THE ASSESSMENT AND TREATMENT OF
COMMON SLEEP PROBLEMS IN SEVERELY LEARNING
DISABLED CHILDREN

MD THESIS

1996

BY

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David John Bramble
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SUMMARY

This thesis explores by means of a small intervention study based on ordinary clinical practice, the effectiveness and acceptability to carers of an intensive behavioural modification approach to the chronic and severe night-settling and night-waking (NS/NW) sleep problems commonly encountered with severely learning disabled (SLD) children. The principal hypotheses tested were: (i) rapid settling and extinction techniques represent an effective means of treating severe and chronic NS/NW sleep problems presenting in severely learning disabled children; (ii) such an approach will also provide useful generalization effects; (iii) such an approach will prove acceptable to the parents of affected children; and (iv) electronic ambulatory movement monitoring (AM) will yield useful data in this context. Fifteen sleep disordered SLD children were recruited into the trial and there were no drop-outs over its course. The results of the study reflect that the intervention was highly successful and that improvements in the children's sleep problems were evident from subjective and objective assessments within a few days of the commencement of treatment. At long-term follow-up, these improvements were sustained in all but three of the families. Generalization effects were seen in that the children's daytime behaviour improved as did the indices of their mothers' subjective wellbeing. The parents were highly satisfied by both the results and the style of the interventional approach. No adverse effects were reported. The AM data confirmed the trends of improvement detected in the parent and investigator assessments (night-waking patterns; settling times; etc.) and also provided novel information about the duration, movement content and temporal distribution of night-waking episodes over the course of the study for five of the children. These findings are discussed in terms of their implications for clinical practice and future research.
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DECLARATION

I certify that this thesis represents original research carried out by me with the help of those persons outlined in the acknowledgements. It does not contain material previously submitted for a degree or diploma in any university and, to the best of my knowledge and belief, does not contain any material previously published by any other person, except where due reference is made in the text.

David John Bramble

David John Bramble
This thesis was inspired by two senior child psychiatrists who both have a special sensitivity to the psychological needs of severely learning disabled children and their families. The first is Professor A. R. Nicol who "firmly but fairly" guided me in his capacity of academic supervisor to this project which for me served as an apprenticeship in academic medicine; the second is Professor J. B. Pearce who was my clinical supervisor for two years and whose highly effective dynamic and humane work with psychiatrically disordered learning disabled children inspired the project which is the subject of this thesis. Dr D. S. Stretch and Mr. S. Dunkley guided my fumbling steps through the minefield of small study design and non-parametric statistics. My colleagues in both the child psychiatry and learning disability services provided a wealth of clinical experience and support while I was undertaking the project field work and I thank Dr. Agnes Hauk, Dr. R. A. Collacott and Dr. S. Bhaumik particularly in this respect. Professor J. Home of Loughborough University and Dr. Lyn Quine of the University of Kent provided invaluable advice and encouragement while I was devising this study. I am indebted to a succession of Academic Department secretaries for their help in improving my clerical and word-processing skills and particularly Mrs. Irene Chenery of the Leicester University Department of Psychiatry and Mrs. Audrey Strong and Mrs. Susan Hooper of the Nottingham University Department of Child and Adolescent Psychiatry. I thank my wife, Michelle, whose support, forbearance and critical comments helped to bring this thesis to the point of submission before its results would be of interest only to medical historians. Finally, I would like to thank all of the families who participated in this study from whom I learnt a great deal more about what it takes to care for severely disabled children than can be presented in this or any thesis. Trent Regional Health Authority provided a grant for this study.
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<td>ABS</td>
<td>Adaptive Behavior Scales (&quot;Vineland&quot;)</td>
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<tr>
<td>AM</td>
<td>Ambulatory Monitor (ing)</td>
</tr>
<tr>
<td>BPI</td>
<td>Behaviour Problem Index</td>
</tr>
<tr>
<td>DB</td>
<td>Dr David Brumle, Principal Investigator</td>
</tr>
<tr>
<td>LD</td>
<td>Learning Disability (Mental Handicap)</td>
</tr>
<tr>
<td>MI</td>
<td>Movement Index</td>
</tr>
<tr>
<td>MMTW</td>
<td>Mean Movement per active Time Window</td>
</tr>
<tr>
<td>MWT</td>
<td>Morning Waking Time</td>
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<tr>
<td>MSS</td>
<td>Maternal Sleep Scale</td>
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<tr>
<td>NREM</td>
<td>Non-Rapid Eye Movement</td>
</tr>
<tr>
<td>NS</td>
<td>Night-Settling</td>
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<tr>
<td>NST</td>
<td>Night-Settling Time</td>
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<tr>
<td>NS/NW</td>
<td>Night-Settling and Night-Waking</td>
</tr>
<tr>
<td>NW</td>
<td>Night-Waking</td>
</tr>
<tr>
<td>NWD</td>
<td>Night-Waking Duration</td>
</tr>
<tr>
<td>REM</td>
<td>Rapid Eye Movement</td>
</tr>
<tr>
<td>SJ</td>
<td>Samantha Jones, Medical Student, Co-Investigator</td>
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<tr>
<td>SOT</td>
<td>Sleep Onset Time</td>
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<tr>
<td>TW</td>
<td>Time Window</td>
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<tr>
<td>TWN</td>
<td>Time Windows per Night</td>
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<tr>
<td>TSP</td>
<td>Total Sleep Period</td>
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<tr>
<td>TST</td>
<td>Total Sleep Time</td>
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SUMMARY

This thesis explores by means of a small intervention study based on ordinary clinical practice, the effectiveness and acceptability to carers of an intensive behavioural modification approach to the chronic and severe night-settling and night-waking (NS/NW) sleep problems commonly encountered with severely learning disabled (SLD) children. The principal hypotheses tested were: (i) rapid settling and extinction techniques represent an effective means of treating severe and chronic NS/NW sleep problems presenting in severely learning disabled children; (ii) such an approach will yield useful generalization effects; (iii) such an approach will prove acceptable to the parents of affected children; and (iv) electronic ambulatory movement monitoring (AM) will yield useful data in this context. Fifteen sleep disordered SLD children were recruited into the trial and there were no drop-outs over its course. The results of the study reflect that the intervention was highly successful and that improvements in the children's sleep problems were evident from subjective and objective assessments within a few days of the commencement of treatment. At long-term follow-up, these improvements were sustained in all but three of the families. Generalization effects were seen in that the children's daytime behaviour improved as did the indices of their mothers' subjective wellbeing. The parents were highly satisfied by both the results and the style of the interventional approach. No adverse effects were reported. The AM data confirmed the trends of improvement detected in the parent and investigator assessments (night-waking patterns; settling times; etc.) and also provided novel information about the duration, movement content and temporal distribution of night-waking episodes over the course of the study for five of the children. These findings are discussed in terms of their implications for clinical practice and future research.
1: INTRODUCTION
Section 1: INTRODUCTION

1.1. INTRODUCTION TO SLEEP DISORDER MEDICINE

1.1.1. Overview of the Field

Sleep disorders are now recognised as representing a major public health problem. Surveys in developed countries have revealed that around fifteen percent of the population experiences chronically disturbed and unsatisfactory sleep (National Institute of Mental Health, 1984). The increasing demands of our 24-hour society promote the intrusion of wakefulness into our sleep and also sleepiness into our waking lives. There is a growing awareness of the importance of having a regularly refreshing night's sleep in meeting the demands and challenges of our busy lives. When this is not achieved, as Moore-Ede describes in his recent review of the effects of sleep disturbance upon Western society (Moore-Ede, 1993), the consequences are usually very serious: poor work records and underperformance; increased accident risk; increased risks of developing significant psychological and physical morbidity.

Sleep disorders are many and varied afflicting people of all ages and levels of ability. To date over eighty different disorders have been described, the largest group consisting of the various, and primarily psychological, forms of insomnia (American Sleep Disorder Association, 1991). Nevertheless, health services have only recently started to afford sleep disorders the importance and attention they deserve, a situation reflected by the fact that the majority of medical and psychological textbooks pay little if any heed to the subject. The majority of British medical schools offer no specific training to their
undergraduates in sleep disorder medicine (Stores, 1990) a clinical area which, in the United States and elsewhere, is now highly developed representing a medical speciality in its own right (Dement & Mitler, 1993).

The subject of sleep disorders within the field of learning disability is probably one of the least researched areas within the general field of sleep research a situation which is surprising considering the close interplay between organic and environmental factors which operates within the psychiatric disorders of this group. Again, most of the current specialist textbooks of learning disability do not refer to the topic despite the fact that sleep problems in learning disabled children have been shown to be very common indeed (Pahl & Quine, 1985; Bartlett et al, 1985; Clements et al, 1986) and represent a common source of significant problems for affected individuals and their families. It must be added that treatment strategies in the form of behavioural modification techniques (see below) are being developed in order to address these problems and appear to show clinical promise, yet only a few as yet have been systematically evaluated (Stores, 1992). It is therefore very probable that these approaches are not yet widely applied in current clinical practice with learning disabled patients and it is only relatively recently that the particular sleep problems of this section of society have stimulated research interest (Espie & Tweedie, 1991).

The intervention study which is the primary focus of this thesis was prompted by this currently unsatisfactory situation and its principal focus is the evaluation of a straightforward behaviour modification treatment approach to severe and chronic night-settling and night-waking (NS/NW) difficulties which present in a cohort of severely learning disabled children. As will be discussed, these particular sleep problems are among the most common types of behavioural disorder encountered in clinical practice
with both preschool non-retarded children and, particularly, significantly developmentally
delayed children.

1.1.2. Caring for Children with Learning Disabilities: a Historical Perspective

Over the last thirty years our society's attitudes towards the rights and responsibilities of
people with intellectual and physical disabilities have changed considerably. It is now the
case that even profoundly and multiply disabled children are expected to remain with their
natural families or, alternatively if this is not possible, grow up in suitable alternative
family settings; the aim being for such children to have the opportunity to experience as
"near normal" a life as possible with the implicit intention of preventing the development
of many of the social handicaps so strongly associated with this section of the population.
This was the chief recommendation of the Jay Report (Department of Health and Social
Security, 1979) which provided both the basis and also the impetus for subsequent
community care legislation in this field. It was expected that flexible and comprehensive
community-based support systems would evolve to facilitate this; these would necessarily
require that health, educational, welfare and voluntary services work together with parents
of affected children in a joint partnership. Currently in Britain only approximately one
percent of disabled children receive long-term specialist residential care (Office of
Population and Census Surveys, 1989) and this figure is expected to further reduce.
Unfortunately, the politically expedited rapid closure of hospitals and specialist units for
the learning disabled has not been matched with a commensurate expansion of community-
based supportive services for these people and their carers. Consequently, many families
caring for severely learning disabled children who, commonly, also have multiple dependency needs are now faced with both a restricted choice of service support and also an increased burden of care, a burden which usually falls most heavily on such children's mothers. Rhetoric which refers to the "community care" of such children tends to obscure this fact. In his early review of the subject, Baldwin (1987) discovered no convincing evidence that the community at large plays any significant part in the overall care of the vast majority of retarded children: most mothers care for their children with little support even from spouses and other members of the immediate family; consequently mothers pay a high price: such high dependency care requires time, financial commitment, thwarted career plans and, often, strained relationships within their families. Financial, physical and emotional health can also suffer significantly (Department of Health and Social Security, 1985).

1.1.3. Behavioural Problems

About half of all children with severe learning disabilities also have significant behavioural problems which commonly take the form of oppositional behaviour, temper tantrums, aggression, overactivity and, particularly, sleep problems (Pahl & Quine, 1984; Corbett, 1979). These types of problems are highly associated with principal carer and family stress, the direction of causality having been established as being primarily the result of the influences of the children's problems upon their families and not vice versa (Quine & Pahl, 1989). Behaviour problems and particularly sleep problems are commonly cited as reasons for urgent requests for help from health and welfare services (Chetwynd, 1985; Quine & Pahl, 1989).
In their large community survey of families with severely learning disabled children, Quine and Pahl (1989) discovered a particularly strong association between night-settling and night-waking (NS/NW) sleep problems and impaired maternal wellbeing confirming Richman's earlier finding for the mothers of sleep disordered non-retarded children (Richman, 1981). Characteristically, such children take a long time, sometimes several hours, to settle to sleep in the evening, many managing to do so only when sharing a bed with their parents. The pre-bedtime period is associated with temper outbursts and screaming fits which are highly distressing for all parties involved. Even if these children settle eventually, they often wake repeatedly during the night and either seek out their parents directly or scream, headbang, shout and even vomit to gain their attention (Pearce, 1991). When this sleep disruption occurs in addition to the high levels physical and emotional stress which parents of demanding and dependent children commonly experience (sleep problems are also strongly associated with daytime behavioural problems Richman et al, 1981), often the result is exhausted and sleepy principal carers whose parenting skills, mood and general effectiveness are further compromised. Douglas (1989) comments on these long-term effects of continuing sleep disruption on affected parents. This chronic cycle of sleep disruption and carer exhaustion with consequent sub-optimal daytime parenting capacities becomes ingrained within affected families and thus a self-reinforcing process. It has been the clinical experience of the author and that of several colleagues with an interest in childhood sleep problems, that successful intervention with such families usually requires the initial treatment of any coexisting significant sleep problems before other behavioural or emotional problems can be successfully addressed. It would also appear to be the case that successful treatment of a severe NS/NW problem often yields beneficial generalization effects in terms of
improved daytime behavioural problems and improved maternal wellbeing (Quine & Wade, 1991).

