Severe Sleep Problems among Infants

A Five-Year Prospective Study

BY

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ABSTRACT


The aim of this thesis was to explore the prevalence of parentally experienced infant sleep problems, with special interest in severe problems, in a total community sample of 2,518 infants aged between 6 and 18 months. Factors associated with severe sleep problems were sought. Parents reported 16% of the infants to have difficulties in falling asleep at night, and 30% to have frequent night waking. Severe sleep problems were associated with frequent night meals, psychosocial problems in the family, exhaustion and depression in the mother, and parental stress. An association with infant difficulty, high activity and problematic behaviour was also found.

In a five-year prospective study a group of children fulfilling specific criteria for severe sleep problems in infancy (N=27) was followed after an interventional sleep programme and compared with a control group regarding sleep characteristics, behaviour and development. One month after an interdisciplinary treatment programme, combining behavioural technique with family work, the average number of times the case babies woke up had diminished from 6.0 to 1.8 times per night. A 92% rate of improvement was reported.

The changes were stable over time. Comparisons with the controls during five years revealed no significant group difference in sleep characteristics. Concerning behaviour and development, however, there were significant differences. At the age of 5.5 years, seven of the children in the former sleep problem group met the criteria for the diagnosis of attention-deficit/hyperactivity disorder. No control child qualified for the diagnosis.

Close follow-ups of infants with combined severe sleep and behavioural problems are recommended.

Key words: Sleep disorders, infant, behavioural therapy, psychosocial problems, attention-deficit/hyperactivity disorder.

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To all sleepy children
LIST OF PAPERS

This thesis is based on the following papers, which will be referred to in the text by their Roman numerals:

I  Thunström M.

II Thunström M.

III Thunström M.

IV Thunström M.
Severe sleep problems in infancy associated with subsequent development of attention-deficit/hyperactivity disorder at 5.5 years of age. Acta Paediatrica. Accepted for publication.

V Thunström M.
A 5-year follow-up of infants with severe sleep problems. Manuscript.

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INTRODUCTION

General background

Sleep plays a major role in children’s well-being and is strongly influenced by the child’s health status, psychological stress and family issues as well as by multiple aspects of his or her culture and environment. Also, children’s sleep patterns affect their well-being within the same wide range of health and psychosocial phenomena.

Research has shown that sleep problems in infants and young children are prevalent. Cross-sectional studies show that between 15 % and 35 % of all children aged between 6 months and 5 years are reported by their parents to show some type of sleep disturbance (22,69,76,98,100,102).

Before six months of age it is doubtful whether the label "sleep disturbance" should be used, since during this period night waking is necessary in order to fulfil the needs of frequent nutrition of the child. This is also the period needed to reorganize the sleep pattern of the newborn to a more mature pattern (85).

Maturation of normal sleep pattern in childhood

Sleep and sleep stages are defined by specific patterns of electrophysiologically recorded brain activity, muscle tone and eye movements. These polysomnographic measures divide sleep into two principal states: sleep associated with rapid eye movements, "REM sleep", and sleep without rapid eye movements, "non–REM sleep" (28). The REM component of sleep, which dominates during the foetal period and the early months of life (50 % of the sleep time), is thought to play a crucial role with regard to brain
The amount of REM sleep decreases to 30% during the first year of life and the REM–non REM state cycle also undergoes maturational changes. In newborns and very young infants the cycle lasts approximately 40 to 60 minutes. At two years of age the cycle has increased to 75 minutes and continues to increase to an average of 84 minutes in 5-year-olds (65).

A newborn baby does not distinguish day from night. Both sleeping and eating take place around the clock, equally distributed over a 24-hour-period, and the two processes are linked in time to each other. The baby wakes up to eat and most often goes back to sleep while suckling and being fed at the breast.

Gradually, in the course of the baby’s first year, a transition to a more regular pattern of eating and sleeping takes place (10). The child acquires a pattern of regular meals during daytime, a period of prolonged sleep during the night and one, or most often two, short naps during daytime. Eating and sleeping are increasingly separated, and, normally, the one-year-old child has grown out of the habit of going to sleep while eating.

Temporary nightwaking in occasional periods of short duration is very common among infants (42), but in the absence of any current physical disease, a normal development and maturing of the central nervous system (10) and normal parental handling (10,125), 90% of one-year-olds sleep all night (85). Prolonged and complicated sleep problems, however, have a tendency to remain, especially from the first year of life to later childhood (1,21,67,92). Moreover longitudinal studies (20,41,60,115) have indicated that the attitudes and (over-) responsiveness of the parents to the signals of the child in the parent–infant interaction could play a role in the maintenance of the sleep problems of the child.
Other possible causative and maintaining mechanisms that have been studied include factors in the genetic, somatic, psychological and psychosocial areas (19-20,32,61,81,89). Prolonged, complicated sleep problems show a picture of multiproblem, where the separate factors seem to amplify each other in vicious circles. Factors affecting the child, the stressed and exhausted parents and the social situation probably influence each other reciprocally and interfere with the normal maturation of the sleep pattern.

**Definition and classification of sleep disorders**

Sleep problems in childhood are classified according to the International Classification of Sleep Disorders: Diagnostic and Coding Manual (ICSD, 1990, American Sleep Disorder Association) into the following categories:

1. Dyssomnia

Dyssomnia comprises disorders that cause either insomnia (most common) or excessive sleepiness (rare). Childhood insomnia is most often seen in *limit-setting sleep disorder* (in which inadequate enforcement of bedtimes by the parent results in the child stalling or refusing to go to bed at an appropriate time) and in *sleep-onset association disorder* (in which the onset of sleep in the child is impaired by the absence of a certain object or set of circumstances). The first conditions is often called "sleep refusal" or "bedtime struggles" (a vivid expression) and has been reported in 5-10% of a normal population of preschool children (44). The second condition is often called "frequent night-waking", when the child needs parental intervention of some form several times every night to go back to sleep, and is seen in 15-20% of all children between 6 months and 3 years of age (44,98). Severity is systematically classified along an axis in the ICSD, 1990, but this classification and coding system has not been widely used in studies.
The ICSD criteria of severe sleep refusal (limit-setting sleep disorder) require five or more episodes per night of stalling, calling out or leaving the bedroom. The ICSD criteria of severe night waking problems (sleep-onset association disorder) require a prolonged sleep latency and over three nightly wakings, or two or three wakings, each lasting over 10 minutes or one lasting over 15 minutes. The awakenings occur at least five nights per week. Corresponding detailed definitions for mild and moderate forms of the different sleep disturbances are also found in the ICSD.

Nocturnal eating syndrome and schedule disorders are other types of insomnia. Nocturnal eating (drinking) syndrome is characterized by recurrent awakenings, with the inability to return to sleep without eating or drinking, preferably breastmilk or formula. In schedule disorder the distribution of sleep in terms of nap frequency and length across a 24-hour period, the sleep-wake cycle, is altered in a way that comes into conflict with social expectations and habits. The child’s sleep occurs at an inappropriate time. Of course these definitions are cultural, as are the expectations of what is "normal" infant sleep. Breast-fed babies throughout the world wake at night and fall asleep at the breast, and in industrialized societies, in which the ICSD 1990 was written, we may have established inappropriate expectations from experience with bottle-fed infants.

Nevertheless, parents of children with the above-mentioned sleep patterns report themselves to be exhausted and in need of paediatric help. Paediatricians, therefore, must have a good knowledge of infant sleep and factors related to severe problems. Excessive sleepiness is seen in the rare disorder, narcolepsy, and more often in the obstructive sleep apnea syndrome.
2. Parasomnia

Parasomnia comprises disorders that intrude into or occur during sleep that are not, primarily disorders of the states of sleep and wakefulness per se. Included here are confusional arousals, sleepwalking, sleep terrors, sleep talking and sleep bruxism. Parasomnia is seen predominantly in children aged 3-12 years, where it encompasses 1-6 % of the population (7).

3. Medical/Psychiatric Sleep Disorders

Medical/Psychiatric Sleep Disorders comprise the medical and psychiatric disorders commonly associated with sleep disturbance. In acute and chronic illness (123), above all in diseases with pains or itching sensations, (38) sleep problems are common, but the most difficult and prolonged sleep problems are seen in mentally retarded children and children with autism, indicating a common factor of dysfunction of the central nervous system (16).

**Assessment of sleep**

Clinical assessment of sleep disorders in children includes developmentally appropriate history-taking of sleep-wake behaviour, including medical, psychiatric as well as family and psychosocial issues (8,42,84). Physical examination should be performed, with special attentiveness to signs of diseases accompanied by pains, breathing difficulties or itching sensations. Questionnaires and sleep logs or sleep diaries are valuable tools in the process of detailed history-taking concerning sleep-wake complaints (8,121).

Actigraphy was developed as a practical measure for long-term quantification of sleep/wake periodicity of movement, since both infants and children have a lower activity level in sleep than in wakefulness. A small motion sensor and
A digital recorder is worn on a wrist and later downloaded into a computerized analysis system. Advantages include noninvasiveness, ease of use and automated scoring. Accuracy for sleep-wake discrimination has been reported to be 85-93 % (111-112).

Audio and video recordings as well as polysomnography or other specialized laboratory tests are seldom used in infant studies. In clinical practice, selected complicated cases are referred to sleep clinics with extended laboratory devices (multichannel recordings, cardiorespiratory video systems, pulse oximetry) (24,58).

Results from studies comparing parental reports with objective sleep measures (109) show that parents are accurate reporters of sleep schedule measures such as sleep onset and sleep duration. They overestimate, however, the time that their infants spend in actual sleep and underestimate the number of their night wakings. According to that result, short sleep duration and many night wakings reported by parents of small infants should be regarded as a minimum rather than be suspected of being an exaggeration by exhausted care-takers.

