepwalking, night terrors, or both.

Of all the affected individuals in the 52

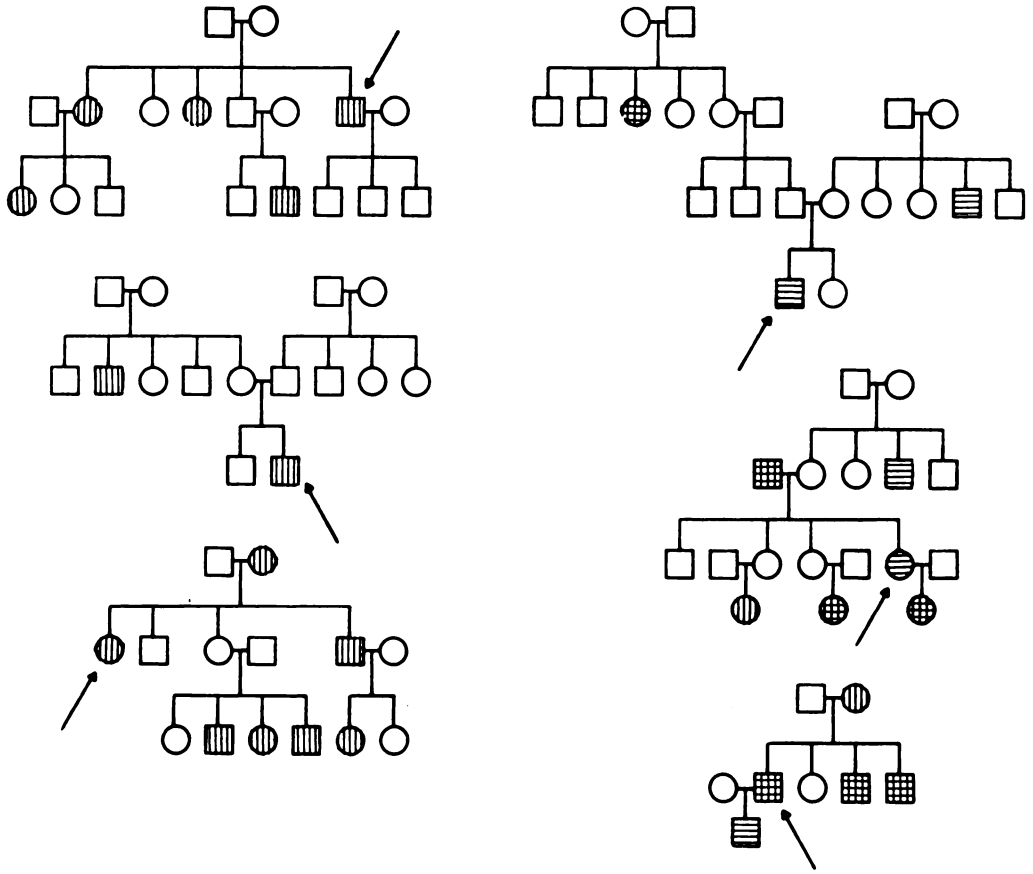

FIG 1.—Examples of pedigrees observed. The proband is identified by an arrow. The three pedigrees on the left represent families of probands with the presenting complaint of sleepwalking, the three on the right represent families of probands with a presenting complaint of night terrors. Squares represent males and circles, females. Vertical shading indicates sleepwalking, horizontal shading, night terrors and cross hatching, the presence of both conditions.

pedigrees, 79 had spouses. Both spouses were affected in only three of these couples. In two of these couples, the wife had sleepwalking and the husband had both sleepwalking and night terrors. Interestingly, both were in the same pedigree. In the third couple, the wife again had sleepwalking alone while the husband had night terrors alone. No twins were noted in any of the families.

Incidence in First, Second, and Third Degree Relatives

The incidence of SW and NT in first, second and third degree relatives is summarized in Table I. First degree relatives of SW individuals were found to have at least a ten-fold greater likelihood of being affected with SW than the general population. Second and third degree relatives also showed a greater risk. These observations clearly support an increased familial occurrence of these two sleep disorders. When Edwards' empirical formula (1969) was applied to our data, a much higher incidence was observed in first degree relatives than would be predicted. Similar observations were made for night terrors in the NT pedigrees and for either sleepwalking or night terrors in both SW and NT pedigrees. In contrast to SW pedigrees, where the incidence of sleepwalking fell sharply with decreasing degrees of relationship, the

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TABLE I

Incidence of sleepwalking and/or night terrors in first, second and third degree relatives of probands with sleepwalking or night terrors

	Incidence in 1° relatives	Incidence in 2° relatives	Incidence in 3° relatives	Edwards' empirical risk in 1° relatives $\sqrt{Q}$
Sleepwalking in relatives of sleepwalking probands	0.26	0.12	0.04	0.16
Night terrors in relatives of night terror probands	0.21	0.11	0.14	0.12
Sleepwalking and/or night terrors in relatives of probands with either sleepwalking or night terrors	0.32	.15	0.07	0.20

second and third degree relatives in NT pedigrees showed essentially the same frequency of night terrors. These observations suggest that factors other than genetic may play a significant role in the expression of these sleep disorders, particularly in night terrors.