1.1.4. Normal Changes of Sleep with Age (Sleep Ontogeny)

Sleep is a reversible behavioural state characterised by predictable, profound, altering shifts in levels of arousal. As with most other vital physiological processes, it changes in both its quality and also, albeit to a lesser degree, its quantity throughout life. It is not known why we sleep although there are several competing theories. Currently the most compelling of which is that proposed by Horne who has conducted a series of studies which have tested subjects' psychological functioning during states of general and also selective sleep deprivation. In his review of his own work (Horne, 1991) he concludes that human adults require only a few (four or five) hours of sleep per day to perform optimally. He refers to this as "core" sleep; the additional sleep time that most people obtain is determined primarily by their personal circumstances and also societal pressures. Horne refers to this extra sleep as "optional" sleep. He uses the analogy of our food needs: we require only a certain amount to survive, the rest we eat for a variety of reasons. Horne speculates that only "core" sleep comprising a high proportion of "deep", or slow-wave (SWS), sleep is required for optimal brain functioning and, specifically, the prefrontal areas of the cerebral cortex. Only when subjects are selectively deprived of this type of non-rapid eye movement (NREM) sleep are the characteristic psychological features of sleepiness are seen: impaired attention and verbal fluency, for example; whereas selective deprivation of rapid eye movement (REM) and "light" sleep (see
below) do not result in such psychological impairments.

Direct and videotaped observation of newborn babies has revealed that they sleep most of the day, waking every hour or so to feed and/or eliminate. A cycle of sleep and wakefulness is quickly established, and by three months of age the infant is sleeping for four hours at a stretch. This gradually extends to approximately twelve hours at six months with additional regular daytime naps totalling about three hours; the implication of this from a clinical perspective is that infants are physiologically capable of sleep through the night by the end of their first year, unless physical illnesses which effect brain function (particularly epilepsy and space occupying lesions) are also operating during this time. From the end of the first year to mid-toddlerhood, daytime sleep declines and the night sleep period slowly reduces to around nine hours by late childhood and eight hours by early adolescence (although there may be a temporary need for more sleep over the period of the characteristic growth spurt of puberty). Children may wake very briefly once or twice during the night, however this is rarely remembered unless waking is associated with a specific disorder such as a nightmare. Early adulthood sees a reduction in the total sleep period down to an average of seven and a half hours with a standard deviation of one hour. This figure remains remarkably constant into old age. Qualitative changes in the relative proportions of "light" and "deep" sleep occur during the normal ageing process: infants' sleep initially comprises high levels of REM sleep and, later, "deep" sleep appears which probably reflects rapid growth and development of the central nervous system (Denenberg & Thomas, 1981). From late infancy, SWS (stages 3 and 4) predominates in the first half of the night, the lighter sleep stages (stages 1, 2, and REM) in the latter half. Non-REM (stages 1 to 4) sleep and REM sleep cycles every ninety
minutes or so throughout the sleep period from late infancy into adulthood. As individuals age there is growing tendency for the quality of their sleep to become progressively "lighter" (i.e. consisting of proportionately less stage 3 and 4 sleep and more stages 1 and 2), a consequence of this in old age being an increased tendency to awaken during the night and be unable to fall asleep again (i.e. maintain sleep). Furthermore, the elderly experience increasing difficulty staying awake during the daytime consequently napping occurs.

1.1.5. Classification of Sleep Disorders

The field of sleep disorder medicine encompasses a most heterogeneous group of conditions and, as will be discussed, community prevalence rates are high. The largest group in adults consists of the various subtypes of "insomnia". In young children the commonest problems are those where children have marked difficulties in either initiating and/or maintaining sleep. More specifically, this involves their not settling at night and / or repeatedly waking up during the night usually involving the soliciting of parental attention (NS/NW problems; these are described above) These types of problem are classified among the "dyssomnias" in the fourth edition of the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 1994). However, they are more helpfully classified as "limit-setting sleep disorder" and "sleep-onset association disorder", respectively, in the latest edition of the International Classification of Sleep Disorders (American Sleep Disorder Association, 1991). This system reflects current understanding about both the aetiology and natural history of these disorders it also provides research criteria for their severity, duration and other characteristics. It is
perhaps indicative of the current geographical variability of the status of childhood sleep disorders that they are not classified independent of other disorders in the tenth, and most recent, edition of the International Classification of Mental and Behavioural Disorders (World Health Organisation, 1992) and childhood NS and NW problems are not identified at all in this scheme.

1.1.6. Epidemiology of Night-Settling and Night-waking Childhood Sleep Disorders

(a) General Population Rates

A nationwide American survey conducted nearly twenty years ago among child health clinicians reported prevalence rates of sleep problems for paediatric patients of 0.2% to 7.8%, whereas child psychiatrists reported rates of 0.2% to 19.4% in their clinic populations (Bixler et al., 1976). The most commonest types were NS and/or NW problems. In the first community-based epidemiological study of its kind, Richman and her colleagues (Richman, 1977) discovered that approximately 20% of a large representative urban British cohort of preschool children were reported by their parents as having significant NS/NW sleep problems and a quarter of these were severe. However, it must be emphasised that subsequent research has revealed that many families with repeatedly night-waking infants do not describe this behaviour as being a problem (Scott & Richards, 1989) a finding which underlines the importance of investigating family ecological factors in childhood sleep problems. Overall, NS and NW problems occur together; however, the latter form is commoner in younger children whereas NS difficulties tend to be more common in older children (Simonds & Parraga, 1982)

(b) Learning Disabled Populations
Several surveys of large community samples of severely learning disabled children (Pahl & Quine, 1989; Clements et al, 1986; Bartlett et al, 1985) have revealed that NS/NW difficulties are also the commonest type of sleep problems encountered in this group and that their prevalence rates more than doubles (to around half of their samples) compared to that for non-retarded children. Hogg and Lambe's (1988) community study of profoundly retarded children and young adults reported a three-fold increase in prevalence, revealing that NS/NW problems appear to be the norm for this group. Furthermore, these researchers discovered strong associations between these sleep problems and daytime behavioural difficulties in affected children and significant levels of stress for carers. Szyndler (1990) discovered an association between disturbed sleep and cerebral palsy in a cohort of young children; however, this was strongest for children who were also learning disabled. Repeated night-waking, in particular, was discovered to be associated with daytime self-injurious behaviour by Clements and his colleagues (1986). Quine and Pahl's (1989) important longitudinal follow-up study of a large community cohort of British severely learning disabled children revealed that behavioural problems, and particularly NS and NW sleep problems, tended towards chronicity in this group. They discovered that between half to three quarters of affected children continued to have marked NS/NW problems at the three-year follow-up stage of their study. This runs contrary to what tends to happen in non-retarded children: the vast majority of them "grow out" of these sleep problems by school age; however, even in this group, a small minority (between two to five per cent of five-year-olds) have persistent NS/NW problems (Kataria et al, 1987) and it is probable, although not yet firmly established, that many of such children will continue to have sleep difficulties and, especially, the various forms of insomnia throughout their lives. Zuckerman et al (1987) followed up a case-matched
cohort of several hundred sleep disordered non-retarded infants and discovered that nearly half of them continued to have these problems by late-toddlerhood; furthermore, these children showed significantly higher rates of other behavioural problems and particularly high rates of severe tantrums compared to their non-sleep disordered controls. These findings serve to emphasise the need for both preventive initiatives and also the elaboration of early detection and treatment strategies in childhood sleep disorder practice (Stores, 1992). In severely learning disabled children it seems that the strongest predictor of chronicity is the previous duration of the problem. Bartlett, et al's (1985) cohort was subdivided by subject's age and prevalence of sleep problems as follows: under six year olds: 86% of 183 had sleep problems; 81% of 172 six to eleven year olds and 77% of 165 eleven to sixteen year olds. This strongly suggests persistence of NS and NW problems with age in the severely learning disabled. The author's clinical experience suggests that persistence of these problems into adulthood is not uncommon, a suspicion supported by the findings of Hogg and Lambe's (1988) survey of several hundred profoundly retarded children. Yet the prognosis for sleep disordered children with lesser degrees of learning impairment is not known. A recent Scottish survey (Espie & Tweedie, 1991) which compared the rates of sleep problems between groups of young adults of all degrees of learning impairment living in community and hospital settings did not reveal excessive rates compared to the general population rates in either group; they did, however, discover that their hospital group did exhibit poorer sleep efficiency (time asleep divided by time in bed) and tended to wake more frequently compared to their community sample. Unfortunately, severely disabled individuals living at home with their parents comprised only a small subgroup of their "community" cohort. Thus it would appear that the prevalence rates and characteristics of the sleep problems of this vulnerable group remain
undetermined.

1.1.7. Developmental Effects of Sleep Problems upon Sufferers

The research field of childhood predictors of adult psychiatric problems is now well established. Most recently, it has expanded into the area of learning disability and has yielded important information. The strongest childhood psychiatric disorder which is predictive of adult problems is severe behavioural or conduct disorder (Rutter, 1985). Given that this particular type of disorder is known to be strongly associated with sleep problems (see above) there would appear to be a strong a priori argument for both the early detection and also effective treatment of these problems when they occur in learning disabled children (see below). It would seem that once well established, these sleep behavioural difficulties become whole-family problems and can add considerably to the burden of caring for handicapped children. Given the sparse nature of the current literature concerning this subject this area merits further systematic enquiry and how this might be achieved will be explored in the "Discussion" section of this thesis.

1.1.8. Co-Sleeping

Another common problem associated with NS/NW disorders concerns affected children co-sleeping with their parents. This can either result from failed attempts to persuade them to settle in their own beds in the evening or, alternatively, represent the eventual routine consequence of a night-walking episode. In the latter case children can gain access to the parental bed covertly; however, it is likely that most co-sleeping occurs with
parental "permission". Whether this behaviour is in itself regarded as being problematical depends upon the wider context of the sleep problem and particularly the parents' attitudes towards it. Historically, intergenerational co-sleeping was the norm until the relative affluence of the Industrial Revolution allowed many people to acquire accommodation which allowed sufficient room for children to sleep separately from their parents. Indeed, in upper-class Western society, it became fashionable for even parents to sleep separately in order to declare their material wealth (Elias, 1937). Across cultures there are marked variations in this practice and even between subcultures within complex Western societies. For example, Afro-American parents tend to allow their children to sleep with them into late-toddlerhood whereas Caucasians tend to encourage their children to sleep separately during the first few weeks of life and, at the latest, by the end of their first year. Some commentators suggest that co-sleeping may protect vulnerable infants from sudden infant death syndrome ("cot death") because there is some evidence of reciprocal physiological modulation between co-sleeping infants and their mothers (Mc.Kenna et al, 1990); however, others suggest that the historically widespread practice of co-sleeping has provided the opportunity for many parents to practice surreptitious post-natal family planning (Danton, 1984).

Clinical experience suggests that severely retarded children tend to co-sleep with their parents over longer periods of their lives compared to non-retarded peers. There is some empirical evidence to support this contention; for example, in Cunningham et al's (1986) cohort of Down's syndrome children revealed a co-sleeping rate of 41%. Quine and Pahl's (1989) longitudinal study of a heterogeneous cohort of severely retarded children revealed a 29% persistence of co-sleeping at the three year follow-up of their cohort; their initial rate was 83% suggesting an eventual resolution of the behaviour for most children.
To what extent co-sleeping practices in this group persist into later childhood, adolescence or even adulthood has not yet been investigated systematically. Nevertheless, it has been the author's experience whilst working in adult learning disability psychiatric services that a small minority of severely retarded adults do indeed continue to co-sleep with their natural parents and this might well represent unresolved separational problems operating between the parents and their children with a persistence of sleep patterns and problems from their children's infancies. The costs of the persistence of this specific behaviour for affected children and their parents are unknown. However, there is a growing recognition of the vulnerability of learning disabled children to various forms of abuse, and particularly emotional and sexual abuse, which would suggest that prolonged co-sleeping is not desirable and families should be counselled and helped accordingly. The author is aware of cases where it was parental recognition of their child's emergent sexual maturation which first brought long-term sleep problems involving co-sleeping practices to the attention of their clinicians. When co-sleeping is also part of a remorseless pattern of severe NS/NW problems, the practice requires urgent therapeutic attention in its own right.