**Epidemiology of sleep problems: prevalence, severity and persistence**

Sleep disturbances in infants and young children, such as night waking and bedtime struggles, are among the most common behaviour problems encountered in paediatric practice. Night waking occurs in 15 % to 35 % of children aged between 6 months and 5 years (17-18,21,59,69,76,94,98,102) and the prevalence of bedtime struggles varies from 6 % to 16 % in the first 2 years (18,76,94) according to previous studies.
These earlier studies show that approximately one preschool child out of four shows some kind of sleep problem, most often of short duration. A small group of children, however, approximately 5%, have more longlasting and complicated sleep disturbances that can have an influence both on the health of the child and on the function of the whole family.

Information on the long-term course of sleep problems in children (33,70-71), and particularly in preschool children, is sparse. Richman, in her survey of cross-sectional and longitudinal studies of disorders of initiating and maintaining sleep (100), concludes that although correlations for waking problems at different ages are reported low in schoolchildren (70), a considerable number of preschool children have persistent sleep problems. Especially during the first 2 to 3 years, the probability of continued waking is high.

Infants who are irritable, sleep little and cry frequently during the early weeks of life are more likely to be poor sleepers during the second year of life (20,23). One survey (60) found that just under half (44%) of those children waking regularly at 6 months were still waking at 1 year. Furthermore almost the same proportion (41%) of the night wakers at 1 year were still waking at 18 months, and from 18 months to 2 years just over half of the children (54%) continued waking. Conversely, 80% to 90% of nonwakers at these ages continued to sleep well. Continuity after 2 years was less marked: 71% of night wakers at 2 years of age slept well at 3 years of age, and 86% of wakers at 3 slept well at 4.5 years of age.

Another community survey (103) found more problems. Of those with bedtime problems at 3 years of age, 25% still had problems at 4 years of age. For night waking problems the figure was 36%. Persistence of the same problems in children 4 to 8 years of age was 40% and 17%, respectively.
There was a small core of persistent wakers, who Richman (100) speculates could be particularly susceptible to irregular sleep patterns because of neurophysiological or temperamental factors.

In another study (129) children with persistent sleep problems from 8 months to 3 years of age (41 %), were found to be more likely to have behaviour problems, especially temper tantrums and behaviour management problems, than were children without persistent sleep problems. Finally, 84 % of sleep-disturbed two-year-olds still had persistent sleep disturbances 3 years later in a study (67) that found that persistent sleep disturbances had a significant relationship with increased frequency of stress factors in the environment.

Different definitions of night waking and bedtime problems in different studies contribute to generating different prevalence, severity and persistency figures, but in summary, longlasting and complicated sleep problems seem to have a tendency to remain, especially from the first year of life to later childhood (1,21,67,92) and to be associated with behavioural problems (119,129) as well as extreme tiredness and stress in the family (1,13,51,53).

**Correlates to disturbed sleep**

Factors in the genetic, somatic, psychological and psychosocial areas correlate to disturbed sleep, as possible causative or maintaining mechanisms. Genetic vulnerability, perinatal and medical disorders, temperamental difficulty in the child, psychological (over-) responsiveness of the parents, parental insecurity and stress, psychosocial problems, and development of vicious circles (19-20,32,61,82,89) have been documented. The effects of insufficient and disturbed sleep in children include daytime behavioural problems. The children do not necessarily indicate their sleep loss by obvious daytime somnolence (119). On the contrary, a hyperactive
behaviour might be the only indicator of sleep deprivation in a child, with resolution of the behaviour problem following successful treatment of the sleep disturbance (46). The effect of chronically disturbed sleep on the parents is extreme tiredness (30,47,98,125,129), and several studies have shown that severe sleep problems in infants and toddlers (pre-school children) are associated not only with extreme tiredness and but also with psychiatric (73) and physical illness in the family, especially depression in the mother. The parents of children with severe sleep problems often seek help, and associations with marital problems (19-20,82,89) and even child abuse have been described (30,61,67). A heightened risk of later development of behavioural problems and school difficulties also exists for children with severe sleep problems (31-32,67,83).

**Treatment of sleep problems in childhood**

The methods suggested in the management of sleep difficulties range from total acceptance (117) (usually meaning taking the child to the parents’ bed) to ignoring all crying, with many variations in between. A number of treatment programs, based on behavioural techniques, have been described in the literature (73). These include establishing night-time routines and rituals, reorganizing settling and waking times, minimizing night-time attention by gradually withdrawing reinforcers, controlled crying by graduated extinction (progressive delay responding) or straight extinction ("crying it out"), and scheduled anticipatory waking where parents arouse and then resettle their child 15-60 minutes before expected spontaneous awakings.

Studies of these techniques show high rates of improvement (80-90 %) when used with support from a therapist (5,63,97,101,118). Sedatives are commonly used and may be helpful in the short-term, but have been shown to have long-term benefits only when used in combination with a behavioural
programme (51). The studies carried out to investigate the success of behavioural programmes rarely have follow-up periods longer than three months (47,56,73,79,81,126,128).

**Attention-deficit/hyperactivity disorder (ADHD) and sleep**

Sleep problems are frequently reported in children and adolescents with attention-deficit/hyperactivity disorder (ADHD) (14,25,66,88,104,124). Parents perceive children with ADHD to have greater sleep difficulty than normally developing children, and anecdotal reports of practising clinicians suggest that the sleep of children with ADHD is often reported by parents to have been disturbed from the very beginning of the child’s life, ever since infancy: "He has never slept through a whole night in his entire life."

Previously, based on clinical experience, sleep disturbance among children with ADHD was so widely presumed that it was included as one of the diagnostic criteria for the disorder (DSM-III; American Psychiatric Association, 1980). However, further research and re-examinations (14) led to the exclusion of the sleep disturbance criterion in the 1987 version of the DSM-III-manual. The present diagnostic criteria for ADHD (DSM-IV, 1994) are presented in Box 1.
Box 1. Diagnostic criteria for attention-deficit/hyperactivity disorder (DSM-IV, 1994)*

A. Either (1) or (2):

(1) six (or more) of the following symptoms of **inattention** have persisted for at least 6 months to a degree that is maladaptive and inconsistent with developmental level:

**Inattention**
(a) often fails to give close attention to details or makes careless mistakes in schoolwork, work, or other activities
(b) often has difficulty sustaining attention in tasks or play activities
(c) often does not seem to listen when spoken to directly
(d) often does not follow through on instructions and fails to finish schoolwork, chores, or duties in the workplace (not due to oppositional behaviour or failure to understand instructions)
(e) often has difficulty organizing tasks and activities
(f) often avoids, dislikes, or is reluctant to engage in tasks that require sustained mental effort (such as schoolwork or homework)
(g) often loses things necessary for tasks or activities (e.g., toys, school assignments, pencils, books, or tools)
(h) is often easily distracted by extraneous stimuli
(i) is often forgetful in daily activities

(2) six (or more) of the following symptoms of **hyperactivity-impulsivity** have persisted for at least 6 months to a degree that is maladaptive and inconsistent with developmental level:

**Hyperactivity**
(a) often fidgets with hands or feet or squirms in seat
(b) often leaves seat in classroom or in other situations in which remaining seated is expected
(c) often runs about or climbs excessively in situations in which it is inappropriate (in adolescents or adults, may be limited to subjective feelings of restlessness)
(d) often has difficulty playing or engaging in leisure activities quietly
(e) is often "on the go" or often acts as if "driven by a motor"
(f) often talks excessively

**Impulsivity**
(g) often blurts out answers before questions have been completed
(h) often has difficulty awaiting turn
(i) often interrupts or intrudes on others (e.g., butts into conversations or games)

A. Some hyperactive-impulsive or inattentive symptoms that caused impairment were present before age 7 years.

B. Some impairment from the symptoms is present in two or more settings (e.g., at school [or work] and at home).
C. There must be clear evidence of clinically significant impairment in social, academic, or occupational functioning.

D. The symptoms do not occur exclusively during the course of a Pervasive Developmental Disorder, Schizophrenia, or other Psychotic Disorder and are not better accounted for by another mental disorder (e.g., Mood Disorder, Anxiety Disorder, Dissociative Disorder, or a Personality Disorder).


A detailed and systematic review of empirical research published since 1970 on sleep disturbances in children with ADHD, encompassing sixteen relevant studies, was published in 1998 by Corkum, Tannock and Moldofsky (35). They found that although subjective accounts of sleep disturbances in ADHD were prevalent (based on sleep diaries, questionnaires and interviews), objective verification of these disturbances (by polysomnographs, actigraphy or video) was less robust (13). In fact, the only consistent objective findings were that children with ADHD displayed more movements during sleep but did not differ from normal controls in total sleep time (39,78,121). Further studies by the same authors have confirmed this picture (34) and pointed to a complex relationship between sleep problems and ADHD. In a recent study (80) the conclusion was drawn that although subjective sleep difficulties are common in ADHD youths, they are frequently accounted for by comorbidity and pharmacotherapy.