Single-Gene Mode of Transmission

Sex-Linked: We determined the proportions of affected parents and offspring to estimate the relative proportions of affected near relatives (Table II). In these families the sex ratio of affected individuals did not significantly differ from unity. In both sleepwalking and night terrors, there was an increased likelihood that one parent of an affected person would be affected. Mothers of both male and female siblings with sleepwalking had at least a two-fold greater chance of having sleepwalking than fathers. A similar effect was noted for female siblings with night terrors. In no instance were both parents affected in NT pedigrees, whereas in three SW families, both parents were affected. In the latter three families, both normal and affected siblings were found. When the pedigrees were considered in terms of sleepwalking and/or night terrors being present, mothers of both males and females were affected by either condition more than fathers.

Autosomal Dominant: Segregation analysis was applied to evaluate the pedigrees for a single gene mode of inheritance (Table III). To determine the probability that sleepwalking may be an autosomal dominant trait, 41 sibships out of the 52 pedigrees were identified in which one parent was affected with sleepwalking and the other was apparently normal and healthy. Out of a total of 127 offspring with one parent affected, 43 (34 per cent) were similarly affected. Twenty-nine sibships of the 52 pedigrees were identified in which one parent was affected with night terrors and the other was not. Out of a total of 87 offspring, 29 (33 per cent) were affected. Assuming autosomal dominance for either group, 50 per cent would have been expected to be affected. With one degree of freedom, chi square should exceed 3.84 to indicate a significant disagreement between the observations and the hypothesis tested. The resulting chi square values (13.2 and 9.7 for SW and NT, respectively) therefore suggest that an autosomal dominant mode of inheritance for sleepwalking or night terrors is unlikely.

Autosomal Recessive: The single incomplete ascertainment method (Fisher, 1934) was applied to the data to determine the probability that these disorders may result from an autosomal recessive mode of inheritance. Forty-four sibships from the 52 pedigrees were selected in which neither parent was affected with sleepwalking. Sixty-three offspring were affected out of a total of 166 with normal parents (16 per cent). Similarly, 24 sibships were identified where neither parent was affected with night terrors. Twenty-nine offspring were affected out of 68 (11 per cent). Twenty-five per cent would have

TABLE II

Occurrence of sleepwalking and night terrors in parents of affected individuals

	Affected offspring	Affected fathers	Affected mothers	One parent affected	Both parents affected
Sleepwalking in parents of sleepwalking offspring					
Males:	62	9 (15%)	20 (32%)	25 (40%)	2 (3%)
Females:	55	5 (9%)	16 (29%)	19 (34%)	1 (2%)
Night terrors in parents of night terror offspring					
Males:	33	10 (30%)	6 (18%)	16 (48%)	0
Females:	25	3 (12%)	11 (44%)	14 (56%)	0
Sleepwalking and/or night terrors in parents of offspring with either sleepwalking or night terrors					
Males:	78	18 (23%)	35 (45%)	43 (55%)	5 (6%)
Females:	72	12 (17%)	31 (43%)	41 (57%)	1 (1%)

TABLE III

Segregation analysis summary

	Number			
	Sibships	Total	Affected	(%)
Dominant				
Sleepwalking	41	127	43	(34)†
Night terrors	29	87	29	(33)†
Recessive				
Sleepwalking	44	166	63	(16)*
Night terrors	24	68	29	(11)*

† $P < .01$ different from autosomal dominant model.
* $P < .05$ different from autosomal recessive model (single incomplete).

been expected assuming autosomal recessive inheritance. The resulting chi square values (5.78 and 4.36 for SW and NT respectively) suggest that the autosomal recessive model for the inheritance of sleepwalking or night terrors is also unlikely.

Multifactorial Mode of Transmission

Reich (1979) has described three different multifactorial models of disease transmission which could account for our data. These models are isocorrelational, environmental and independent. Four correlations of phenotypic liability are referred to test these models. Using our data, these are: (1) correlation between SW/NT probands and SW/NT relatives (r_{11}), (2) correlation between NT probands and NT relatives (r_{33}), (3) correlation between SW/NT probands and NT relatives (r_{13}), (4) correlation between NT probands and SW/NT relatives (r_{31}). In addition, the correlation between familial determinants of SW and NT (r_p) is required.

The four correlations of phenotypic liability were calculated for all first degree relatives of the night terror and sleepwalking probands (Table IV). The environmental and independent models of Reich assume that correlations of phenotypic liability of first degree relatives are dissimilar, whereas they are similar in the isocorrelational model. Since our four correlations are approximately equivalent, the isocorrelational model was accepted and the other two rejected. Given these correlations, the extent of overlap of the familial factors between these two disorders was found to approach unity ($r_p = .89$). These data support the hypothesis that familial transmission relevant to the development of both sleepwalking and night terrors is similar.