1.1.9. Aetiology of Sleep Disorders in Learning Disabled Children

(a) Neurological Impairment

Speculation that learning disabled children as a group should have physiologically abnormal sleep patterns because they have higher rates of gross brain abnormality than
non-retarded children has been confirmed only in part. Heterogenous populations of
learning disabled children have been shown to have sleep electroencephalographic
evidence of differences in certain features of sleep architecture (the characteristic
electroencephalographic changes related to the sleep state). The "spindle" density of stage
2 sleep and also the total amount of REM sleep, for example, tends to be reduced
compared to that seen in age-matched controls to a degree which is proportionate to the
degree of intellectual impairment assessed by psychometry (Grubar, 1983). These findings
led to a degree of therapeutic nihilism when it came to the subject of the treatment of the
sleep problems presenting in severely retarded children (Osaka, 1987). Nevertheless, the
great majority of even the most profoundly retarded children who may also have gross
electroencephalographic abnormalities are capable of establishing sleep and wakefulness
patterns which are virtually identical to those of non-retarded children which are described
above. Also, the ontogeny of their sleep patterns appears to progress at a similar rate
(Stores, 1992). It is only in cases of a few specific handicapping conditions such as
tuberous sclerosis where there is disruption or even obliteration of the mid- and hind-brain
centres which control the circadian sleep-wake cycle (the suprachiasmatic nuclei of the
hypothalamus, for example) and also through the effects of severe epilepsy (or its
treatment - see below) where very high rates of NS/NW and other sleep problems are
seen (Hunt & Stores, 1994). Certain mucopolysacharidoses and Down's syndrome are
also known to be associated with obstructive sleep apnoea which can result in daytime
hypersomnolence and, consequently behavioral problems in sufferers. The Prader-Willi
syndrome is also associated with daytime sleepiness. Nevertheless, for the great majority
of severely learning disabled children it seems that organic factors play only a minor role
in the aetiology of their commonest form of sleep problem which is the NS/NW type.
Having conducted a large follow-up study of psychiatrically-disordered learning disabled children, Reid (1980) was the first clinician to emphasise that the persistence of such problems owed more to the children's high dependency needs and their carers' capacities to meet these rather than factors linked directly to their handicapping condition. This theme will be further discussed below.

(b) Epilepsy, Learning Disability and Sleep

The high rates of epilepsy in the learning disabled population (Corbett, 1979) may account for some sleep problems in this group by either the condition's direct effects upon sleep (nocturnal fits) or through the effects upon sufferers' sleep of anticonvulsant medication (Stores, 1991). The older agents phenytoin and phenobarbitone and also the longer-acting benzodiazepines are particularly sedating anticonvulsants which can produce excessive daytime somnolence with prolonged napping and thus result in a reduced need for sleep at night. Withdrawal phenomena occurring as a result of discontinuation or accidental omission of these types of medication could also, theoretically, disturb sleep. Thus when assessing any learning disabled child with NS and / or NW problems the possibility of such factors operating needs to be borne in mind and, where appropriate, addressed in their own right.

(c) Developmental Influences

Factors relating to emotional development, such as fears of the dark, are well known to influence bedtime behaviour and those severely retarded children who are in the upper
range of ability having a mental age of over three years are psychologically capable of experiencing this. Temperamental factors are known to be correlated with sleep and other behavioural problems (this is particularly the case for children with the so-called "difficult" temperament (Thomas & Chess, 1977); however, the research relating to the effects of temperament has been conducted primarily with non-retarded children and its applicability to learning disabled children is uncertain. The behavioural interpretation of NS/NW problems interprets them as being primarily the result of children not having learned a good sleep habit, lacking the appropriate routines and associations which promote sleep. Rough play, television and other forms of pre-bedtime overstimulation particularly interfere with sleep onset. These poor habits being reinforced by both parental overresponsiveness and also inconsistent limit-setting at bedtimes (Quine, 1991). The psychodynamic perspective emphasises separational anxieties and distorted object relations as being at least important maintaining factors in the NS/NW problems of childhood (see below). A family systems theoretical perspective might suggest that inadequate limit-setting by the parent, a generally disorganised parenting style and the lack of distinct boundaries resulting from emotional enmeshment between parents and their affected children at bedtime as being causally implicated. Quine and Pahl (1989) discovered another strongly predictive factor for the emergence of these types of sleep problem: in a pathway analysis of their data involving the factors associated with sleep problems they discovered that their cohort's impaired communication skills were a key factor. Indeed, these authors were prompted to speculate that the night-waking behaviour itself is a type of dysfunctional communication between affected children and their parents. Other commentators have speculated that the normal transient awakenings and fussing which children show in the second half of their first year of life which correspond
to the points of the phasic transitions from NREM to REM sleep can be misunderstood particularly by anxious or inexperienced parents as demands for food or comfort (Ferber, 1985). Should the infant receive these pleasant rewards routinely at these times, a self-reinforcing pattern of increasing alertness and expectation in such infants may be induced inadvertently thus setting the scene for a possible sleep problem when the infants' demands exceed their parents' willingness to respond to them thus leading to stress and the perception of a "problem" and, possibly, requests for professional help. This specific factor of the parent's presence in the child's bedroom when the child awakens during the night has been shown to have the strongest association with recurrent NW problems in non-retarded children and also serves as the most potent reinforcing factor (Adair et al, 1991). Particular aspects of the retarded child's emerging relationship with his parents may increase the likelihood of sleep problems emerging and these are discussed more fully in the next section.

(d) Influence of Parent-Child Attachment Processes

Intrapsychic developmental factors which operate within the severely retarded child and also his or her principal caregiver (usually the biological mother) are now recognised as being implicated in both the creation and also the maintenance of NS/NW problems (Daws, 1991). All of the theoretical frameworks alluded to previously can explain, at least in part, some of these factors, however in order to understand the quality of the feelings these problems tend to engender in mothers and their affected children it is necessary to comment on attachment theory and ego and object relationship perspectives within psychoanalytic theory. In this context the impact of retardation on the child and
its relationship with its caregivers is important for several reasons: (i) retardation involves delay and/or distortion of neurological development which can affect attachment responses in the child; (ii) illnesses and both sensory and psycho-motor handicaps can impair ego-integration and intensify dependency and the related feelings of conflict; (iii) additionally, through the powerful emotional states that this induces in parents and other caregivers (low self-esteem, guilt, denial, anger, despondency, etc.) it can indirectly affect the child's experience and hence the evolution of its internal world. Most of the work in this field has concentrated on the mildly or moderately retarded child because they have the capacity, albeit to a limited degree, to comment on their internal states and also to use play in an imaginative way thus allowing therapists to have some access to their subconscious processes. Severely retarded children, by implication, are usually incapable of this or, at best, only capable of communicating in these ways to a very limited degree. It is this particular difficulty which has led many psychodynamic psychotherapists to regard such children as "untreatable". This may be part of the explanation why it has also been the case, until relatively recently, that working with disturbed severely intellectually retarded children has not been popular with child psychiatrists generally.

In a review of this field, Levitas and Gilson (1987) attempted to show how the area of the psychopathology of mentally retarded children provides fertile ground for object-relations theorists. These authors refer to the "primary psychosocial deficit" of retarded individuals which is defined as their passivity, self-absorption, novelty avoidance and their very evident developmental deficiencies in terms of showing expressive nuance and comprehension of emotion. They argue that it is these precise elements that make caring for such children such hard work from a psychological point of view. They point out that many workers in this field (for example, Baker and Baker, 1987) have emphasised the
importance of the period of birth to late-toddlerhood in establishing a sense of individuality and then the eventual autonomy of the child through successful separation from the primary caregiver. It is axiomatic in object-relations psychology that successful separation of the child requires the prior development of successful attachments and intimacy formation. It is at this developmental stage where most severely retarded and all profoundly retarded children remain for the rest of their lives from an intrapsychic developmental point of view; therefore it would be impossible for a therapist who is aware of these developmental issues to engage families with psychologically disturbed severely or profoundly retarded children without interpreting any presenting "problem" as being underpinned by issues directly related to distorted and perhaps arrested object-relations transitions. Levitas and Gilson believe that there needs to be a coherence between the child's skills and talents and its parents' satisfaction with these skills in order to promote intrapsychic individuation to even a limited degree. In order for this to occur all parties have to relinquish primitive satisfactions however distressing this may be. Resistance to change in this way from the caregiver may further impede change in the child and this is a situation which is commonly encountered in NS/NW problems.

Unresolved parental grief can also influence the process of establishing effective attachments: Solnit and Stark (1961) comment on the "...helplessness and sense of failure" seen in such parents. Grieving and attachment processes having to occur simultaneously, usually over several years (and sometimes decades) and both processes can be exacerbated by the predictable developmental crisis points of everyday life such as starting school, for example. Additionally, many of these children experience repeated separations and stresses resulting from co-existent physical illnesses (which they are far more prone to compared to non-retarded children) and hospitalizations. It is very
probable that child health professionals are also vulnerable to become subconsciously caught up in these powerful emotional states which operate within many of these families. If unrecognised, such feelings may cloud professional judgement and lead to inappropriate action such as over investigation or the pursuit of "miracle" cures rather than the necessary exploration and promotion of greater understanding and coping skills. This may also help to explain, at least in part, previous attempts of professionals to try to put unrealistic pressure on such families to encourage their children to achieve developmental milestones at a near "normal" time. Additionally, this form of professional psychological enmeshment with these families and denial of their children's developmental needs may also help to understand the calls for the uncritical and global "normalisation" of life for all such children by certain groups of child health, care and welfare professionals. Hodapp and Zigler (1989) warned of the probable deleterious consequences of these pressures for such children's fragile emergent sense of autonomy.

The retarded child's intrinsic passivity which has already alluded to usually, but not invariably, results in a distortion in the relationship with the caregiver. The mother (and it is usually the mother) necessarily becomes much more directive within the mother-child interactions and this directiveness has implications for the child's intrapsychic development: the severely retarded child is usually unable to incorporate aspects of significant others into themselves, and then psychologically individuate successfully. Levitas and Gilson suggest that because this separation cannot occur in retarded children in a normal way: they incorporate the caregiver as an ever-present "executive" ego. This situation is maintained by the principal caregiver's assertive, directive and actually continuing presence in the child's life. The caregiver, figuratively, becomes both a lens and filter for such children in their perception and relationship with the outside world;
thus, in being so, remain crucial for their children's continuing survival. This ongoing vital intimacy only stops when desertion, severe illness or death intervenes. The power of the relationship is reflected in the prolonged and often pathological grief these individuals demonstrate when they eventually lose their principal carers.

The quality of this state of intimacy depends upon the mother both perceiving and responding to signals from her child at a pace and in a manner which is adaptive and follows their respective levels of development within the growing parent-child relationship. When this does not occur anxiety is a common consequence. When this is intense or too sudden, severe distress occurs and this precise factor of communication inadequacy was shown to be strongly implicated in the maintenance of severe NS/NW sleep problems in Quine and Pahl's cohort of severely retarded children (1989). This flaw in the relationship can trigger the mother to assuage her child's distress habitually in order to prevent strong, and commonly unresolved, feelings of failure, guilt and grief reemerging in herself. If the mother and child relationship is already compromised by insecure attachments and extreme carer stress this can serve to magnify the feelings engendered by the child's distress and lead to excessive soothing of the child and, also, the carer's avoidance of potentially distressing stimulations or situations. A cyclical, self-reinforcing interaction involving handicapping overprotection is thus established.

MacKeith (1973) comments upon the "lavish caring" often seen in such families and the important aetiological role this can have in their children's behavioural problems. Here is where the NS and NW sleep problems which are the focus of this study come in: if ever there was a scenario where comfortable and controlled early separations between parents and their children need to be reenacted routinely in everyday life it is at bedtime and during the night. As has been described previously, any child who is sleepy and tired
is also more likely to be less self-controlled and, consequently, less controllable by others. Mothers who are stressed and sleep-deprived are less likely to control night-time behaviour in a calm and consistent manner. If this occurs within the context of ongoing attachment difficulties which themselves evoke strong anxieties within the mother then she is far less likely to tolerate the child's fussing and cries for attention during the night. On the child's part he remains stuck at a stage of requiring immediate solace for his distress with delays in receiving this resulting in increasing distress and demands. This situation may be further compounded by the transient disruption of sleep commonly encountered in children returning from hospital where the child tends to demand even more of the reassurance of his parents' presence and, for their part, the parents are more likely to overcompensate anxiously for this by giving even more comfort. At the upper ability range of the older severely retarded group they are capable of "blaming" their parents for sending them to hospital and thereby adding to their parents' sense of guilt (Moore, M.-S., 1989). Thus when sleep disturbance is present in a child there also exists a mutual difficulty for the child and his principal carers (Daws, 1991). For the parents of severely learning disabled children this difficulty is often compounded by other potent and often unacknowledged anxieties relating to feelings of grief, loss, failure and anger. Recognising these issues may engender pessimism in therapists when it comes to treating such problems. It is therefore reassuring that treatment success appears to be very much the rule rather than the exception when it comes to helping such families.