However, when convincing evidence exists of sleeplessness in an overactive child, attempts should be made to increase or improve the sleep because, in some cases, the ADHD features may be the result of poor sleep (37,121). The effects of sleep loss in children include tiredness, difficulties with focused attention, low threshold to express negative affect (irritability and easy frustration), and difficulty modulating impulses and emotions (37). In some cases these symptoms may resemble ADHD. This has been reported in children with obstructive sleep apnoea, with improvement of behaviour following treatment (53).
ADHD-associated sleep disturbances may be improved by medication with clonidine (93,127). Improvements can also be achieved by administration of stimulant drugs late in the day (29,93,121,127) even if insomnia secondary to stimulant medication is a well-known adverse effect in some children (15,35,120). Prospective, randomized, double-blind, placebo-controlled, long-term studies of stimulant treatment for childhood attention-deficit problems have, however, showed that for this patient group in general, early awakenings and disturbed sleep tend to occur less frequently with stimulant treatment than at baseline, i.e. without treatment (49).

In their review of the literature (35), Corkum, Tannock and Moldofsky conclude that the exact nature of the sleep problems in children with ADHD remains to be determined. Many relevant issues need new and better methodological approaches. One interesting field is Periodic Limb Movements in Sleep (PLMS) in children with ADHD, which has been recently studied by Picchietti et al (90-91). PLMS are repetitive flexions of the toes, feet, legs, thighs and/or the arms which recur periodically during sleep. Sleep disruptions often accompany these movements, which in studies using polysomnography and electromyography have been shown to be more prevalent in children with ADHD than in control children.

Studies using retrospective data on sleep patterns in infancy among children with ADHD are scarce. Trommer et al (124) compared 48 schoolchildren with ADD (with and without hyperactivity) with 30 controls using retrospective parental questionnaires which covered the ages 0 to 12 months and 1 to 4 years. The ADD children were reported to have had more frequent arousals than the controls had both in the first year and from 1 to 4 years. However, there is an obvious risk of selective memory bias in such studies.
Other prospective studies than the one included in this thesis on children with severe sleep problems in infancy, concerning the risk of later development of inattention and hyperactivity-impulsivity, have to our knowledge not yet been performed.
Aims of the studies

The aims of this 5-year prospective study were:

1. to provide epidemiological data on infant sleep-wake characteristics and the prevalence and severity of sleep problems in a general population, as perceived by parents (Study I),

2. to explore medical, psychological and psychosocial child and family characteristics associated with severe infant sleep problems (Studies I och II),

3. to investigate the changes in sleep in a group of severely sleep-disturbed infants after a specific treatment programme combining behavioural technique and interdisciplinary family work (Study III),

4. to prospectively follow and compare a group of children identified with severe sleep problems in infancy with a control group regarding the development of symptoms of attention-deficit/hyperactivity disorder (ADHD) (Study IV) and regarding further sleep pattern (Studies III and V),

5. to explore factors in infancy which are associated with subsequent development of ADHD in later childhood (Study IV).
METHODS

Design and subject selection

Study I

A total community sample of 2,518 infants aged between 6 and 18 months were approached using a parental questionnaire. The response rate was 83%. Data from the collection procedure point to a non-selective drop-out, not reflecting presence or absence of sleep problems. Severe sleep problems as defined by the ICSD were found in 129 of the children (6.2%) who were studied in detail, with the rest of the population as controls.

Studies II-IV

Those 6 to 12 month-old children in the community sample who fulfilled specific inclusion criteria for severe and chronic sleep problems (N = 27) created the case group.

Inclusion criteria were:

- the child should be reported by parents to have a prolonged sleep latency and over three nightly wakings occurring at least five nights per week. This pattern of disrupted sleep should have prevailed for a period of at least 6 months.

- infant age between 6 and 12 months at inclusion in the study. (The reason for restriction of the study to this age group was limited resources and the ambition to investigate all children with severe sleep problems in a specific population with home visits by the same paediatrician).
- full-term pregnancy with a birth weight of greater than 2 500 grams and gestational age of at least 37 weeks.

- the parents should report the need of help. (This criteria turned out to be unnecessary. All parents of children with the above-mentioned sleep problems reported themselves to be in need of help. This was true even for the non-European parents in this group, who co-slept with their breastfed child according to their cultural norms. They all reported much need for help with their child’s sleep within the context of the Swedish industrialized society).

- the parents should be able to communicate in Swedish or English.

The exclusion criterion was:

- signs of significant organic causes of the sleep problem identified by hospital examinations and laboratory tests.

Among the 28 families with infants fulfilling the set inclusion criteria, (i.e. 2 % of the sample of 1 259 children aged between 6 and 12 months), one family was not able to participate because they moved to another part of the country. The other 27 families agreed to participate in the study. Sixteen of the children were boys, eleven girls. None of them fulfilled the exclusion criteria. All were basically healthy and went further in the study.

A control group was matched with regard to age and sex. The infant of a particular sex who fell next in age sequence to the case infant in the Child Health Center (CHC) register was chosen as a control. A further criterion was that no sleep problems were reported on the questionnaire. Of 33 families contacted, 27 agreed to participate.
**Study V**

In addition to the original control group, an extended control group was created from the questionnaire – study I, consisting of four control children for each case child. The controls were matched for age and sex, had no reported sleep problems in the questionnaire at 6 to 12 months and were still living in Sweden five years later.

**Procedure and measures**

**Study 1**

In January 1996 data were collected in a parent population study in the urban district of Uppsala (the fourth largest municipality in Sweden). A questionnaire, constructed especially for this study, was sent to all parents with an infant born during the period from July 1994 to July 1995.

**Questionnaire on sleep and background factors**

The instrument covered the following areas: social and demographic background, pregnancy and birth, sleep-wake characteristics, parental behaviour, and feeding problems.

Most items in the questionnaire had a 5-step response scale with verbal definitions of each scale step. A few items had response categories with yes/no answers. The items were constructed in close accordance with the ICSD classification in addition to subjective parental rating scales of the severity of the problem. No validation process was possible for these scales, since the explicit aim of the questions was to measure early sleep problems as perceived by parents.
Study II, III, V

A home visit was scheduled when parents had received information about the study and agreed to participate in the research project. The author, who is a clinically active paediatrician, interviewed the parents about the infant’s sleep characteristics and family members’ previous and present state of health. The infant’s interaction with the parents and normal activity were observed, and the development of the child was assessed. A thorough somatic examination of the child was also performed. Information about demographic variables, perceptions of parenting and infant temperament, life events and social support was collected in a questionnaire that the mothers filled in after the home visit. The following week the parents kept a special Sleep Diary of the sleep of their child. This diary was brought to the special CHC center where the parents of the children in the case group received advice, counselling and support in the form of a behavioural training programme, in order to overcome the sleep problems of their child. The sleep programme was carried out as detailed later. The paediatrician was of course aware of the group status of the families visited. In order to reduce the possible influence of this, the assessments of the psychosocial problems were compared with data from the CHC and the medical records of all the children. This was done by two independent assistants who were blind to infant group status.

One month after inclusion in the study the parents in the problem group were sent another 7-day sleep diary and a general questionnaire about their opinion of the help received. At one year, two and a half years and five years, respectively, from inclusion, contact was made by mail, and a questionnaire and sleep diary were filled in by the parents of both the problem group and the control group. For ethical reasons the study design did not include any placebo intervention group (i.e. untreated children in the problem group), as
studies of behavioural programmes have shown high rates of improvement (80-90 %) (5,63,97,101,118).

**Questionnaires on sleep, family and infant characteristics, and sleep diaries**

The home visit covered questions about sleeping characteristics and the state of health of the family and comprised open-ended questions, scored on yes/no, 3-step or 5-step response scales. Sleep characteristics were covered in five questions that allowed a classification of the child’s sleep pattern/problem according to the definitions of ICSD, 1990, in a 24-hour recall of the infant’s sleep, and questions about the circumstances surrounding getting the child to sleep. The following week the parents filled in a special Sleep Diary of the sleep of their child in which the amount of night and daytime sleep was recorded. In addition information was recorded about clock time into bed, sleep latency, settling time, arousals (time and length), morning wake time, in bed awake – and out of bed time, and day time naps (time and length). All data were measured in minutes. Parental and sibling sleep problems during infancy were also probed. In order to assess the infant’s general development, the procedure used at the CHC was employed (National Board of Health and Welfare, 1991), including an assessment of gross and fine motor skills, as well as language, social, cognitive, and neurological development. Development was rated on 5-step scales ranging from very late for age to well above chronological age level. The health status of the family was addressed by two questions about medical pregnancy and delivery complications, and by four concerning the family’s contact with medical care and health status during the infant’s lifetime. Answers to questions about health were supplemented with information from medical records.
Assessment of the psychosocial situation of the family was based on self-disclosure during the medical interview (e.g. reports of unemployment, financial restraints, conflicts between the infant’s parents, conflicts with the infant’s grandparents, family health problems, substance abuse). Psychosocial problems were rated by the author as none, minor or major. In order to reduce the possible influence of the authors’ knowledge of the group status of the child, the assessments of psychosocial problems were compared with data from the CHC and medical records. This was done by two independent assistants who were blind to infant group status. The findings showed an interrater correlation of $r = 0.95$ when one assistant’s ratings of the listed observations of the paediatrician (author) was compared with the other assistant’s ratings of data from the CHC and medical records. The interrater correlation between the author’s ratings and the assistant’s ratings was $r = 0.98$.

Information about family demographic variables (ethnicity, age and education, employment, housing and financial situation, and number of children) was gathered by a questionnaire mothers completed after the home visit. Maternal perceptions of parenting were obtained through semi-structured questions scored by mothers on 5-step response scales. The inventory, a revised Swedish version of R. Abidin’s Parenting Stress Index (PSI; Abidin, 1990) (2,74,130), contained 34 items in 5 subscales that focused on parental feelings of competence/incompetence, restriction by parental role, social isolation, spouse relationship problems, and health problems.