As a further evaluation of the isocorrelational model, the four correlations were calculated for each type of first degree relative (offspring, siblings and parents). The purpose of these correlations was to determine whether the transmission was consistent across types of

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TABLE IV
Phenotypic correlation in liability

	First degree relatives			
	Total	Offspring	Siblings	Parents
r_{11}	.64 ± .06	.82 ± .12	.60 ± .06	.56 ± .08
r_{ss}	.56 ± .08	.62 ± .13	.59 ± .10	.50 ± .10
r_{1s}	.45 ± .07	.49 ± .12	.43 ± .08	.46 ± .09
r_{s1}	.62 ± .07	.68 ± .09	.64 ± .11	.50 ± .10

relatives. Within and between sets of first degree relatives, the correlations were consistently approximately equivalent, with the one exception being r_{11} within the offspring. All four correlations for the offspring were higher than those obtained for the other types. This may reflect an increased environmental role or a possible ascertainment problem or may be suggesting an intrauterine effect.

Finally, there appears to be a strong relationship between an affected mother and affected offspring. The correlations relating affected offspring of either sex to affected father (r_{mm} = .60 ± .10 and r_{fm} = .50 ± .09 for male and female offspring, respectively), were similar to each other as well as similar to the previously reported correlations. In contrast, the correlations relating affected male and female offspring to affected mothers (r_{ff} = .80 ± .07, r_{mf} = .82 ± .08 for female and male offspring, respectively), although again similar to each, were considerably higher than those previously reported.

Discussion

Prevalence of sleepwalking and night terrors in first degree relatives of an affected individual is at least ten times greater than that in the general population. However, our data were collected under circumstances different from those reported elsewhere for the general population. This difference should be taken into account when considering our interpretations.

Each family in our study was referred because of one affected person with well defined clinical symptoms. Ascertainment of other affected family members was based on information given to us by at least two or more family members who were themselves often affected. Sleepwalking and night terrors are generally assumed to be 'all-or-none' phenomena and sufficiently characterized so as to be readily recognizable. Thus, the diagnosis of these conditions in various family members on the basis of the way in which the histories were obtained, may be accepted as being reasonably reliable.

In our review of the pedigrees of 25 families with sleepwalking and 27 with night terrors, we found remarkable variation in the apparent pattern of heritability. The high prevalence of night terrors in pedigrees of sleepwalking probands and of sleepwalking in the pedigrees of night terror probands and the neurophysiological and clinical similarities between the two conditions suggest that they have a common genetic and neurophysiological substrate. Moreover, individuals may develop both conditions at the same time or at different times in their lives. In some families, affected members appeared in successive generations, and in others, one or even multiple affected siblings had apparently normal parents. In several families, affected individuals were scattered throughout the pedigrees with apparent skipped generations. These findings indicate the remarkable heterogeneity of these traits in the families studied.

Pedigrees were selected for segregation analysis on the basis of an apparent autosomal dominant or recessive mode of transmission. These analyses indicated that a single gene mode of inheritance was unlikely. In those families with affected members scattered among different generations, a markedly reduced level of penetrance of the trait would be required to explain a dominant gene effect. It remains to be determined if some of the families do in fact represent the expression of a single gene trait.

Visual inspection of these pedigrees suggested from the start a possible multifactorial basis, and several general findings from both the literature and the current study support this hypothesis. A number of studies have shown that both night terrors and sleepwalking arise from an identical physiological substrate (i.e., they are both disorders of arousal out of slow

wave sleep) and that they share common clinical manifestations. Night terrors have been shown to be more severe than sleepwalking, however, in terms of physiological parameters such as heart rate and GSR, and in the higher degree of general psychological disturbance (measured by the MMPI) among adults with these disorders. While night terrors are less prevalent than sleepwalking in the general population (1.5 per cent and 2.5 per cent, respectively), the proportion of affected first degree relatives within the current study was greater among NT probands (29 per cent vs 34 per cent for SW and NT probands, respectively). These findings are in agreement with the concept that the NT probands would be more extreme cases along the liability continuum. The proportion of affected offspring within our sample increased in relation to the number of affected parents (i.e., 22 per cent when neither, 45 per cent when only one, and 60 per cent when both parents are affected), supporting the hypothesis that there are more abnormal genes present if both parents are affected than if neither or only one parent is affected.

When our data were tested using the multifactorial models described by Reich, they appeared to fit a 'two threshold' isocorrelational multifactorial model of inheritance (Reich, 1979; Cloninger *et al*, 1978; Falconer, 1965; Carter, 1976; Leckman and Gershon, 1976). While application of this model provides a test of this hypothesis, it cannot unequivocally establish the mode of inheritance involved in the transmission of these traits within families. Using the same model, a similar relationship between REM narcolepsy and hypersomnia has been previously suggested (Leckman, 1976; Kessler, 1974).

Our data imply an increased 'liability' of relatives of an affected individual to develop sleepwalking and night terrors. On the basis of Bakwin's twin study (1970) and our family studies, it appears that heritable factors may predispose an individual to these sleep disorders. However, the relatively high frequency of these disorders in near relatives may also be influenced by environmental factors that may precipitate the expression of the trait. Although the relative importance of environmental factors has yet to be studied in our families, a high degree of psychopathology occurs in adults with these conditions (Kales *et al*, 1978a, b). Thus, it is highly probable that multiple factors, both genetic and environmental, influence the expression of the trait.

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