(d) Summary
The various developmental perspectives of NS/NW problems presented in this section are probably not mutually exclusive of one another and several of these theoretical views might be applicable in a given case. It must be borne in mind that these factors may also be the result of a severe sleep problem rather than its cause. For example, effective limit-setting at night-time may be extremely difficult when the parents themselves are chronically sleep deprived and thus exhausted. In the opinion of the author it is probably most helpful to conceptualise NS/NW sleep problems as representing a final common pathway of a range of possible extrinsic and intrinsic aetiological factors. A comprehensive clinical history will reveal these and such influences may have to be addressed during treatment. It is also the author’s view that NS/NW problems represent the prototypical behavioural problem seen in children in developed societies: the establishment of a satisfactory sleep habit is necessary for optimal daytime performance and, of equal importance, is that this represents the first major step infants have to make in order to achieve adequate self-control and, ultimately, degrees of independence from their principal caregivers. When this is not achieved to a satisfactory degree a series of internal and external developmental decompensations may ensue which can contribute to later interpersonal and psychological dysfunction manifesting as a range of behavioural and emotional problems as the children develop. In their large population prospective survey Butler and Golding discovered that sleep problems in infancy predicted feeding, aggressive behaviour and other behavioural problems at school age (Butler & Golding, 1986). A recent study of chronic sleep problems (night-walking) in normal five-year olds predicted a constellation of emotional difficulties five years later (Pollock, 1994). Unfortunately none of these workers elaborated theories or pathways of causality for these associations. As has been stated previously, the influence of severe and chronic sleep
problems operating upon the developmental potentials and trajectories of severely retarded children can only be guessed at present. Nevertheless it is possible to predict on a priori grounds that prevention or early detection and treatment of these problems may well help to optimise such children's prospects.

1.2 TREATMENT OF NS/NW PROBLEMS

1.2.1. Behavioural Treatment

Despite the fact that sleep problems in childhood have been recognised for several decades and several eminent child care specialists have discussed their treatment (Spock, 1950; Anders, 1985; Ferber, 1985) it is therefore surprising to discover that there have been very few treatment studies involving sleep disordered children. Nevertheless, the results of the few which have been conducted support the clinical view that they are amenable to straightforward interventions which, for the most part, involve behavioural modification techniques delivered by the children's parents who act as co-therapists with the clinician or principal therapist. The behavioural modification approach to childhood NS/NW problems can take several forms however the most frequently practised (and researched) technique involves inter alia a negotiated gradual withdrawal of parental attention from the child at bedtime and offering developmentally appropriate rewards to the children for appropriate bedtime behaviour. This "graded changes", gradual extinction, approach assumes that parental attention is the most important reinforcing factor for the childrens' sleep problem. Treatment effectiveness being commonly assessed by comparing outcome
measures with, often lengthy, baseline measures a process which is probably somewhat removed from everyday clinical practice where parents and carers are often desperate to receive rapidly effective help. Overall success rates for the behavioural treatment approach to NS/NW problems in clinical studies have been shown to be high: Richman (1985): 77%; Seymor (1983): 78%; Jones and Verdyun (1983): 84%; Minde et al (1993): 83%. Unfortunately, the criteria employed to establish treatment success in these studies varied and also tended only to concentrate upon symptom reduction rather than overall consumer satisfaction. A study which compared treatment outcome of health visitors using specific behavioural modification techniques with those offering only general advice and support did not show any advantage (Weir & Dinnick, 1988); however, more recently, a far larger retrospective audit of a primary care sleep clinic run by health visitors reported generally excellent results in terms of target symptom reduction and also high service-user satisfaction (Boomer & Deakin, 1991).

1.2.2. Treatment Studies in NS/NW Learning Disabled Children

Until very recently the literature concerning treatment studies of sleep disordered children with significant general learning difficulties rested primarily on individual case studies; for example, Howlin (1984) described the successful behavioural management of a severely sleep disordered six year-old autistic boy. However, larger scale studies involving cohorts of affected children have been conducted over the last decade. Quine and Wade’s (1991) intervention study involving twenty-five sleep disordered severely learning disabled children had an overall success rate of 80%, furthermore, they demonstrated that the children’s parents were highly satisfied with the approach used.
This evidence supports the clinical impression that learning disability \textit{per se} is not a barrier to treatment success. Several other groups of workers have shown that parents of learning disabled children particularly can be helped by child health professionals to teach their children to adopt more satisfactory behaviour patterns by employing straightforward behavioural modification approaches (Cunningham \textit{et al}, 1986; Yule, 1975; Gath, 1979) and the various strategies employed with NS/NW problems are described below. Quine and Wade's study (1991) employed standard behavioural techniques again within a framework of negotiated gradual (or "graded") changes in which the parent(s) withdrew gradually from the child's bedroom and respond to their children's demands once settled, again, in a graded, attention-reducing, manner. This treatment study was prompted by a previously conducted survey (see above) which revealed high rates of chronic sleep problems in this group and also significantly associated factors, such as high levels of maternal stress. In this elegant study, the researchers trained a group of health visitors during a three-day workshop to both assess and also treat a selected cohort of NS/NW sleep disordered children. Following this, each health visitor was allocated two families with affected children and treatment was started after several weeks' worth of baseline data were obtained. The results reflected that the intervention was successful in most of the families and that there were significant changes in the families and children's wellbeing over and beyond those attributable to the treatment advice given. This was manifested in terms of improvements in the childrens' daytime behaviour and also reduced levels of stress and improved coping skills were reported by the childrens' parents. The average contact time between the therapists (health visitors) and the families was several weeks and, in some cases, several months (Quine, personal communication). An earlier study by Hewitt (1985) was similarly
successful although he himself was the only therapist involved in the treatment of his study cohort of ten learning disabled children, eight of whom were cured by his intervention. Again, the graded-changes approach employed required several weeks' worth of patient therapist contact. In Rickert and Johnson's treatment study (1988) which compared a progressive extinction approach involving training affected children's parents to ignore their children's demands at bedtime "cry it out") to scheduled awakening (briefly rousing the NW children before they usually tend to awaken during the night) a few of their children in addition to their sleep problems had learning difficulties and it was shown that these children were as responsive to the systematic ignoring techniques as the non-retarded children were. This study which resulted in successful outcomes for both types of intervention provides the only example within the literature of treatment studies which has evaluated short-contact, focused therapeutic techniques which also included learning disabled children. Given the high prevalence of sleep problems in this group this is surprising. It has been the author's clinical experience and also that of senior colleagues who have an interest in children with learning disabilities suggests that focused, rapid-settling techniques are highly effective in most sleep disordered children irrespective of the presence of any co-existent developmental delays.

1.2.3. "Gradual" versus "Rapid" Extinction Approaches

Anxieties expressed by Bax (1981) concerning the possibly dangerous consequences of "...training a parent to ignore their child" through using rapid extinction techniques with
NS and NW problems have not been supported by any reports concerning this clinical practice nor by any treatment studies. Indeed, an often observed maintaining (and perhaps aetiological) factor concerning NS/NW problems is that of inappropriate parental overresponsiveness to their children during bedtime (Blurton-Jones, 1981; Adair et al., 1991). More recently, in a review of the field of childhood sleep disorders by Skuse (1993), expressed the belief that approaches aimed at rapid extinction of the NS/NW sleep problems by settling the child quickly and thereafter ignoring the child's demands for attention are "...rarely practicable" pointing out that the necessary distress involved with this technique would prove unacceptable to parents (and therapists? -see above). In her therapeutic guide for the parents of sleep disordered infants, Elizabeth (1988) warned of several possible side-effects of extinction techniques such as the induction of states of fearfulness and feelings of rejection and insecurity in affected children yet without giving empirical evidence in support her contentions. Unfortunately, these beliefs concerning the problems of rapid extinction techniques, appear to be held by many child therapists, yet do not appear to be supported by any evidence and one of the stated aims of this thesis is to investigate the acceptability (and safety) of this type of treatment approach. In his small cohort study, France observed behavioural and emotional improvements with rapid extinction techniques when they were used with sleep disturbed non-retarded infants (France, 1992) However, an example of where extinction methods can indeed fail predictably is when the parents eventually "give in" to their children's extreme demands for attention and, in doing so, paradoxically reinforce the target behaviour; a process made even worse if this happens repeatedly and intermittently. On the other hand, gradual extinction or "graded changes" approaches while often successful in carefully conducted clinical trials such as Quine and Wade's study (see above), in everyday clinical
practice run the risk of losing momentum over a protracted treatment period and also, unless the therapist remains vigilant, children can readily and suddenly increase or alter their attention-seeking strategies at any time during this period and thus catch vulnerable parents or carers unawares, and thus pushing back the likelihood of success or, indeed, wrecking the plan entirely. This latter phenomenon tends to be more common when treating older and more able children. Lawton et al. (1991) discovered that "graded changes" approaches can also be stressful for infants' parents and particularly when "extinction bursts" of acute behavioural deterioration occur during treatment. Intermittent countertherapeutic reinforcement of the target behaviour can also occur in this way (Skinner, 1953). In the specific case of severely retarded children other factors such as intercurrent physical illness or disability or the presence of other severe behavioural problems are more likely to undermine any protracted programme. Factors relating to therapist contact, continuity of treatment and also consistency of approach style are additional sources of potential difficulty which can contribute to failure with this technique. Therefore, theoretically, an intensive and focused time-limited approach such as a rapid extinction technique should reduce the possibility of these confounding factors operating and thus increasing the possibility of therapeutic success and thus may represent the best therapeutic option for the sleep disordered severely learning disabled child.

To date, the behavioural modification treatment literature concerning childhood NS/NW problems has been dominated by "graded changes" approaches which, as has been discussed, while they have been demonstrated to be highly effective in carefully conducted studies, have not been shown to be as cost-effective as other, more focused and time-limited, approaches. The few studies which have compared approaches have not revealed any therapeutic advantages to using lengthy (often lasting many months) techniques
indeed one such study suggested an advantage in using time limited-techniques including the use of intensive telephone follow-up reviews (Rickert & Johnson, 1988). Boomer and Deakin (1991) also discovered advantages for time-limited intervention in their study. Like most other branches of medicine, child psychiatry employs a wide variety of therapeutic techniques, however few of these have been tested scientifically in properly conducted trials to support claims of their efficacy or superiority over other techniques. Examples of this in child psychiatry are the ongoing debate concerning the efficacy of "pure" forms of family therapy over more eclectic, short-term approaches to disturbed families presenting with given problems as is the precise role of antidepressant medication in the treatment of depressed prepubertal children. This situation is changing and, indeed, needs to change in this era of clinical audit and a growing awareness of patients' rights to receive effective treatment. Another important factor is the availability of improved techniques for measuring change in complex clinical situations.

Primary prevention of such problems is an area which has until only recently received scant attention. Parents of all children should be sensibly advised about the need for their children to learn healthy sleep habits before problems develop ideally this should occur at an early stage. Hewitt and Galbraith (1987) have described and tested an effective scheme which addresses this particular need, however their impressive results have yet to be replicated by other workers in this field.

1.2.4. Drug Treatment

No review of the field of childhood NS/NW disorders would be complete without reference to drug treatment. Nevertheless, although widely practised by the non-specialist,
pharmacotherapy has only a small role to play in the treatment of paediatric sleep disorders. Clinical experience suggests, however, that sedative drugs, particularly the antihistamine phenothiazines (trimeprazine tartrate and promethazine hydrochloride) and chloral hydrate, remain the mainstay of treatment for night-time behavioural problems despite there being no evidence for their efficacy in chronic and severe cases or, indeed, their safety (Anders & Keener, 1983; Biban et al., 1993). In a recent American community survey of a thousand pre-adolescents, 4% of the 132 habitually "poor" sleepers received sedative medication regularly (Kahn et al., 1989). Another American worker, Johnson (1991) discovered a point prevalence rate for sedative prescription in children of 6% and also a paradoxically high level of parental disapproval concerning this practice in her community survey which was conducted by telephone interview. Only in a small number of very specific clinical situations has drug treatment been proven to be effective: for example the use of antidepressants and psychostimulants in narcolepsy (Parkes, 1985). Other specific physical treatments, although gaining clinical popularity have yet to be subjected to scientific trials (for example, the use of tonsillectomy in children with symptoms of obstructive sleep apnoea- children with Down’s Syndrome appear to be particularly prone to this condition). Anticonvulsant medication can prove effective in controlling symptomatic (i.e. sleep disrupting) nocturnal forms of epilepsy and prophylactic antiasthmatic drugs similarly for nocturnal asthma. Chronic infestation with threadworms (Enterobius vermicularis) which is not uncommonly encountered in severely retarded children because of their poor personal hygiene skills, can cause intense nocturnal pruritus ani which can severely disturb sleep. This condition is easy to diagnose and can be treated effectively with mebendazole (Farthing & Rolston, 1990).
Proof of the long-term efficacy of medication has not been demonstrated for childhood NS/NW problems. In a double-blind trial of trimiprazine tartrate in 22 non-retarded toddlers, Richman (1985) demonstrated that sedative medication offered, at best, only temporary respite for affected families and then only for a few responsive children. At six month follow-up of 14 of the children there was no evidence of any lasting effects of sedative treatment. However, as Richman acknowledges, her study was flawed methodologically in that there were no checks that the children were actually taking the medication neither was the accuracy of the parents' diary records of their children's sleep records confirmed independently. In another controlled trial of the same agent with a small and highly selected cohort of night-walking non-retarded children, Simonoff and Stores (1987) demonstrated a significant, albeit transient, positive drug effect; nevertheless, the authors urged caution at the blanket use of such medication suggesting that it is best reserved as a temporary respite measure rather than a "cure" for persistent NS/NW problems. More worrying are occasional reports paradoxical reaction to sedatives which children may show, which involves their exhibiting extreme hyperarousal and behavioural disturbance with consequent increased distress for their families (Valman, 1981). In his recent review of drug treatment in child psychiatry, Prendergast (1993) strongly recommends using behavioural treatment for NS/NW problems in preference to sedative drugs pointing out recent concerns about the risk of children developing tardive dyskinesia when taking trimiprazine (which is related to the phenothiazines) over long periods. The author's clinical experience suggests that children with severe learning disabilities might be particularly vulnerable to paradoxical reactions to sedative medication when it is used in this context (see "Results" section). The most extreme example of pharmacological mismanagement of severe NS and NW problems in a child with severe
learning disabilities encountered by the author was that of a 12 year-old girl who had been receiving a hundred milligrammes of trimeprazine tartrate (a dose recommended for surgical premedication) for eleven years. Despite this, her sleep problems had persisted; furthermore she was noticeably oversedated in the mornings and, in being so, most prone to be physically aggressive at this time. Her daytime behaviour improved considerably once this drug had been stopped and her longstanding sleep problems responded quickly to the rapid extinction technique tested in this thesis. It is not widely appreciated that the British National Formulary does not recommend to use of sedatives for children (British National Formulary, 1994) and the product information sheets for these agents state that they are specifically contraindicated for young children (Association of the British Pharmaceutical Industry, 1994). Given that the prescription of a sedative agent remains the first, and often the only, treatment approach for NS and NW problems employed by primary care and paediatric clinicians, this practice further serves to highlight the general deficiencies concerning training in the assessment and management of common childhood sleep problems.