To measure infant temperament, the Baby Behaviour Questionnaire (BBQ) and the Toddler Behaviour Questionnaire (TBQ) (54-55) were used. In the methodological work with the TBQ, a construct corresponding to difficultness has been developed, consisting of low manageability as the most important
characteristic. This construct, based on 5-step response scales, was used in the present study.

The occurrence of infants’ problematic behaviour during the previous month was reported by mothers in the home interview. Questions in the interview were related to sleeping habits, thumb or dummy sucking, screaming and whining, shyness, activity, restlessness and lack of concentration, temper tantrums and the mothers’ perception of the child as troublesome, difficult to handle. Maternal ratings on 2-point response scales (yes, no) were used.

**Treatment programme**

The methods used were based on the following premise which was explained to parents: "During the night all children have some spells of light sleep during which they easily wake. The problem is not that the child wakes up but that it cannot slip back to sleep without the help of a parent rocking or feeding it. The way the baby is settled to sleep in the evening will be its preferred way of returning to sleep after natural wakings. In order to have a child that returns to sleep by itself, you have to accustom it to settle on its own".

The programme included:

*Controlled crying*

Controlled crying, or more positively called controlled comforting, implies that the parent spends a few minutes settling the child, then leaves it for gradually increasing lengths of time with brief and warm, but firm, reassurance and resettling in between, yet avoiding taking the child out of its cot. No feeding is given, and the child should be drowsy but still awake when put into bed. The intervals between reassurance increase from initially one minute up to five minutes at most. This procedure was used (if needed) every
time the baby was settled by day or night, and whenever it woke and cried at night. Consistency was emphasized. A nurse and a child psychologist supported the parents by daily telephone calls for 1-2 weeks and then tried to help them to develop confidence in the programme and to manage on their own.

**Day and night routines**

Overtired babies do not sleep well at night. Two daytime sleeps of about 1-1½ hours were encouraged, together with a regular pattern of feeds, play times (with plenty of parental involvement) and sleep. The precise times could be varied somewhat to fit in with each family situation, but it was important to ensure that feeds and sleep were separated.

General help and information were given as appropriate, e.g. about nutrition, feeding practices and play techniques. The great range of normal behaviour and development was discussed with the parents.

Parents were encouraged to be agreed about the programme management and to find ways to get support and time out for themselves in order to maintain the energy needed to care for a baby or toddler. Problems such as relationship stresses, social problems, depressive moods and parental health problems were addressed if appropriate.

**Medication**

Five children with deeply depressed and exhausted parents were given sedatives (alimemazine) for two weeks at the beginning of the programme, in combination with the behavioural programme, in order for the parents to be able to sleep directly and thereby regain some mental strength.
Study IV

A screening procedure for ADHD was carried out 4.5 to 5 years after inclusion in the study, when the children had reached the age of 5.5 years. In case of screen positivity, further assessment methods were used to diagnose ADHD.

Screening instrument for ADHD

The screening procedures were identical for all children in the study and comprised the following:

(i) a Parent Psychomotor Questionnaire (PPQ) containing four questions pertaining to the child’s psychomotor development for completion by parents and rated on a scale of normal/abnormal (95).

(ii) a previously validated Preschool Questionnaire (PSQ) comprising six items pertaining to the child’s attentional, motor, language and perceptual-conceptual capacity for completion by the preschool teacher and rated on a scale of normal/abnormal (96).

(iii) a motor examination, performed by a nurse who had received special training. The examination was carried out at the CHC, according to a standardized screening of motor abilities that comprised six items rated on a scale of normal/abnormal (48).

The screening procedures were in accordance with the battery recommended by the Swedish National Board of Health and Welfare for screening for minor neurodevelopmental/psychiatric disorders in preschool children at the CHCs (72,96).
The criteria for screen positivity were: (a) one or more abnormal scores on either the PPQ or the PSQ in combination with one or two abnormalities on the motor examination or (b) three abnormalities on the motor examination. Children not fulfilling these criteria in the screening procedure were considered screen-negative. The criteria for screen-positivity were chosen so as to identify children with ADHD as well as those with other neurodevelopmental and neuropsychiatric problems (72).

The screening methods used in this study have been psychometrically examined in previous Swedish studies (45,48,50,96) and have been found to have good inter-rater and test-retest reliability.

**Assessment methods**

The assessment methods used for diagnosis comprised a detailed history, psychiatric and neurodevelopmental examination, neuropsychological assessment and speech/language evaluation.

The first assessment was performed by the local multidisciplinary district CHC teams including the local psychologists, speech therapists, physiotherapists and paediatricians outside the original sleep study organization. Those teams were thus unaware of the group status of the children examined. The psychological examination included an assessment of development with Griffiths’ Developmental Scale II (52) in addition to a detailed history and a clinical neuropsychological assessment of symptoms of inattention, hyperactivity, impulsivity and behavioural problems. The speech therapists used clinically accepted assessment methods of language comprehension, phonological and grammatical problems, and the physiotherapists administered the Scandinavian motor-perceptual scale MPU
The local paediatrician performed a paediatric physical examination of the child with the focus on neurological functioning.

The second examining paediatrician with special training in child neuropsychiatry (author M.T.) was unaware of the results of the previous assessment performed by the local team members while performing the second step in the assessment procedure, a home visit.

A medical, developmental and behavioural history was taken at a home interview with the parents, using a standardized interview schedule. The criteria for ADHD according to the DSM-IV were checked during this interview, and each criterion was rated as 'definitely met', 'possibly met' or 'not met'. Inter-rater reliability of results obtained at this type of ADHD interview is excellent (72).

Routine paediatric psychical examination and a brief neurodevelopmental examination in accordance with the method outlined by Gillberg et al. (48) were also performed at the home visit. Inter-rater reliability for this standardised assessment is good to excellent with values of Pearson r ranging from 0.68 to 1.00 for individual items (50).

**Diagnosis**

A comprehensive diagnosis was made in each case after all neuropsychiatric assessments had been completed on the basis of the available information.

The diagnosis ADHD was set in children meeting the criteria for this diagnosis of the DSM-IV (Box 1).
Statistics

For analysis of the data, the statistical program SAS (113) was used in all studies. Group comparisons for dichotomous variables were executed with Chi-square analyses or Fisher’s exact test (two-tailed) when an expected cell value was less than 5. For variables with 3-, 5- or 7-step response scales, comparisons were made with t-tests. In group comparisons, the more conservative alpha level of 0.01 was used because of the large number of tests.

RESULTS

Prevalence and classification of sleep problems (Study I)

Sleep problems: Descriptive data

Previous sleep problems, irrespective of duration or type, were reported for 48.3 % of the total child population. At the time of inquiry, 40.8 % of the children were reported to have episodes of evening sleep refusal, including minor and transient problems. The sleep refusal was considered a severe or very severe problem by 3.8 % of the parents.

With respect to night-waking, 62.3 % of the children had current problems, irrespective of severity and duration. The night-wakings were considered a severe or very severe problem by 5.6 % of the families.

Table 2 gives descriptive data for sleep refusal and night-waking problems measured with parental rating scales. Table 1 gives descriptive data classified according to the operational definitions of ICSD. The frequency of severe sleep refusal and severe night-waking problems in the total child population varied between 3.8 % and 6.2 % in these measurements, with the parents
being slightly more tolerant in their judgement and opinions than the ICSD scales.

**Table 1.** Percentages of infants that fulfilled the ICSD criteria of severe sleep refusal (limit-setting sleep disorder\(^a\)) and severe nightwaking problems (sleep-onset association disorder\(^b\)).

<table>
<thead>
<tr>
<th></th>
<th>Ages 6-12 months, N = 1048</th>
<th>Ages 13-20 months, N = 1018</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Tot(^c)</td>
<td>Chronic(^d)</td>
</tr>
<tr>
<td>Severe sleep refusal</td>
<td>4.9</td>
<td>3.2</td>
</tr>
<tr>
<td>Severe night waking</td>
<td>8.6</td>
<td>2.7</td>
</tr>
</tbody>
</table>

Note: More than one problem can be reported for a child.

a) The ICSD criteria of severe limit-setting sleep disorder require five or more episodes per night of stalling, calling out or leaving the bedroom.

b) The ICSD criteria of severe sleep-onset association disorder require a prolonged sleep latency and over three nightly wakings, or two or three wakings each lasting over 10 minutes or one lasting over 15 minutes. The awakenings occur at least five nights per week.

c) Percentages of children for which problems were reported.

d) Proportion of children with chronic problems.

Chronic sleep refusal (limit-setting sleep disorder) requires a duration of 3 months or longer. Chronic night-waking problems (sleep-onset association disorder) require a duration of 6 months or longer.
Table 2. Percentages of infants for which current sleep problems were reported by parents. 

N = 2066.

<table>
<thead>
<tr>
<th>Sleep refusal</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>no problem</td>
<td>59.2</td>
</tr>
<tr>
<td>minor problem</td>
<td>24.6</td>
</tr>
<tr>
<td>moderate problem</td>
<td>12.4</td>
</tr>
<tr>
<td>severe problem</td>
<td>2.6</td>
</tr>
<tr>
<td>very severe problem</td>
<td>1.2</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Night waking</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>no problem</td>
<td>37.7</td>
</tr>
<tr>
<td>minor problem</td>
<td>32.3</td>
</tr>
<tr>
<td>moderate problem</td>
<td>24.4</td>
</tr>
<tr>
<td>severe problem</td>
<td>3.4</td>
</tr>
<tr>
<td>very severe problem</td>
<td>2.2</td>
</tr>
</tbody>
</table>
Family and infant characteristics associated with severe infant sleep problems (studies I and II)

Comparison of severe sleep problem group \( (N = 129) \) with a control group \( (N = 1937) \) (Study I).