1.3. ASSESSMENT OF NS/NW PROBLEMS

1.3.1 Diary Monitoring of Sleep Disorders
Throughout the treatment literature of NS and NW sleep problems there is testimony to the value of persuading parents to keep diary records of their children's sleep patterns and that this simple measure can sometimes, by itself, result in the amelioration, or even the disappearance, of the sleep problem (Largo & Hunziker, 1984; Boomer & Deakin, 1991). There are various forms that such a diary record may take but they all have in common certain features such as requesting data concerning the time the child is put to bed, night-settling times and waking times, for example. Additionally, they usually require that parents note down their reactions to their children's behaviour and what resulted from this. In effect, they undertake a simple functional analysis of the problem. Performing this exercise, in itself, is probably an important explanation of the reported success of diary monitoring in these circumstances. Nevertheless, it is well recognised that parents' abilities to keep such records vary enormously; families which are characterised by high levels of familial disruption and also those with low parental literacy skills can preclude the effective use of this means of monitoring. Even when diary keeping is possible and complete data sets are available, the reliability of such data is influenced by several factors. Simonoff and Stores (1987) reported good reliability of parent reports compared to ambulatory polysomnographic data; however, these parents comprised a highly selected group within their study cohort. In a much larger study of children's general behavioural patterns, Griest et al (1980) discovered marked discrepancies between mothers' reports of their children's behavioural difficulties and those of independent observers: stressed and depressed mothers particularly tended to overestimate these problems compared to non-stressed mothers. The reliability of sleep diaries is therefore probably similarly suspect given that stressed parents are the rule rather than the exception in cases of severely sleep disordered retarded children (as has been discussed elsewhere). Andersen, Keenan and
Carson (1989) compared activity monitor data obtained from 30 children with parents' diary assessments of their children's rest-activity patterns: a low \( r (28) = .42 \) but only just significant \( (p < .05) \) correlation between the two measures was obtained. Thus a degree of caution is required when interpreting sleep diary data particularly in treatment studies which explicitly require that parents ignore their children, once settled in bed, as will be the case with the treatment approach examined in this thesis.

1.3.2. Activity Monitoring

(a) Movement in Sleep

Along with its predictably varying degrees of unconsciousness sleep, from a behavioural perspective, is also characterised by relative and absolute periods of whole body inactivity. The exact points at which sleep begins and ends are not known: sleep involves a process of diminishing consciousness and reduced awareness of the environment which occurs over a (usually) brief period not exceeding a few minutes. Prerequisites for sleep onset is a prior state of quiet wakefulness around the time when the intrinsic circadian sleep/wake cycle, controlled by the suprachiasmatic nucleus of the hypothalamus, tips towards sleep promotion. With sleep onset both body movement frequency and intensity further reduce and continue to do so as sleep deepens. Sudden, brief jerks, or "hypnic shakes", are sometimes experienced over this period. "Deepest" (NREM stages 3 and 4) sleep shows long periods of little movement other than those associated with shifts in posture or a few brief gross extensive movements around the periods of shift from deeper to lighter sleep which occur at around ninety minute intervals from sleep onset throughout
the night (Lugaresi et al, 1988). REM, or "paradoxical", sleep is associated with paralysis of skeletal musculature with loss of tendon reflexes which precludes whole body movements (which is just as well because it prevents unpleasant dreams being enacted physically). REM sleep does, however, allow muscular micromovements and these were first detected by means of wrist placed EMG electrodes in a study conducted by Wolpert (1960). Thus it can be seen that normally the sleep period consists of little or no movement compared to that seen in wakefulness; what little movement occurs tends to be concentrated around the periods corresponding to the cyclical changes from "deep" to "light" (stages 1, 2 and REM) sleep. This adult-type pattern of sleep movement is usually established in children by the end of their first year of life irrespective of any degree of learning disability (given that subjects' motor systems are both intact and functional). When this is not the case, when movement is detected to a significant degree at nighttime, this usually (but not invariably) the result of subjects waking up and mobilising. The most important exception to this is with children (and some adults) who experience repeated sleep-walking and/or night terror parasomnic episodes. Other rarer movement problems are associated with the nocturnal epilepsies and other neurological conditions such as childhood periodic limb movements. Even when these non-waking conditions are severe their impact on sleep in terms of the amplitude and duration of movement involved is usually very small compared to the movement levels associated with wakefulness. Thus the investigation of certain movement patterns during sleep can yield objective evidence about certain sleep parameters. The various analyses of the activity monitor data obtained from some of the children in this study will attempt to do this, the precise details are presented in the "Method" section of this thesis.
(b) Ambulatory Activity Monitoring

Activity monitoring (AM) employed as a research tool applied to physiological measurement has a history which spans nearly seventy-five years. The first significant contribution to the subject was by a Viennese psychiatrist, Szymanski (1918) who obtained systematic mechanical analogue data from man and other higher mammals. The prototypical devices worked on mechanical principles similar to those found in self-winding watches and pedometers; modern devices employ solid state ambulant microcomputers which measure movement electronically by means of accelerometric or piezoelectric transducers. The increasing technical sophistication of these devices and also, importantly, the research paradigms in which they have been used places them in the forefront of objective techniques for measuring physical movement in both normal and pathological states.

This review will concentrate on the use of AM in sleep disorders generally and, more specifically, those commonly found in children. However, it must be emphasised that AM has been used in the investigation of a wide range of conditions which feature abnormalities (or observable changes) of movement (neurological conditions: Parkinson's Disease, epilepsy, hyperkinesis, spasticity, chorea, dystonias and rigidity; cardiorespiratory disorders: obstructive sleep apnoea; psychiatric disorders: anorexia nervosa and depression; rehabilitation of the chronically disabled; pharmacological research, especially the development of tranquillising and sedative agents; human performance studies; human metabolism; animal research (Tryon, 1991)).
From Szymanski's earliest investigations of movement in sleep, the field of sleep and its disorders has provided fertile ground for the application of various AM techniques and has yielded valuable information. In recent years the pace of research has accelerated. Sleep is currently conceptualised as a dynamic, actively regulated behavioural state with associated predictable physiological correlates. Neurophysiological changes during this state provide the basis for staging and thus classifying the different levels of sleep (Rechtschaffen & Kales, 1968). As has been discussed previously, sleep is primarily recognisable by a relative or absolute absence of whole body movement when compared to activity patterns characterising wakefulness. Activity monitoring is therefore well-placed to measure these specific features of sleep. Unfortunately, the technique cannot provide detailed information concerning sleep staging: polysomnography which involves parallel electroencephalographic, electromyographic, oximetric and other techniques remains the investigation technique of choice for this. Nevertheless, AM has been found to be a sensitive means of detecting the microarousals associated with certain specific sleep disorders (obstructive sleep apnoea and narcolepsy, for example) which polysomnography can sometimes miss (Horne, personal communication). AM has been shown to be reasonably accurate in assessing sleep onset and total sleep time in young children and infants when compared to polysomnographic data. The margin of error between the two systems is approximately 10% (Sadeh et al, 1989). One of the major problems in this area is the lack of any generally agreed algorithms with which to define sleep onset or, similarly, points of wakefulness following sleep even when polysomnographic measures are used. Part of an explanation for this is that these transitional states are dynamic and operate over several minutes: there is no precise points
of transition of sleep to wakefulness and vice versa. By definition AM only measures movement and AM systems' sensitivity to movement vary from device to device and also depend on anatomical monitor placement. However, when these monitors are used in sleep studies the general rule which applies is that whichever criteria are employed to assess these parameters in a given study they need to remain constant during that study and are required to be explicitly stated by the investigators. Sadeh et al. (1989) developed a scheme for computerised automatic scoring of sleep and wakefulness onset which compared very favourably with polysomnographic assessments of these parameters but this technique has not yet been applied to a treatment study setting. As has been discussed, AM is far less accurate at differentiating between REM and non-REM sleep stages; polysomnography remains the best means of obtaining this information. Given these reservations, AM represents a useful means of assessing sleep and its disorders from another perspective. The technique has several advantages over traditional polysomnography. These include:

(i) Monitors are portable and generally well-tolerated by adult subjects. Modern devices such as the Gaehwiler Electronic monitor (used in this study) are roughly the size of a man’s wrist watch and being approximately double the weight.

(ii) They can be worn on the wrist, the upper arm or leg; placement is determined by both the type of movement parameter being measured and also the type of subject. For example, infants and toddlers' sleep movement patterns would be best monitored by placing the devices on the upper arm whereas an elderly person with periodic leg movements best assessed with leg placement. Unfortunately normative data concerning the various monitor placements is not currently available.

(iii) Software development allows modern devices to start and stop monitoring away from the laboratory or clinic for a variable time depending on whichever
sleep movement parameters are being investigated and also according to study design. This allows minimal "first night" effects which delays sleep laboratory work (acclimitisation to the setting) because many studies can be conducted in the patient's normal domestic sleep environment. Even in the sleep laboratory wearing a monitor is far less uncomfortable than EEG, EMG and EOG electrodes. Thus the data so obtained is far more "naturalistic", representing the subject's usual sleep patterns. Depending on the memory capacity and software specifications of the instrument employed several nights' or even weeks' worth of data can be obtained without requiring the subject to return to the clinic. A cost/benefit analysis of AM as compared to sleep laboratory polysomnology would be probably greatly in favour of AM when simple sleep movement-related parameters are being studied.

(iv) Given the variable reliability of traditional sleep diary reports which has been discussed previously, the AM data can provide accurate "real time" data of the subject's sleep patterns which is not influenced by major interpretive biases (although care must be applied to detect monitoring artifacts and periods when the monitor is either deliberately (whist the child is being bathed or is swimming, for example) or inadvertently not attached to the child by the parent. Therefore, in order to avoid such confounding events parental diary records should be obtained in addition to any AM-derived data.

(d) AM and Childhood Sleep Disorders

There have only been a few research projects conducted in the field of childhood sleep disorder research which have employed AM devices to assess treatment effects and there have been no large-scale studies with non-disordered children to provide robust normative
data. Sadeh and his colleagues (1992) used AM in their study which compared sleep disordered infants to non-disordered controls and discovered that computerised analysis of the AM data sets alone allowed accurate (80-90%) discrimination between the two groups. Other interesting results from this study were that there was low within subject stability for night-waking children compared to controls; also, the sleep efficiency (that is the time asleep divided by the time in bed) between the two groups differed significantly. A criticism of this study was that although there were nearly a hundred children in the study they were each monitored for an average of only four nights which probably explains low-within subject stability for some of the sleep criteria. Additionally, "night-of-the-week" effects which can influence night-waking frequencies (see "Results" section) were not considered in their analyses. Nevertheless, their work is the most exhaustive to date concerning this particular application of AM. Sadeh's group go on to speculate that AM could well prove to be the assessment method of choice in research or even clinical work in the future; however, it is more likely that AM and polysomnographic data collection will seen to complement each other, each technique having advantages in specific areas. When used together, for example in the assessment of obstructive sleep apnoea, their combined data may offer more information than that obtained by using either technique alone.

To date, AM techniques have not been used with children in a treatment study prior to the interventional study described in this thesis.