Sex, siblings, age distribution, pregnancy and delivery

There were more boys in the problem group (60 %) compared with controls (51 %) \( (X^2 (1, N=2066) = 4.01, p < 0.05) \). No significant difference was found for the presence of siblings. The age distribution of children in the problem group was uneven, with an overrepresentation of children aged 8, 9 and 10 months (42%) compared with controls (26%) \( (X^2 (1, N=2066) = 15.09, p < 0.001) \). There were no significant group differences in prematurity or birthweight.

Family sleep problems

No significant group differences were found concerning previous childhood sleep problems within the family (mother, father and siblings analyzed separately). In the problem group 12 % of the mothers, 10 % of the fathers and 27 % of the siblings were reported to have had sleep problems in the first years of life. Corresponding figures among controls were 10.8 and 23 %, respectively.

Parental behaviour

Significant group differences were found concerning kinds of parental intervention when the child refused to go to bed in the evening and when the child needed to go back to sleep after waking up in the night. In the problem
group the parents to a significantly lesser degree left the child alone to settle by itself in the evening (8 %) compared with controls (16 %). The parents in the problem group more often sang to their children, pulled the child in a baby carriage, or preferably breastfed the child or offered it a bottle to settle with in cases of sleep refusal in the evening, as shown in Table 3.

Table 3. Percentages of families who used different kinds of parental intervention to get the child to sleep in situations when this was problematic.

<table>
<thead>
<tr>
<th></th>
<th>Problem group</th>
<th>Control group</th>
<th>X²</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N = 129</td>
<td>N = 1 937</td>
<td></td>
</tr>
<tr>
<td>In the evening</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Leaves the child to settle alone</td>
<td>8</td>
<td>16</td>
<td>6.32 *</td>
</tr>
<tr>
<td>Sings for the child</td>
<td>53</td>
<td>40</td>
<td>7.70 **</td>
</tr>
<tr>
<td>Pulls the child in a baby carriage</td>
<td>8</td>
<td>3</td>
<td>9.92 **</td>
</tr>
<tr>
<td>Breastfeeds the child / gives a bottle</td>
<td>63</td>
<td>38</td>
<td>31.13 ***</td>
</tr>
<tr>
<td>In the night</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Puts their hand on the child</td>
<td>39</td>
<td>26</td>
<td>10.90 ***</td>
</tr>
<tr>
<td>Rocks/holds the child in their arms</td>
<td>36</td>
<td>23</td>
<td>11.59 ***</td>
</tr>
<tr>
<td>Sings for the child</td>
<td>23</td>
<td>12</td>
<td>14.89 ***</td>
</tr>
<tr>
<td>Takes the child to the parents’ bed</td>
<td>65</td>
<td>50</td>
<td>10.72 **</td>
</tr>
<tr>
<td>Breastfeeds the child / gives a bottle</td>
<td>70</td>
<td>48</td>
<td>36.04 ***</td>
</tr>
</tbody>
</table>

Note: Chi-square statistics are given from comparisons of the problem group with the control group with df = 1 in all tests.

* p < 0.02
** p < 0.01
*** p < 0.001
During the night the parents in the problem group also intervened more than the control group parents in situations when the child woke up and needed to go back to sleep. Table 3 shows significant group differences in the following interventions: putting parents’ hand on the child, rocking/holding the child in parents’ arms, singing for the child, taking the child to the parents’ bed and, above all, breastfeeding the child or giving the child a bottle.

**Feeding problems**

Feeding problems were defined here as difficulties in eating solids, with refusal and preference for breast-milk or formula. Comparisons of the group with severe sleep problems and the control group revealed significant differences. Feeding problems were reported for 33 % of the problem group compared with 15 % of the controls \( (X^2 (1, N = 2066) = 29.33, p < 0.001) \), and 18 % of the problem group parents reported themselves as being in current need of help with the feeding problem of their child, compared with 6 % of the control parents \( (X^2 (1, N = 2066) = 25.03, p < 0.001) \).

**Health**

Comparisons of the presence of eczema, allergy and nutritional intolerance between the problem group and the control group showed no significant differences. Neither could any differences be found concerning frequency of occurrence of diarrhoea, constipation, colds, otitis or fever. However, more worries and anxiety concerning infant health were reported in the problem group than in the control group. Although there was no difference in the frequency of colds or ear infections between the groups, the parents of the affected children in the problem group were significantly more worried over the diseases than were corresponding control parents: colds \( t (df=125) = 3.37, p < 0.001; \) otitis \( t (df = 551) = 3.69, p < 0.001. \)
In this study severe sleep problems were found to be correlated with parental worries and anxiety concerning infant health (although the children were reported as being as healthy as the controls), infant feeding problems and intensive parental interventional behaviour (especially feeding) during the evening and night. A common factor of insecurity in the parental role is suggested.

**Comparison of severe and chronic infant sleep problem group (N = 27) with a control group (N = 27) (Study II)**

**Sleeping characteristics**

More case infants were breastfed (59 %) than were controls (15 %) ($X^2 (1, N = 54) = 11.44, p < 0.001$). Most case infants (78 %) were fed immediately before going to sleep, whereas this was true for only 37 % of the controls ($X^2 (1, N = 54) = 9.16, p < 0.01$).

Feeding was used by the parents of case children as a method of getting the child to sleep. Consequently most case infants (18 out of 27, 67 %) fell asleep in the arms of their mother, whereas most of the control children fell asleep alone in their own beds (16 out of 27, 59 %). These differences, however, were not significant.

During the night most of the case infants (56 %) slept in their parents’ bed (co-sleeping), whereas this was very uncommon among controls. Only one of the control children slept in the parents’ bed; the remaining control children, 26 out of 27 (96 %), slept in their own bed ($X^2 (1, N = 54) = 17.41, p < 0.001$). Frequent night meals (5 meals or more) were given to 44 % of the case infants. This pattern was not seen in any of the control children. The majority of the controls (63 %) was not fed at all during the night.
Evaluation of sleep data (except that polysomnographic monitoring was not used in this study) showed that 22 of 27 children in the problem group had a sleep pattern consistent with the diagnosis, nocturnal eating (drinking) syndrome. Four children in the problem group fulfilled criteria of sleep-onset association disorder, and one child exhibited a sleep pattern characteristic of schedule disorder.

Significant group differences were found concerning the childrens’ sleeping time during the night based on a 7-day parental report in the form of a detailed Sleep Diary (Table 4).

The case children’s mean sleeping time during the night was one and a half hours less than that of the controls, and the case infant group woke up significantly more often. A tendency towards group difference was also seen concerning the children’s sleeping time during the day, with the case children taking shorter day time naps. Over a 24-hour period the mean sleeping time for the case children group was two hours less than that of the control group.

**Table 4.** Results from analyses of variance with means and standard deviations for sleeping characteristics.

<table>
<thead>
<tr>
<th></th>
<th>Cases (N = 27)</th>
<th>Controls (N = 27)</th>
<th>p value*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Night-time sleep (in minutes)</td>
<td>557.7</td>
<td>78.78</td>
<td>647.6</td>
</tr>
<tr>
<td>Number of night wakings</td>
<td>6.0</td>
<td>3.01</td>
<td>0.7</td>
</tr>
<tr>
<td>Night time awake</td>
<td>71.8</td>
<td>37.39</td>
<td>8.5</td>
</tr>
<tr>
<td>(length of arousal time at night in minutes)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Daytime sleep (in minutes)</td>
<td>107.6</td>
<td>61.11</td>
<td>138.1</td>
</tr>
<tr>
<td>Sleep latency at bedtime</td>
<td>30.0</td>
<td>32.67</td>
<td>10.6</td>
</tr>
<tr>
<td>Total sleep during a 24-hour period</td>
<td>665.3</td>
<td>91.54</td>
<td>785.7</td>
</tr>
</tbody>
</table>

* p value from t-tests
The clock time into bed did not differ between the case and the control group, but the sleep latency period was longer and, consequently, the settling time later in the case group compared with the controls. No significant group difference was found concerning morning wake time.

Previous sleeping problems in the family were more common among case families (67%) than among controls (7%) \((X^2 (1, N = 54) = 20.33, p < 0.001)\).

**Demographic characteristics**

All mothers, except three in the case group and one in the control group, lived with the infant’s father. A significant difference was found with respect to parents’ ethnic origin \((X^2 (1, N = 54) = 7.48, p < 0.01)\); a higher proportion of case infants had a non-Swedish parent. Even more uneven was the proportion of non-European parents in the groups; 37% of the case children had one (7%) or both (30%) parents who originated from a non-European country (Africa, South-America, Asia), whereas this was true for only 4% of the controls \((X^2 (1, N = 54) = 9.24, p < 0.01)\).

During the same time as this study was made, data were presented from the Swedish Central Bureau of Statistics (SCB), in which Uppsala, where our study was performed, was divided into areas of high, medium, and low-income structure, based on family income. The majority of the case children lived in low income areas, and a significant group difference was found concerning family income, with financial problems reported in 48% of the case families compared to 11% of the controls \((X^2 (1, N = 54) = 8.88, p < 0.01)\).
p <0.01). Regarding housing conditions, the majority, 81 %, of the children with sleep problems lived in blocks of flats as did the majority of the controls, 59 %. However, 37 % of the control children lived in a detached house in residential districts compared with 11 % of the cases. This difference in housing conditions was, however, not significant.

No significant differences between cases and controls were obtained for the other demographic characteristics.

**Health characteristics**

A significant group difference was found concerning the psychosocial situation in the family; case families had more psychosocial problems than the control group. Comparisons of other variables related to somatic health in the family and of infant development revealed no significant findings. Chi-square analyses showed no significant differences in pregnancy and delivery complications.

The parents of the case children reported symptoms related to chronic loss of sleep with extreme tiredness and lack of energy (81 %). Feelings of hopelessness, restlessness, inability to relax and irritation were common. Many parents also reported sadness and crying and a sense of being close to mental breakdown. Somatic and psychosomatic complaints were very frequent (59 %) among parents of case children, such as headaches, pains in shoulders and back or "the whole body", vertigo and dizziness, heart-palpitation, fainting fits and loss of weight. Sexual problems were common (33 %).

The parents of case children suffered from exhaustion, both physically and mentally, and 11 case mothers (41 %) fulfilled the ICD-10-criteria for a severe depressive episode. None of them had sought psychiatric help. Fifty-
two per cent of the case mothers reported feelings of guilt in relation to their child, compared with 11 % of the controls ($X^2$ (1, N = 54) = 10.39, p <0.01). The pattern of interaction with the child was affected, and 78 % of the case mothers reported feeling that the locus of control of the situation and interaction was in their child. This feeling of being controlled by the child was very uncommon in the control group, 11 % ($X^2$ (1, N = 54) = 24.30, p <0.001).

Although depressed and in expressed need of help, only 20 of the 27 case parents (74 %) had asked for help at the Child Health Center. In the parent questionnaire population study, the corresponding figure was 93 % among parents who had experienced any sleep problem, including minor and transient in their children. The reasons why the parents of the children with the most serious sleep problems did not seek help were reported by them to be difficulties with language, lack of energy and a sense of guilt that the sleep problem of their child was their own fault.

**Perceptions of parenting**

Maternal perceptions of parenting differed significantly between groups. Mothers of case infants had less positive perceptions of parenting and experienced more parental stress, in particular feelings of incompetence and restriction by the parental role, as well as perceived health problems.

**Infant difficultness and problematic behaviours**

Comparisons between case and control infants regarding difficultness and problematic behaviours showed a significant group difference for temperamental difficultness. Regarding problematic behaviours, a significant group effect was found; cases had more problems than the control group. In
the assessment of the development of the children the paediatrician also rated their activity level. More case infants were rated as very active (30 %) compared with controls (0 %) \( (X^2 (1, N = 54) = 9.39, p < 0.01) \).

Independent ratings by the mothers revealed the same group difference; more case infants were rated as very active (52 %) than were controls (0 %) \( (X^2 (1, N = 54) = 18.90, p < 0.001) \).

Infants with severe and chronic sleep problems thus showed normal health and general development, and had a normal growth pattern. Early severe sleep problems were, however, found to be associated with a higher incidence of breast-feeding, frequent night meals as a method of putting the infant back to sleep, co-sleeping, and previous sleeping problems in the family. They were also associated with non-European origin, living in a low income area and with psychosocial problems in the family, including financial problems, exhaustion and depression in the mother, less positive perceptions of parenting and more parental stress. There were also feelings of incompetence and restriction by the parental role, as well as perceived health problems. An association with infant difficulty, high activity and problematic behaviours was found.

**Short-term and long-term outcome of an interdisciplinary sleep treatment programme (Study III and V)**

Three sets of parents found that they could not agree with the premise on which the programme was based, and therefore chose to leave the programme without even trying it. Their children were reported to have unchanged severe sleep problems one month later.
At the evaluation one month after the treatment programme had been initiated in the 24 motivated families, significant changes had taken place. The average number of times the babies woke up had decreased from 6.0 to 1.8 times per night and night-time sleep had increased by an average of 67 minutes (Table 5). The difference in total sleep during a 24-hour period between the problem group and the control group had been reduced by 55 %, and the difference in night-time sleep between the two groups had been reduced by 75 %. Ninety-two percent of the parents reported their children to have a good sleep after treatment and were satisfied with advice and support. The sleep pattern remained unchanged for two of the 24 problem children treated, and no child had exaggerated sleep problems after treatment.

Table 5. Results from analyses of variance with means and standard deviations for sleeping characteristics in the problem group, before and after treatment. N = 24.

<table>
<thead>
<tr>
<th></th>
<th>Before treatment</th>
<th>After treatment</th>
<th>p-value*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M</td>
<td>Sd</td>
<td>M</td>
</tr>
<tr>
<td>Night-time sleep (in minutes)</td>
<td>557.6</td>
<td>82.39</td>
<td>625.5</td>
</tr>
<tr>
<td>Number of night wakings</td>
<td>6.0</td>
<td>3.17</td>
<td>1.8</td>
</tr>
<tr>
<td>Night time awake (length of arousal time at night in minutes)</td>
<td>76.2</td>
<td>37.13</td>
<td>22.6</td>
</tr>
<tr>
<td>Daytime sleep (in minutes)</td>
<td>102.5</td>
<td>60.66</td>
<td>106.1</td>
</tr>
<tr>
<td>Total sleep during a 24-hour period</td>
<td>660.1</td>
<td>95.92</td>
<td>731.5</td>
</tr>
</tbody>
</table>

*p value from t-tests
One year after the accomplishment of the sleep programme, the previous problem group still had a good sleep. No significant differences were found between the sleep duration of the formerly sleep-disturbed children and their controls.

The follow-up after two and a half years showed no significant group differences. The former problem group still slept as well as the controls.

Thus the changes were stable over time. The families in the case group also managed to maintain the achieved changes in infant sleep behaviour on their own; continuous therapist support was not necessary. This was true even for formerly depressed and psychosocially burdened parents.

Five years after the initial contact the former problem group still slept as well as the controls according to the parental ratings of sleeping characteristics (Table 6). Concerning sleeping behaviour there was a tendency for the case group to be judged by their parents to move more and be more restless during sleep than were the controls. For the children in the problem group that at the age of 5.5 years fulfilled the criteria for the diagnosis of attention-deficit/hyperactivity disorder, this group difference in nocturnal restlessness and movements reached significance ($p < 0.0001$). With respect to the remaining sleeping behaviours as well as sleeping characteristics, the children with ADHD as a group did not differ from the controls.
Table 6. Results from analyses of variance with means and standard deviations for sleeping characteristics and behaviour during sleep, five years after initial contact

<table>
<thead>
<tr>
<th></th>
<th>Cases (N = 24)</th>
<th>Controls (N = 26)</th>
<th>p value*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M</td>
<td>Sd</td>
<td>M</td>
</tr>
<tr>
<td>Night-time sleep (in minutes)</td>
<td>628.8</td>
<td>56.08</td>
<td>639.5</td>
</tr>
<tr>
<td>Number of night wakings</td>
<td>0.5</td>
<td>0.67</td>
<td>0.3</td>
</tr>
<tr>
<td>Night time awake (length of arousal time at night in minutes)</td>
<td>4.7</td>
<td>8.10</td>
<td>1.2</td>
</tr>
<tr>
<td>Daytime sleep (in minutes)</td>
<td>0.0</td>
<td>0.00</td>
<td>0.0</td>
</tr>
<tr>
<td>Total sleep during a 24-hour period</td>
<td>628.8</td>
<td>56.08</td>
<td>639.5</td>
</tr>
<tr>
<td>Behaviour (5-step scales):</td>
<td>Cases (N = 24)</td>
<td>Controls (N = 107)</td>
<td></td>
</tr>
<tr>
<td>Snoring or breathing difficulties during sleep</td>
<td>0.3</td>
<td>0.45</td>
<td>0.5</td>
</tr>
<tr>
<td>Head banging</td>
<td>0.2</td>
<td>0.65</td>
<td>0.0</td>
</tr>
<tr>
<td>Restless movements</td>
<td>1.2</td>
<td>1.34</td>
<td>0.4</td>
</tr>
<tr>
<td>Nightmares</td>
<td>0.7</td>
<td>0.65</td>
<td>0.5</td>
</tr>
<tr>
<td>Sleep terrors (Pavor Nocturnus)</td>
<td>0.3</td>
<td>0.54</td>
<td>0.1</td>
</tr>
<tr>
<td>Sleep walking (Somnambulism)</td>
<td>0.2</td>
<td>0.67</td>
<td>0.1</td>
</tr>
<tr>
<td>Sleep talking (Somniloquy)</td>
<td>0.8</td>
<td>0.83</td>
<td>0.5</td>
</tr>
<tr>
<td>Tooth grinding (Sleep Bruxism)</td>
<td>0.3</td>
<td>0.78</td>
<td>0.5</td>
</tr>
<tr>
<td>Bedwetting</td>
<td>0.8</td>
<td>1.39</td>
<td>0.3</td>
</tr>
<tr>
<td>Anxiety in the settling situation</td>
<td>0.1</td>
<td>0.29</td>
<td>0.1</td>
</tr>
<tr>
<td>Protests against going to bed</td>
<td>1.0</td>
<td>1.41</td>
<td>0.5</td>
</tr>
</tbody>
</table>

* p value from t-tests

** All values are the same for one class level
Factors associated with subsequent development of attention-deficit/hyperactivity disorder (Study IV)

Of the 27 children with previously severe sleep problems in infancy (cases), 25 participated in the screening procedure for ADHD. Two families had moved (returned to their home countries). Eight children in this group were screen-positive and went further to assessment. Of those, one child did not qualify for any diagnosis of neurodevelopmental/neuropsychiatric disorder. The rest, seven children in the former sleep problem group, met the criteria for ADHD. Five of those children also had additional motor-perceptual problems including gross motor, fine motor, perceptual and speech/language dysfunction. Severe behavioural problems, dominated by conduct problems, were seen in three of the children. One of the children was diagnosed by the district paediatrician and the local CHC team before the screening study (at 4.5 years of age).