1.3.3. Other Methods of Objective Assessment of Sleep Behavioural Problems

(a) Polysomnography
Combined ambulatory monitoring of brain, muscle, heart and eye movement activity (EEG, EMG, ECG and EOG, respectively) and also oximetry is now widely used in clinical research; the multichannel recording technique is referred to as "polysomnography". In severe sleep behavioural problems the technique can detect underlying or contributory medical conditions such as epilepsy or obstructive sleep apnoea but the vulnerability of even the latest devices to damage at the hands of disturbed retarded children is high. Additionally, the tape cassettes need to be changed every day, requiring regular contacts between affected families and investigators which might, in itself, affect the target behaviour. Interpretation of the data so obtained can be time consuming, however computerised analysis has recently greatly improved this.

(b) Television monitoring

A recent study by Minde et al (1993) investigated response to a behavioural treatment by means of over-night time-lapse infra-red videotaping of the child's sleep pattern. This yielded important information concerning the sleep patterns of their children and the patient controls: the treated children slept for an hour less than their controls, on average. They also discovered that even their controls were waking almost as frequently as their sleep disordered subjects, however, they usually settled within a few minutes without being distressed. Elias et al (1986) discovered acceptable degrees of reliability ($r = .7$) when they compared videotape and sleep diary records of infant sleep/wake activity. Home videotaping is undoubtedly an accurate means of assessing a subject's sleep pattern however there are certain practical drawbacks to this method: (i) it requires at least a one-night acclimatisation period whereby the subject gets used to the presence of the camera and its attendant noise and lighting; (ii) the videotape has to be replaced every night and
any faults (focus, orientation, damage, etc) attended to; (iii) the videotape may have to be checked after every session to detect any faults which would require researcher contact with the families which might contaminate studies taking a strongly "naturalistic" approach; the most important drawback is the large amount of time required to view the tape and code data. Kligman et al (1975) were the first researchers to comment upon the intrusiveness of home video monitoring in their investigation of infants' sleep patterns.

1.4. INTRODUCTION TO THE INTERVENTIONAL STUDY

The quasi-experimental clinical treatment study which is the focus of this thesis arose from the author's general interest in the psychiatric care of children with marked learning difficulties and their families. More particularly, it arose from a recognition that many of these children were being referred to child psychiatric services with chronic, severe and usually combined NS and NW problems which were causing their families great distress and difficulties. Commonly, the children's parents admitted to being severely stressed, sometimes expressing explicit fears concerning their own mental health and capacities to continue to care for their children. Occasionally, and more worryingly, some feared for the safety of their children at their hands should the sleep problems persist. Characteristically, many families reported having sought advice previously from many sources: family and friends, primary care staff, paediatricians or even other child psychiatrists but finding their advice ultimately of little or no benefit (see "Results" and "Discussion" sections). Despite these often dramatic presentations, of chronically sleep-deprived and exhausted parents and children who are out of their control at bedtimes and often also during the day, a straightforward therapeutic approach consisting of several behavioural modification techniques (see above and, more specifically, the "Method"
section) aimed at rapidly establishing positive bedtime routines and firm bedtime limit-setting by the parents involving concentrated initial daily therapist contacts (usually by telephone), usually proved to be successful within a few days notwithstanding the prior duration of the sleep problem. Furthermore, results were commonly sustained at follow-up appointments and positive improvements in other aspects of the children's behaviour were seen without their having been addressed specifically. When this form of treatment was employed the usual contact time between families and clinicians (which included an initial assessment and treatment combined session) was seldom longer than two or three hours. When compared to "graded changes" approaches (see above) this technique appeared to provide a considerably more efficient (and also more cost-effective) means of dealing with such common problems. Despite the usually predictable, although in no means invariable, minor distress this approach causes children during the initial phase of treatment, both parents and, sometimes, the children (who were capable of expressing an opinion on the matter) were usually highly satisfied with the results. In the minority of cases when this approach did not work which accounted for around one in ten families so treated, the family dynamics which prevented this (for example, the various reasons why some parents found it impossible to separate from their children at bedtime) became very evident and were addressed accordingly. Thus this treatment approach could also yield important diagnostic data concerning refractory families' functional dynamics. In such cases other treatment approaches such as scheduled awakening, negotiated "graded changes" or formal family therapy would be used as indicated. Generalization effects from this treatment were commonly observed: effective limit-setting by the carers at night time would also occur during the day. As a consequence of obtaining regular undisturbed sleep, often for the first time in several years, parents were generally more alert and thus more capable of meeting their children's demands during the day. The children
themselves also appeared to benefit in that they would often become more settled during the daytime, presumably being unhindered by tiredness and sleepiness following an undisturbed and settled night's sleep and also becoming more responsive to the limits set by their carers. Anecdotal reports of children showing improvements in various adaptive behavioural or cognitive skill domains were sometimes related by parents after such a treatment programme had finished.

Over a year's training attachment to an adult learning disability psychiatric service the author also discovered several severely retarded young adult patients living at home who, according to their parents, had also exhibited severe NS/NW problems throughout their lives in addition to range of chronic and severe "challenging" daytime behaviours. This led one to speculate about how these intense problems impacted upon their families and also how they contributed to these individuals' patent failure to achieve even their restricted developmental potentials. Interestingly, once identified, these patients and their families also benefitted from the general behavioural modification approach previously described which needed to be slightly adapted to take into account the specific needs of adult patients (see "Discussion" section).

A review of the available literature revealed that this type of clinical problem and its focused treatment had not attracted a great deal of systematic enquiry (see above) therefore this study was devised in order to meet this deficiency. This study is the first to investigate the efficacy of a brief behavioural modification approach which is based principally upon the behavioural therapy principles of extinction and stimulus control in a mixed cohort of severely learning disabled sleep disordered children. Also, for the first time in a treatment research paradigm, some of the childrens' sleep patterns were monitored electronically over the course of the study which, as will be seen, yielded objective information about the effects of treatment as well as about other aspects of their
sleep. Analysis of the data obtained in this way required the elaboration of several novel concepts in respect of the assessment of night-waking episodes. Of necessity, certain minor adaptations of the precise form of the intervention had to be made in order to comply with the constraints of a research paradigm which would allow any meaningful interpretation of any results. Nevertheless, every attempt was made to keep the treatment approach as close to that used in normal clinical practice in order to conduct, in effect, an audit of this behaviour modification technique in the hope that its results would be of direct relevance to clinical practice in this area.
1.5. HYPOTHESES TESTED IN THE THESIS

Four principal hypotheses are tested in this thesis. These are listed below:-

1. Night-settling and night-waking problems presenting in severely learning disabled children can be treated effectively by a focused brief behavioural modification approach.

2. There are beneficial generalization effects demonstrable in both the children and their principal carers as a result of treatment in parameters not explicitly addressed by the treatment.

3. The approach is acceptable to the principal carers of such children.

4. Ambulatory electronic activity monitors can yield useful objective data concerning changes in the children's sleep patterns over the course of the study.
2: METHOD
2.0 OVERVIEW OF METHOD SECTION

This section will begin with a description of the selection criteria employed and means of recruiting suitable subjects for the study; this is followed by a description of the treatment advice package and the rationale for choosing it; study design is then presented followed by a description of the study's various outcome measures and how, in turn, they are used to test the four principal hypotheses underpinning the study. The section closes with a description of the various means of analysis of data which are used to interpret and present the data in the following "Results" section.

2.1. COHORT SELECTION

2.1.1. Selection Criteria

There follows a concise description of the selection criteria used in this study to identify suitable sleep disordered subjects:

(i) A severely learning disabled child whose primary behavioural problem is a severe and chronic night-waking and / or night-settling problem (by both parent and investigator assessment criteria - see below). The child may also have other behavioural problems.

(ii) There is no evidence of any form of abuse or neglect of the child by his or her carers.

(iii) The child's parents or principal carers are adjudged to be capable of both
understanding the treatment programme and also of carrying out the tasks required of them.

(iv) There is no co-existent extreme marital disharmony and the relationship between carers is reasonably stable. As far as is practicable, there should also be no major upheavals facing the routine of the household (major holidays, starting school, etc.).

(v) The child should be in good physical health or any chronic physical illnesses such as epilepsy or asthma should be well controlled by medication.

(vi) The child should not be receiving any other conventional or unconventional forms of treatment for the sleep problem and this is particularly the case for sedative medication.

Fifteen children fulfilling these criteria were sought from a range of referral sources: specialist health visitors, paediatricians and psychiatric colleagues. Referrals to all of the Leicestershire Child and Adolescent Psychiatry teams were scrutinised for suitability. Once identified, subjects were then screened at their homes or in clinic and, if they proved suitable, consent obtained from their parents or principal carers. Copies of the recruitment letters and consent form are presented in the "Appendix" section.

2.2 INTERVENTION TECHNIQUE

2.2.1. Behaviour Modification Programme

Having established a trusting relationship with the affected families, the primary aims of the intervention were to help children settle quickly at night and also to learn not to demand attention from their parents should they awaken during the night. The behaviour
modification approach employed in the study consisted of the following components and is based upon the scheme suggested by Pearce (1991):-

(i) The parents are asked to set a regular bedtime for the child and stick to it.

(ii) A consistent "winding down" routine is devised for the hour before bed-time consisting of the promotion of relatively unstimulating activities and the avoidance of stimulating ones (for example: rough play, loud music and television).

(iii) The child's bedroom is made as safe and unstimulating as possible (toys, hi-fi's, televisions, etc., are removed or locked away out of the child's attention). Outside sources of light, the sun or street lighting, should be blocked out by heavy curtains or blankets. These would also serve to dampen external street and traffic noise.

(iv) At bed-time the child is settled quickly in order to prevent exciting games and extensive bargaining developing in the sleeping environment. A very brief (less than two minutes) story or alternative unstimulating interaction is recommended; this is followed by a consistent settling phrase ("Good night, God bless", for example). Immediately, the parent leaves the child.

(v) The bedroom light is turned off and the bedroom door closed leaving the child in as close to complete darkness as possible.

(vi) Thereafter the child is ignored by the carers unless physically unwell. Should the child endeavour to leave the bedroom and demand attention the carer either continues to ignore him (or her) or, if this is not practical, puts the child back emphatically without any affective contact with the child other than the usual settling phrase.

(vii) A regular waking time and its associated routine are also decided upon.
Parents and principal carers (if plural) agree to back one another up and to not give in to their child's demands unless they are legitimised by illness. Should physical or psychological illness intervene, appropriate dispensations are made; however upon the child's return to health the programme is recommenced. Older children are counselled at a developmentally appropriate manner about the treatment and what is expected of them. Siblings are also encouraged to participate.

Carers are warned that during the initial phase of the treatment there is a strong likelihood that the severity of the bed-time behaviour problems will worsen and any neighbours (when appropriate) warned accordingly. Should this happen carers are urged to see this in an optimistic light: as a favourable response to treatment.

Finally, the carers are asked to repeat the agreed instructions to the therapist and also demonstrate that they can discern the difference between their child's inappropriate demands for attention and those which reflect that he or she is "legitimately" distressed.

The treatment commences immediately and the carers telephone in to the clinic (or are telephoned by DB) on the three subsequent mornings thereby any progress or problems are discussed and any changes made accordingly without deviating from the general principles of the agreed treatment plan. Extra telephone contacts are arranged on a case-by-case basis.

2.2.3. Treatment Rationale

This approach incorporated several different but not exclusive techniques: stimulus control techniques which help the child to associate the bedroom with comfortable sleep onset; sleep hygiene practices which promote sleep and restfulness rather than arousal in and
around the subject's bedtime; the rapid extinction of unreasonable requests for parental attention by complete non-reinforcement of this target behaviour. This eclectic and selective multi-technique approach is commonly employed in the treatment of several other common sleep disorders, especially the psychophysiological insomnia of adulthood (Reite et al, 1991).

2.3. STUDY DESIGN

2.3.1. Limitations Imposed by Employing a Small Cohort

The general experimental approach of this study was that of a multiple case "A/B/A" design with multiple baseline measures using the individual children as their own controls. A "no treatment" control group was not used in this study because the perceived high clinical effectiveness of the treatment as well as the high levels of chronic distress experienced by affected families dictated that any withholding of treatment was considered to be ethically unacceptable. Also, a case-controlled study would have been extremely expensive to conduct particularly since there was very little empirical research evidence that rapid extinction techniques are effective in NS/NW problems in severely learning disabled children. Naturally, such a study would be required in order to properly establish the treatment approach's efficacy if the present study demonstrates this. Ideally this would be best achieved using less severely sleep disordered children who, in turn, demonstrate a range of degrees of learning disability; this theme will be developed further in the "Discussion" section of this thesis. Therefore, in effect, this study aims to audit this form of treatment in a more naturalistic way and it was hoped that its results would indicate directions of subsequent more specific research activity.
In an A/B/A design conventionally the letter "A" represents the pretreatment, baseline, condition; "B" represents the treatment condition and "A1" the post-treatment condition. During the baseline phase the target behaviours (delayed night-settling and repeated night waking, etc.) occur spontaneously and are measured repeatedly in "real time" by activity monitoring and closely retrospectively by the parents entering these data into the Sleep Diary. These combined data provide the standard with which to compare the outcome of the intervention (the settling and ignoring advice). The "B" phase involves data collection during the treatment stage; this continues over the "A1" phase. Detection of any treatment effect is demonstrated by showing a difference between these latter data sets and those in the "A" phase. An additional phase, "A2", of two weeks' worth of diary and monitor data collection occurs in this study after a break of four months from the treatment phase in order to assess maintenance of treatment effects. These data were also compared to the "A" phase data. Point sleep problem severity scores, carer stress, behavioural problem measures and other study parameters (see below) were assessed at the initial assessment stages and also at the follow-up stages. The final follow-up stage ("A3") comprised a very brief postal questionnaire enquiring about the children's sleep pattern rating it in a similar way (visual analogue scale) and enquiring whether or not the carers required any further help specifically for any remaining sleep problem or with any other psychological problems with their children.

2.3.2. Study Procedure

Initially, there was a full explanation of the study and its principal aims (which included a statement to the effect that no drugs or other physical treatment methods will be employed in the Treatment phase of the study. This was followed by a typical child
psychiatric interview where the child and family’s suitability for inclusion into the study is determined. If this proves to be so then consent is obtained and they enter the Pretreatment phase directly. (NB: Parents were informed that they could opt out of the study at any stage without having to give any reason for doing so in the knowledge that the principal investigator would arrange alternative help for their children’s problems.)

There now follows a concise scheme of the principal phases of the study (the outcome measures referred to are described later):

(a) Stage 1: Pretreatment Phase

(i) Baseline assessments of outcome measures (Sleep Problem Questionnaire, Behaviour Problem Index, Vineland Adaptive Behavioural Scale; Malaise Inventory; Maternal Sleep Scale).

(ii) Entry into programme (consent obtained—see "Appendix" for copy of consent form)

(iii) Two weeks of diary data (and activity monitoring of those children who accept wearing the monitor).

(b) Stage 2: Treatment Phase

(i) Behavioural Modification Advice given (see above) on day 15 followed by at least three telephone calls on subsequent days to reinforce advice and "trouble-shoot" any emerging problems

(ii) Two more weeks of diary and monitor data

(c) Stage 3: Initial Follow-up Phase (two weeks after advice session)
(i) Follow-up Sleep Problem Questionnaire; Maternal Sleep Scale; Malaise Inventory.

***** (Four Month Discontinuity :- no contact unless requested by carer)******

(d) Stage 4: Four-Month Follow-Up Phase

(i) Two further weeks of diary and activity monitoring

(ii) Questionnaires: Follow-Up Sleep Questionnaire; Maternal Sleep Scale; BPI; Vineland ABS; Malaise Inventory; Advice Audit Questionnaire (to be returned by post)

(iii) Further treatment option

(e) Stage 5: Eighteen-Month Long-Term Follow-Up

(i) Postal enquiry: short questionnaire assessment of sleep problem severity (including a visual analogue scale) and enquiry as to whether further psychiatric help is required for the child.

2.3.3. Summary

The "Pretreatment", "Treatment" and "Four-month Follow-up" phases are each of two weeks duration. The first two phases are continuous with one another; the treatment intervention commences on the first night of the "Treatment" phase (night 15) and finishes on night 18, followed by ten more days without further treatment contact, unless this is requested by the families. The last phase commences four months after the end of the "Treatment" phase (five months after the beginning of the "Pretreatment" phase. The final follow-up consists of a simple postal questionnaire sent out eighteen months after the
"Treatment" phase finishes. Activity monitoring and diary records are undertaken over the three two-week phases, only.

2.4. MEANS OF TESTING THE HYPOTHESES

(a) Introduction

This study aimed to assess as closely and naturalistically as possible treatment outcomes for a focused behavioural modification programme with a group of severely learning disabled children who demonstrate severe and chronic night-waking and settling problems. Methods for assessing change in sleep disorder research is generally notoriously difficult given the qualitatively subjective nature of most sleep problems: a repeatedly poorly settling and night-waking child may represent "a problem" in one family but may not be so in another despite demonstrating similar behaviour. The complex interplay of factors which result in a "sleep problem" are discussed in the "Introduction" section of this thesis. Nevertheless, disorders of night-waking and night-settling occurring in childhood ought to provide more robust data given that desired settling times can be stated and a time for an affected child to settle and fall asleep following this time readily recorded. Similarly, night-waking episodes should be easily scored in terms of their frequency and duration. However, given that the children's parents and not laboratory technicians were required to collect these data it was anticipated that there might be a degree of inaccuracy involved in this process. Having stated this, sleep questionnaires have an accepted place in sleep disorder research and, generally, have acceptable levels of reliability particularly in large surveys (Bootzin et al, 1981). There are dozens of sleep rating scales available for
for assessing adult sleep problems but, again, none have emerged as powerful instruments however the screening questionnaire developed for children by Simmonds and Parraga (1982) seemed to be the best available in terms of comprehensiveness and face validity (see below). A multidisciplinary workshop held under the auspices of the University of Oxford in May, 1991, examined the strengths and weaknesses of all of the rating scales and assessment instruments being used at the time in childhood sleep disorder research and came to the conclusion that, as yet, there were no "gold standard" measures available, hence the use of multiple measures in sleep studies, however the Simmonds and Parraga questionnaire appeared to be closest to this ideal therefore the principal screening instrument employed in this study (the "Sleep Problem Questionnaire" - see below and "Appendix") was based upon this.

(b) Specific Measures Employed

This study required several sleep measures and other instruments, however an important factor influencing selection of these was that they could be completed reasonably quickly particularly at the follow-up stages so as not to diverge too far from normal clinical sessions.


(a) A means of establishing the overall severity of the presenting child's sleep problem in the context of their family: two means of characterising the severity of the problem were employed the first was a visual analogue scale presented as a 10 cm line
having the response limits "no problem" corresponding to zero on the left hand side and
"very severe problem" at the ten centimetre right hand limit. This measure was repeated
at the three follow-up stages and the same scale was used for the night-by-night diary
ratings of sleep problem severity. This technique has the advantage of being easily
interpretable by both the parent raters and also the principal investigator (DB). When
used in this way for repeated assessments visual analogue scales have been shown to have
acceptable face validity (Peck, 1992) In addition to this, categorical closed questions
enquiring about specific night-time behaviour were taken from Clements et al, (1986)
population screening questionnaire who, in turn, employed the night problem questions
from Wing and Gould's "Handicaps, Behaviour and Skills (HBS) Schedule" (Wing and

There follows a precise scheme for the rating of the HBS Sleep Problem Items with the
changes in the scoring already described:-

(i) **Disturbance of Sleep**

Score:

0 = "usually sleeps well"
1 = "sleeps poorly but lies quietly when awake"
2 = "sleep disturbed, wants attention, but does not scream or cry"
3 = "sleep disturbed occasionally- screams and needs attention"
4 = "sleep disturbed once or twice a week- screams and needs attention"
5 = "sleep disturbed most nights- screams and needs attention"

(ii) **Limited Hours of Sleep**

Score:

0 = "always or nearly always goes to sleep and wakes at a reasonable hour"
1 = "occasionally late sleeping or early waking but usually no problem"
3= "usually goes to sleep very late at night, but wakes at a reasonable hour in the morning"

3= "usually wakes very early in the morning but goes to sleep at a reasonable hour at night"

4= "usually goes to sleep very late and wakes very early in the morning"

Clements et al, collapsed these ratings into these two levels, with a score of two plus signifying "difficulty". In this study the two components were combined to yield a maximum score of nine; a score of six or more was taken to represent the presence of a significant sleep problem. The results of this enquiry for the group are shown in the "HBS Sleep Item Scores" graph ("Results": figure 2).

These are very similar to the sleep problem severity ratings used by Richman and Graham in their "Behaviour Screening Questionnaire" (Richman & Graham, 1971) which has been used widely in child behaviour research. In the present study these factors were rated during the initial interview and, as above, at the next two follow-up stages. Both the visual analogue scales and the HBS measures were included in a general screening questionnaire based largely on that devised by Simmonds and Parraga (1982). This assessment schedule was derived from a review of the available literature of the time concerning children's sleep problems and by obtaining an expert consensus opinion about what relevant questions should be asked of carers to cover this area. These authors devised questions which could be answered simply by principal carers taking into account the inevitable distortions revealed by such enquiry. Part 1 of the instrument enquires about demographic data and specifically the characteristics of the child's current sleeping pattern (time in bed, time to settle, etc.). Part 2 covers the range of commonly encountered sleep problems (for example: snoring, nightmares, and sleepwalking) and are
phrased in everyday language. Parents were asked to both indicate whether such problems are present in their children and, if so, their frequency. Spearman correlation studies of data collected by this method from nearly four hundred families yielded high reliability scores such that it was assumed to have high face validity. In the field it had the advantage of taking less than ten minutes to complete the questionnaire when it was applied in the form of a semi-structured interview. A condensed form of this instrument was used at the initial assessment stage (the "Sleep Problem Questionnaire") and at the follow-up stages, a shorter version was used ("Sleep Problem Follow-up Questionnaire"). (These and most of the other instruments employed in the study are included in the "Appendix" to this thesis).

(b) Night-by-night changes in night-time behaviour

These were rated by the parents and principal carers by means of a standard sleep diary similar to that used by Richman and Douglas in their treatment study (Richman and Douglas, 1985). This allowed daily entries of data concerning settling and waking times as well as details of carer responses to their children's nocturnal demands. They were also invited to rate their children's sleep problem severity on a daily basis using a visual analogue scale in a similar way to that described above for the children's general sleep problem severity. In addition to this they could also rate up to two other separate coexisting challenging daytime behaviours. The previous night to the initial assessment day was rated in the clinic and this was used to illustrate the use of the diary to the carer. Thereafter for a total of six weeks the carer would rate their child preferably at the same time each morning (the advantages of sleep diaries are discussed in the "Introduction").
2.4.2. Testing the Second Hypothesis: (Generalisation Effects will be Observed)

(a) Changes in maternal or principal carer sleep patterns

These were assessed by means of a brief, ten question, inventory devised by De Diana (1976) and developed by Visser et al (1979) which enquired about specific aspects of the children's mothers' sleep patterns. These questions took the form of statements which the respondent was invited to agree or disagree with. For example: "I think that I usually enjoy my sleep". Negative statements were also included to avoid response set conformity. In addition to these an additional question: "I often feel sleepy during the day" was included by the investigator (DB) in order to enquire about this specific possible sequel to an unrefreshing night's sleep - an area of enquiry which was a conspicuous omission from this inventory of subjective sleep quality. This yielded a maximum score of eleven. This amended version of the De Diana instrument is called the Maternal Sleep Scale (MSS) in this study. Validation data for the original unadapted version has been published (Visser et al, 1979).

(b) Changes in maternal or principal carer levels of stress

Over the course of the study this parameter was assessed using Rutter's "Malaise Inventory" (MI) which was adapted from the Cornell Medical Index. This is a quickly administered 24-item instrument which has been employed to assess carer stress in a number of studies of learning disabled children (Gath, 1978; Burdon, 1980; Cook et al, 1982; Quine & Pahl, 1985). Most recently, Bebbington and Quine (1987) employed factor analysis techniques which verified this instrument's capacity to measure a
unidimensional moderate "stress" factor. This was undertaken in response to Hirst's (1983) criticism of the MI's sensitivity to do this. When using the MI a score of greater than or equal to 5 but less than 7 reflects that the individual is significantly "stressed" and a score of 7 or above "severely stressed". Respondents are invited to indicate whether the various statements apply to them and their "yes" or "no" responses are scored one or zero respectively (maximum score: 24).

(c) The detection of Changes in Daytime Behavioural Problems (or "Challenging Behaviours")

The occasionally observed improvements in co-existing behavioural problems noted by parents during treatment of their children's NS/NW problems in ordinary clinical practice and, more specifically, also demonstrated in Quine and Wade's study (1992) justified using a specific an instrument to rate these problems as an outcome measure too. There are several behavioural rating scales available for use with non-retarded children for example the Behaviour Screening Questionnaire (BSQ), and the Child Behaviour Checklist; however, they were not suitable for this group because they were standardised on non-retarded children. Whereas the most frequently employed instruments in studies of learning disabled individuals tend to be either too complex and detailed to administer quickly or, otherwise, biased towards more able subjects. The best compromise available at the study design stage was the questionnaire developed by Cunningham for his longitudinal study of Down's Syndrome children (Cunningham et al, 1986) and was derived from Richman's BSQ which has already been referred to. This adapted instrument was called the "Behaviour Problem Index" (BPI). Its parent instrument, the BSQ has been shown to have adequate reliability and validity (Richman, Graham and Stephenson, 1982). The version
of the BPI employed in this study consisted of twenty-one questions which cover most of the commonly encountered behavioural and emotional problems which can present in severely retarded children. Each item is scored "no problem" (zero), "mild problem" (one) and "major problem" (two). There is also the facility for scoring the total numbers of problems in several items. Additionally, specific problem areas could be subdivided and each of the components rated in a similar way. In this study the maximum BPI score was sixty-four. This instrument produces a range of scores for each subject and has a good internal reliability (Cronbach's alpha score of 0.87 which compares well with that of the BSQ: 0.72). Most importantly, the BPI was also employed in Quine and Wade's study (see above) which allows comparisons to be made between their data and those obtained in the current study. The BPI was used in this study at the beginning of the Pretreatment phase and, again, at the beginning of the Four-month Follow-up phase.