In the group of 27 control children, 25 agreed to participate in the general screening. One family had moved and one refused to participate, referring to lack of time.

One child among the controls was screen-positive but in the following assessment procedure did not qualify for any diagnosis. To summarize, seven of the former sleep problem children met the criteria for the diagnosis of ADHD, compared with no child in the control group.

The diagnosis ADHD was thus significantly more prevalent in children who previously had severe infant sleep problems than among the controls, 7/25 versus 0/25 (p < 0.01).

Factors associated with later development of ADHD in children with severe infant sleep problems were sought by comparing infant data of those children in the sleep problem group that later developed ADHD (N = 7) with infant
data from the rest of the problem group (N=20). Given severe sleep problems in infancy, the following characteristics were associated with later diagnosis of ADHD; psychosocial problems in the family, behavioural problems at settling time (bedtime struggles) and a long sleep latency at bedtime (Table 7 and 8). There was also a tendency towards more behavioural problems during daytime (problematic behaviours) in infancy, according to parental ratings four and a half to five years earlier, in the children that later met the criteria for ADHD. These children also tended to have been rated by the paediatrician as very active as infants (Table 7).
### Table 7. Means and standard deviations for health characteristics, infant difficultness, problematic behaviours and sleeping characteristics at 6-12 months of age

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Children with ADHD (n=7)</th>
<th>Rest of former sleep problem group (n=20)</th>
<th>p-value*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean</td>
<td>SD</td>
<td>Mean</td>
</tr>
<tr>
<td>Health characteristics</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Infant health problems</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Severe episodes</td>
<td>0.7</td>
<td>0.76</td>
<td>0.1</td>
</tr>
<tr>
<td>Moderate</td>
<td>1.1</td>
<td>1.21</td>
<td>0.6</td>
</tr>
<tr>
<td>Common, banal</td>
<td>2.0</td>
<td>1.00</td>
<td>1.5</td>
</tr>
<tr>
<td>Infant’s contact with medical care</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Specialist, paediatrician</td>
<td>1.7</td>
<td>1.38</td>
<td>0.9</td>
</tr>
<tr>
<td>General practitioner</td>
<td>1.9</td>
<td>1.46</td>
<td>1.1</td>
</tr>
<tr>
<td>Psychosocial problems</td>
<td>1.0</td>
<td>0.00</td>
<td>0.5</td>
</tr>
<tr>
<td>General development of the infant</td>
<td>3.3</td>
<td>0.76</td>
<td>3.2</td>
</tr>
<tr>
<td>Activity level in infant</td>
<td>1.8</td>
<td>1.10</td>
<td>0.6</td>
</tr>
<tr>
<td>Difficultness</td>
<td>3.8</td>
<td>1.70</td>
<td>3.9</td>
</tr>
<tr>
<td>Problematic behaviours</td>
<td>3.8</td>
<td>1.92</td>
<td>2.0</td>
</tr>
<tr>
<td>Sleeping characteristics</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bedtime struggles (7-step scale)</td>
<td>6.6</td>
<td>0.55</td>
<td>4.9</td>
</tr>
<tr>
<td>Sleep latency at bedtime (min)</td>
<td>55.4</td>
<td>46.6</td>
<td>21.1</td>
</tr>
<tr>
<td>Night-time sleep (min)</td>
<td>567.9</td>
<td>116.14</td>
<td>554.2</td>
</tr>
<tr>
<td>Number of night wakings</td>
<td>6.3</td>
<td>3.20</td>
<td>5.9</td>
</tr>
<tr>
<td>Night-time awake (length of arousal time at night) (min)</td>
<td>76.7</td>
<td>47.26</td>
<td>70.1</td>
</tr>
<tr>
<td>Daytime sleep (min)</td>
<td>116.1</td>
<td>85.72</td>
<td>104.6</td>
</tr>
<tr>
<td>Total sleep during a 24-h period</td>
<td>684.0</td>
<td>137.53</td>
<td>658.8</td>
</tr>
</tbody>
</table>

*p value from t-tests

SD, standard deviation
Table 8. Number of families with different kinds of psychosocial problems when the child was 6-12 months old

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Children with ADHD (n = 7)</th>
<th>Rest of former sleep problem group (n = 20)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Unemployment</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>Insecure employment</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Single mother</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Chronic disease/disability in parent</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Maternal depression</td>
<td>2</td>
<td>9</td>
</tr>
<tr>
<td>Serious family conflicts</td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>Refugee status</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Substance abuse</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Previous assault and battery against mother in the family</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Father previously in prison</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>No psychosocial problem</td>
<td>0</td>
<td>11</td>
</tr>
</tbody>
</table>

Note: More than one problem can be reported per family
DISCUSSION

General discussion

Study I, performed on a large total community sample, confirms results from other populations concerning the proportion of sleep problems as perceived by parents (20-22,67,69,92). The designation of infant sleep problems as an area of major parental concern is obvious as parental reports showed that 16 % of the infants aged between 6 and 18 months were judged to have moderate to severe difficulties in falling asleep at night, and 30 % were reported to have frequent night waking.

A core group of children with severe or even very severe sleep problems, encompassing around 5 % of the population, could be identified in different ways. About 4 % of the infants had sleep refusal problems that were considered severe or very severe by parental rankings and by the more operational definitions of ICSD. The corresponding figure for night-waking problems was 6 %. Five per cent of the parents reported themselves as being in current need of help.

Almost all parents had sought help at a CHC or more seldom at a paediatric clinic. Only approximately half of the parents were satisfied with the help obtained. Obviously something is lacking. Further studies of the interaction between parents and professionals regarding sleep problems are needed in order to determine the nature of the problem. Is there a problem in the information provided, in the way it is provided, or are the parents’ expectations not in concordance with those of the profession? Further research is urgent.
The delineation of factors associated with severe sleep problems was one of the major aims of the study. Results both from study I and from study II indicate that severe sleep problems are related to a number of factors. The most striking factors associated with severe sleep problems in the parent population study I were intensive parental interventional behaviour (especially feeding) during the evening and night, parental worries and anxiety concerning infant health (although their children were reported as being as healthy as others) and infant feeding problems. The parents of the children in the problem group seemed to be more insecure in their parental role, a finding that was confirmed in study II. It is of course always difficult to ascertain whether the behaviour of parents actually contributes to the development of the sleep problem of the child or whether, because the child acts in a particular way (will not settle or wakes and cries), the parents adopt certain behaviours (eg rocking, feeding) to get the child to sleep. Works undertaken by Kerr et al (68) in the area of prevention, however, support the first view. Altering the parent’s behaviour in the early months of the child’s life in a preventive study produced a statistically significant reduction in the incidence of sleep problems in the intervention group when compared to a control group at 9 months of age.

In study II, the closer study with in-depth interviews and examinations of the sleep-disturbed infants, a number of factors associated with severe sleep problems were found: characteristics of the child, of the parents and of their psychological and social situation.

Infants with severe and chronic sleep problems showed normal health and general development and had a normal growth pattern. Early severe sleep problems were, however, found to be associated with a higher incidence of breast-feeding, frequent night meals as a method of putting the infant back to sleep, co-sleeping and previous sleeping problems in the family. They were
also associated with non-European origin, living in a low income area and with psychosocial problems in the family, including financial problems, exhaustion and depression in the mother, less positive perceptions of parenting and more parental stress. There were also feelings of incompetence and restriction by the parental role, and perceived health problems. An association with infant difficultness, high activity and problematic behaviours were found.

There are potential dependencies of certain of the demographic variables (socio-economic status, ethnicity) and other variables (such as the rate of co-sleeping, breastfeeding and nap frequency) which make it more difficult to evaluate the contribution of each factor. However, we do not make any causal conclusions but believe that it is of clinical importance for prevention and early identification in child health care and clinical paediatric settings to be able to point to and identify common patterns and risk conditions associated with severe sleep problems.

Earlier findings were replicated in study II. More frequent night-waking has been reported in breastfed infants, (3,9,26,40,71,89) and in children, most often breastfed, who are given frequent nightmeals (3,89). An association between co-sleeping all night and problems with bedtime struggles and night-waking has also been reported by several studies (3,62,67,75-77,86,98,114,125,129). Previous sleeping problems in the family have been less investigated, and as an experienced researcher concluded: "little is known about how variations in social expectations and norms are related to sleep patterns or sleep problems" (100). A clear hereditary component is only known in parasomniacs (64).

The association of sleep problems with psychosocial problems in the family, including financial problems and overcrowding, is well known. (3) Exhaustion (30) and depressive state, especially in the mother, have previously been widely documented (73,76,98,129) as have less positive
perceptions of parenting (3,19,76). The parent-child interaction has been studied, and oversensitivity to the signals of the child (20,89) as well as insecurity in the parents have previously been found. Other contributors include inadequate stimulation of the child, overanxiety, rigidity or inconsistent handling from the parents (85) and health problems in the family (76).

In recent years the temperament of the child has come into focus, and infant temperamental difficultness in sleep problems has been documented, (3,9,12,27,82,98-99) as well as problematic behaviour, (6,82,98,129). The question of hyperactivity in the sleepless child is still controversial. It has been stated that there is no convincing evidence that sleepless children in general are hyperactive per se, and many of them "calm down" when their parents change their attitudes and establish consistent routines (100). A core group of true hyperactive children with sleep problems most certainly exists, but in the majority of very active children, this activity level has been seen as a consequence rather than a cause of their sleeplessness.

In study II we did not measure infant temperament before the sleep problem started. This means that we cannot rule out the contribution of infants’ temperamental characteristics, and in study IV we later found that approximately one in four children with severe sleep problems in infancy will later qualify for the diagnosis of ADHD.

The results of study II are thus in line with earlier findings. What may be new is the massive impact. Together, our results give a very complex, large-scale picture of how the infant and family situation develops in cases of very severe infant sleep problems. This is a picture in which multiple factors may amplify each other in vicious circles. Characteristics of the child, of the parents and of the situation may influence each other reciprocally. Of course there are methodological limitations here. Interpretation of a multifactorial genesis of
the problem could not strictly be assigned from univariate analyses but a not too improbable *speculation* is that when a vulnerable child with a temperament characterized by difficulty, high activity, liveliness, strong will and stubbornness, and with heredity for difficulties in the regulation of sleep and wakefulness, is born into a family with great psychosocial problems, health difficulties or a tendency to depression in the parent(s), there emerges a situation of excessive parental stress. Living in a foreign country, with other child-rearing traditions and far away from relatives, as for the non-European families (half of them refugees), adds even more stress to the situation. The higher risk of sleep disturbances in children of first generation immigrants, possibly due to a periodic effect of the stress of cultural change, has recently been discussed by Rona et al (106). In such cases, the child’s needs may exceed the parents’ capacity to meet them.

A feeling of insufficiency and insecurity in the parental role, desperation, and finally depression, in the parent can be the result. The child, who probably more than other children needs regularity and routines, is met instead by resignation and easy solutions, such as pacifiers, on-demand breast-feeding and co-sleeping. A chronic lack of sleep may be the result for the parents with such consequences as extreme tiredness, physical and mental exhaustion, irritability and relationship problems. For the child, there may be exaggerated difficulties and behavioural problems that maintain the vicious circle. Sleepless and depressed parents may have neither the energy to solve the situation themselves nor an ability to grasp the situation and seek help.

Our study, with its very small drop-out rate, has identified an unselected population of very difficult cases. Out of these, three quarters had spontaneously sought help. The remaining quarter of the families had all regularly visited their CHC for weight and length controls, and vaccinations, but they had not mentioned the severe sleep problems of their child.
Thus, in order to discover the severe cases of sleep problems among infants and young children, paediatricians and CHC need to actively ask parents about the sleep of their children. Severe problems are otherwise underreported.

In study III, infants with severe and chronic sleep problems showed significant improvements in all night-time sleep characteristics after a relatively short, but intensive and multi-disciplinary, intervention sleep programme. The improvements proved to be stable over time, lasting the whole follow-up period of five years. Continuous therapist support was not necessary to maintain the changes in sleep behaviour. In fact no contact between the special CHC team and the case families took place during the follow-up period, besides the follow-up questionnaires and a few telephone calls from three of the families.

Furthermore, earlier findings were replicated in this study. The benefits of a brief behavioural intervention programme for improving sleep in infants and young children have previously been documented (4,36,43,79,116,128). Of course it cannot be ruled out that a gradual achievement of a normal sleep pattern would have been the natural history for some of the children in the problem group, even without treatment.

Controlled crying was only one part of the programme, but is perhaps the most controversial one. It has been argued that leaving a child of this age to cry for even short periods may affect its long-term feeling of security (117). There is no support for this belief in our results. On the contrary, no case child showed any signs of anxiety in the settling situation one year after treatment, and the control children were more prone to protest against going to bed than were the cases at the two-and-a-
half-year follow-up. However, further research on follow-ups of behavioural sleep disturbances is needed.

Tendencies towards parasomnia (restless movements during sleep, head banging, sleep walking) were seen in the case group at the follow-up at one year but not at the two-and-a-half-year follow-up. After five years there was only a slight tendency for the problem group to move more during sleep than the controls. Further research will be necessary in order to determine whether the infants with early sleep problems are more prone to develop true parasomnia as they grow older than are the controls.

We believe the interdisciplinary approach in the clinical interventions to be crucial (87,107). The whole family situation must be taken into consideration (11) in order to overcome vicious circles of insecurity in the parental role, feelings of insufficiency, desperation and, finally, depression. Given understanding and the tools of a more secure way of parenting a child, the majority of even heavily psychosocially burdened parents were able to manage to help their child sleep well.

**Validity of the results**

The source of information is a crucial issue in the evaluation of sleep disturbances in early childhood. The reliability and validity of parental reports in the form of Sleep Diaries, the most common assessment instrument in clinical practice, have been questioned in the literature. It has been stated that parents might be unaware of many aspects of the sleep behaviour of their infants during the night (6,108,111). Consequently, a number of objective sleep measures have been employed, including polysomnographic studies, pressure-sensitive mattresses, timelapse video recordings and actigraphy (activity-based monitoring).
Results from studies comparing parental reports with objective sleep measures (109) show that parents are accurate reporters of sleep-schedule measures such as sleep onset and sleep duration. They overestimate, however, the time that their infants spend in actual sleep and underestimate the number of their night wakings. According to that result, the short sleep duration and many night wakings initially in our problem group should be regarded as a minimum rather than be suspected of being an exaggeration by exhausted parents. It could of course be argued that parents who co-sleep with their children are likely to be aware of more night-time wakings than would parents who sleep in different rooms. And since the parents in the problem group co-slept with their children much more frequently than did parents in the control group, one might suspect an artefact in reporting. However, the difference in number of night wakings between the groups is very large, as is the difference in length of reported time spent awake in the night. Even if an artefact cannot be ruled out, it is not reasonable to believe it to have had a major impact on the results.

The total community sample of 2,518 children, with a response rate of 83%, does not exhibit any known bias. Hence, results based on data from this sample are assumed to be generalisable in children from similar sociodemographic backgrounds, i.e. Scandinavian and probably North-European larger cities.

Concerning reliability it has to be noted that strict alternative answers give increased reliability but could be a threat to the validity by loss of information. We tried to solve this dilemma by combining survey data with interviews with both strict alternatives and open-ended questions.
The most controversial results in this thesis emerge in study IV, in which a discussion of methodological issues is urgent. According to our results, approximately one in four children with severe sleep problems in infancy will later qualify for the diagnosis of ADHD. This is a figure which clearly exceeds the estimated prevalence rate in this age group in a normal Swedish population, 2.4-4 % (72). However, the small numbers of children involved in the study, both at the screening and diagnostic stages, constitute an obvious drawback. The small sample size limits the extent to which conclusions can be drawn. We therefore regard our findings as preliminary and suggestive of a need for larger-scale corroborative studies.

Nevertheless, even though numbers were small, the sample examined was population-based and thus more likely to be representative of children with severe infant sleep problems than most studies reporting on clinical case series. Moreover, the drop-out rate at various stages of the study was very small. The longitudinal prospective nature of the study is also an advantage.

Individual, family and social factors were associated with later development of ADHD. Most striking were the associations with bedtime struggles and psychosocial problems in the family. ADHD has been described as a "biopsychosocial disorder", raising critical questions concerning the relations between genetic, biological and environmental factors (122).

The findings that there were a number of family psychosocial problems associated with the development of ADHD in infants with severe sleep problems need a comment. It has been stated that parents of children with ADHD have a high likelihood of ADHD themselves, and thus the family dysfunction may be a by-product of the parents’ own adult type of ADHD-related psychosocial difficulties.
What is then measured with the criteria of ADHD? Behaviour reflecting genetic predisposition to a deviant functioning of the synapse–transmittor system with difficulties in the regulation of sleep and wakefulness? Behaviour reflecting psychosocial stress? Behaviour reflecting vulnerability and difficultness in temperament? Potentially neuropathogenic events during the pregnancy, perinatal and postnatal periods, which are known to be associated with a heightened risk of development of ADHD (96), had been excluded through the inclusion criteria. Again we return to the speculations in page 56.

And why do we see a positive development in sleep characteristics but not the same concerning behaviour and development? This is an urgent task for future research.

**Conclusions and clinical implications**

Sleep problems are a common parental concern during infancy, and many parents seek advice and help from a CHC or paediatrician. Severe sleep problems in infants and young children are complex problems with both psychological and social factors involved, besides the strictly paediatric issues. In addition, many families are not satisfied with the help they obtain today. Even more serious, the severe cases are underreported by parents.

These findings have implications for the consultations at CHCs and for the paediatric consultation in cases of severe sleep problems in infants and young children. In order to discover the severe cases, health care personnel have to actively ask parents about sleep problems. In addition to obtaining a thorough history and making a complete medical examination of the child, physicians also need to pay great attention to the situation of the parents, their worries, and psychological and social conditions.
The possibility of a vicious circle developing deserves attention in clinical interventions, which need to be interdisciplinary, taking the whole family situation into consideration.

With a relatively short but intense treatment programme, combining behavioural technique with family work, even severely sleep disturbed infants and children, coming from families with depression and psychosocial problems, can be helped to sleep well. Long-term improvements in sleep pattern after the programme, extending over years, have been documented.

The results concerning the development of attention-deficit/hyperactivity disorder call for heightened attentiveness and awareness among nurses and doctors working with infants with sleep problems at CHCs and paediatric clinics. Infant activity level, problematic behaviours and parent-infant interaction have to be considered seriously, and both psychosocial, behavioural and sleeping characteristics have to be taken into consideration. Close follow-ups of children with combined severe and prolonged sleep, activity and behavioural problems are recommended as neurodevelopmental problems seem to be prevalent in this category of children.
ACKNOWLEDGEMENTS

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REFERENCES