(d) The Children's General Development

An objective assessment of the children's developmental attainments was necessary in order to confirm their eligibility for entry into the study (they were required to have an overall developmental level equivalent to less than half of their chronological age) and also for the purpose of assessing whether the intervention affects development generally or in specific ways over the course of the study. This would also provide objective data in order to assess the occasionally clinically observed carers' claims of specific changes in their children's adaptive behaviour resulting from this type of intervention. In this study the survey version of the revised "Vineland Adaptive Behavioural Scales (VABS)" (Sparrow et al, 1984; Harrison, 1985) was employed. This allowed assessment of the four general domains of adaptive ability: communication skills, daily living skills, socialisation
skills and motor skills and these were rated at both the initial interview and, like the BPI, also at the beginning of the Four-month Follow-up phase. These ratings yield developmental age-equivalent scores. The children’s four major domain scores were averaged out and compared to their chronological ages, yielding an overall "developmental quotient" for each child. The inter-rater reliability of this test as well as its correlation with standard intelligence tests, such as the "WISC-R" is only moderate (Atkinson, 1990); however, it does assess adaptive behaviour specifically and is thus suited to the needs of the study. It also has the practical advantage that a trained interviewer can complete it within a few minutes; therefore, given these advantages, it was the instrument of choice for this study paradigm.

4.2.3. Testing the Third Hypothesis: Acceptability of Treatment Approach to Parents

(a) Consumer satisfaction

Given that one of the principal hypotheses of this study is that an approach incorporating rapid-settling and systematic ignoring techniques when applied to a sleep disordered child will prove to be acceptable to affected children's parents and carers, means were devised to try and rate this factor and its components. There is an ongoing debate concerning the ethical basis of this forthright treatment approach (Bax, 1981; Skuse, 1994) and this has been described in the "Introduction" and will be further explored in the "Discussion" of this thesis after the relevant results are presented. Therefore the author believed it was important to solicit "consumer opinion" concerning these issues specifically whilst undertaking the treatment study. This is also in keeping with the current trend within the National Health Service which actively encourages consumer choice in health care.
Parental satisfaction with the outcome and style of the treatment approach was assessed in two specific ways:

(i) Specific questions included in the "Sleep Problem Follow-up Questionnaire" enquired about which (if any) components of the treatment approach worked and, if so, these should be stated. Estimation of how helpful the advice was thought to be was rated on a visual analogue scale which had the limits: "not helpful at all" (zero score) and "extremely helpful" (scores ten). This was rated at the end of the Treatment phase and, again, at the commencement of the Four-month Follow-up phase.

(ii) A specifically devised "Advice Audit Questionnaire" (see "Appendix") which is presented in the appendix to this thesis was given to the principal carers at the start of the Four-month Follow-up phase and this presented the individual components of the advice package (see "Intervention Technique" above) in a list and invited rating of each component as either "very helpful", "helpful", "neutral", "unhelpful" or "very unhelpful". These categories were scored +2, +1, 0, -1, and -2, respectively, yielding a maximum score for each item (eg: "setting a regular bedtime and sticking to it") of two. Parents were also invited to state and rate any other factors not included in the list but of relevance in their children's cases. Finally, in order to assess the acceptability of the approach style from the carers' points of view, several phrases were presented at the end of the "Advice Audit Questionnaire" and the respondent was invited to circle the one which best reflected their views about the style of the intervention. The options provided were: "too tough", "rather tough", "just right", "rather soft" and "too soft". It must be emphasised that these instruments were purpose-designed and have not, to date, been subjected to specific tests of their reliability or validity, however it is assumed for the
2.3.4. Testing the Fourth Hypothesis: Electronic Ambulatory Activity Monitors Will Provide Objective Evidence of Treatment Effects

(a) The Children's Night-Time Activity Patterns

This study attempted to assess changes in the children's sleep patterns objectively in a naturalistic situation and the cheapest and most convenient means of achieving this was by ambulatory electronic activity monitoring (AM). The choice of monitor for this study was influenced by several factors which have been discussed in the introduction of this thesis. Chief among these were the technical specifications (its ability to measure movement sufficiently sensitively to differentiate from the data collected between sleep and wakefulness); the instrument's physical robustness bearing in mind of the generally high activity levels expected with the children in the study; the availability of other researchers using a similar device locally; good after sales service (quick repair times so as to avoid delays during study) and, finally, the cost.

(b) Choice of Activity Monitor for this Treatment Study.

At the time of protocol development for this study there were nearly a dozen devices to
choose between and there were major differences between these devices. The device which most closely fitted the requirements of this study proved to be the Gaehwiler Electronic device Z80-32k V1 (Gaehwiler Electronic, Switzerland). This instrument was chosen for a variety of reasons some of which were not directly related to its technical specifications:

(i) The device had the capacity to record both day and night time activity data over the periods prescribed by the study.

(ii) Menu driven software for MS-DOS computer systems was provided with the device allowing simple data analysis and also easily interpretable day by day printouts of activity.

(iii) Other local researchers were using the device for sleep-related work and recommended the device. They were also prepared to advise during the study should practical problems emerge.

(iv) The cost of the device compared to its technical specifications represented the best value for money available at the time.

(v) Despite the fact that the device is produced by a Swiss company the turn around time for repairs was very short (2-3 weeks) and the terms of the aftersales guarantee are very generous.

(vi) Software for more sophisticated data analysis was being developed at the beginning of the study (the menu driven "ACTPLAN" MS-DOS system) and became available during the data collection phase of the study for use with these data.

1Actplan software package: developed by Dr. C.G.S. Kramer, University of Leiden, Dept. Clinical Neurophysiology, Rijnsburgerweg 10, NL-2333 AA Leiden, The Netherlands. To be used only under licence. Examples given in "Appendix" of thesis.
(c) Using the Gaehwiler Electronic Device

This monitor can be worn comfortably on subjects' wrists or upper arms during the study. The device contains a 32 kByte solid state memory for data storage. After completion of the data collection, the data are downloaded to an IBM, or IBM compatible, personal computer by means of a purpose designed interface connected to the serial RS 232 port. Special menu-driven software allows selection of a movement measurement interval (sampling time) between the range 0.375 seconds to 3600 seconds at steps of 0.375 seconds. A start delay may be specified (once programmed, the monitor has an intrinsic real-time clock) in order to measure activity over a predetermined period (as was the case in this study). One drawback to the device is that there is no event-marker facility which could be employed to indicate night movement events however the principal thrust of the treatment approach is to encourage parents to ignore their children during the night therefore for the purpose of this study this factor was not a problem. In order to indicate other abnormal nighttime movement phenomenae such as somnambulistic episodes, nightmares and seizures, event-marking would be important. The monitor is powered with standard or rechargeable batteries. This study used mercury batteries which last for approximately a year. Battery charge levels can be checked by means of a liquid crystal display voltmeter within the interface device in order to reduce the risk of the power failing during the monitoring period. The monitor employs an intrinsic Z80 microprocessor which allows rapid data transfer. Movement is measured by a monoaxial piezoelectric sensor attached to a balanced metal rod which, as will be described, is vulnerable to mechanical damage when monitors are attached to children for long periods.

The data movement obtained may then be analyzed by purpose devised menu-driven
software (the "ACTPLAN" software package). The applications of this program with the study monitor data is described later.

(d) Analysis of Activity Monitor Data

In this study the devices were programmed to continuously measure movement both by day and at night over the three fourteen-day periods in the study (see above). The device was worn around the upper non-dominant (where discernable) arm or wrist of as many children who would accept them. The data obtained over the combined Pretreatment and Treatment phases (which were continuous with one another) would allow the detection and thus analysis of any night-by-night changes in specific sleep problem parameters which might suggest treatment effects. Specialised software developed for this device the "ACTPLAN" software package, allowed analysis of the nightly activity patterns as well as the creation of databases which were analyzed by statistical software (SAS and MINITAB statistical software). Direct observation of the day-by-day continuous graphically-displayed bar charts of activity for the fourteen (or in one subject seven) day monitored periods allowed visual assessment of Sleep Onset Time (SOT) and Morning Waking Time (MWT), the Number and Duration of any discrete Night Waking Episodes (NWEs), which nights of the week they occurred on as well as the precise timing of these episodes: the Night Waking Times (NWTs) and when they occurred in relation to the SOT for each child. The "ACTPLAN" software allowed the investigator to "zoom-in" on night, or part of night, movement segments and store selected time sections in a data base for later analysis (see below).

After consultation with other researchers who were using this type of monitor in other
projects the following algorithms were employed throughout the study to define SOT, MWT and NWEs:

(i) Sleep Onset Time (SOT): "A point corresponding to five minutes after a continuous or near continuous high activity pattern of at least an hour which is followed by a period of little (not qualifying as a Night Waking Episode) or no activity of at least half an hour which cannot be accounted for other than by the child being asleep (ie: the monitor being removed for the purpose of bathing the child)". The records of adults and children obtained prior to the start of the study showed a gradual decline of activity interspersed with time windows showing no movement which occurred over several minutes before sleep onset. Removal of the monitor usually showed an abrupt change from a peak of relatively high activity to none at all.

(ii) Morning Waking Time (MWT): "A point corresponding to five minutes after a no or little activity pattern of movement of at least thirty minutes duration, which is followed itself by a continuous or near continuous period of activity lasting at least an hour which cannot be accounted for other than by the child being awake or, as might be indicated by an abrupt period of no activity, if the monitor is temporarily removed".

(iii) Night Waking Episode (NWE): "A discrete period of activity occurring between the SOT and MWT lasting at least fifteen minutes and having within it at least one peak of activity (acceleration per timewindow) of 50 units of movement or more. This event also requires to be temporally separated from other similar events by periods of at least thirty minutes of little or no movement which is assumed to reflect that the subject is asleep. The beginning and the ends of such episodes are measured similarly to MWT and
SOT but without the same activity / non-activity buffers*. As has been emphasised previously, sleep and wakefulness onset are processes occurring over several minutes therefore somewhat arbitrary but nevertheless consistently applied algorithms are employed. For the night waking episodes associated with the subjects of this study, these are usually characterised by high levels of arousal reflected by high activity levels per time window which are in marked contrast to those seen during sleep. This particular algorithm was derived from parental reports of night-waking episodes as well as from direct observation of the activity monitor data. Additionally, they were discussed with other researchers who were working with activity monitors and they confirmed that the algorithms had acceptable face validity. Other workers have derived more complex and reliable algorithms for sleep and wakefulness onset which have allowed the automatic scoring of night-waking frequencies and other sleep parameters from the data obtained by means of another type of activity monitor (Sadeh et al, 1992) however this highly specialised technique was beyond the scope of this study.

(iv) Total Sleep Period (TSP): This corresponds to the period between the SOT and the MWT.

(v) Night Waking Duration (NWD): This corresponds to the total time spent awake in NWEs per night.

(vi) Total Sleep Time (TST): This is calculated by subtracting the NWD from the TSP

\[ TST = TSP - NWD. \]
(vii) **Night Waking Time (NWT):** These times are calculated in minutes from the initial SOT for the night. This was investigated in order to discover whether any sleep biorhythmic factors (especially sleep rest-activity cycles) relate to the NWEs. More specifically, whether they correspond to the transitional points from deep to light sleep or, if they do not, to any external factors such as the time when the principal carers go to bed. These data are also analyzed in relation to whether the temporal arrangements of the NWEs change over course of the study.

(e) **Using the "ACTPLAN" Software**

Included within the "ACTPLAN" software are simple statistical procedures which can give night-by-night values for the following movement parameters:

(i) **Number of "Time Windows" per Night (TWN),** that is: the number of computer memory bytes covering the period between the SOT and the MWT. This figure can be multiplied by the time duration of each time window (a variable which is set before the device is activated: this is 36.5 seconds for this study unless otherwise stated) to yield a value for the Total Sleep Period (TSP) in seconds which, in turn, can be easily converted to hours.

(ii) **The Total Number of Movements (units of acceleration) per Night (TNM).** This refers to the summed movement data from all of the time-window bytes occupying the TSP for any night.

(iii) **The Mean Number of Movements per Time Window (MNTW).** This is obtained by dividing the TNM by the TWN data
(iv) The "Movement Index" (MI) is the percentage of time windows per night containing greater than or equal to one unit of movement (ie: the proportion of the night in which any activity is occurring). This is calculated automatically by the "ACTPLAN" programme once the SOT and MWT are set by the investigator.

(v) **Further Analyses**

In addition to deriving these simple parameters, other, more sophisticated, indices of treatment outcome were also derived for this study. The mean duration of night-walking episodes (ie: the mean time the subject spends awake per night-walking episode) can be easily calculated; however, the amount of movement per walking episode or, more usefully, the amount of movement per unit (minute) of time of night waking which might reflect the degree of arousal during these waking episodes, required that a series of novel calculations to be derived because the literature relating to the use of AM in sleep problems does not cover this topic from these perspectives. The calculations of duration and movement content of averaged night-walking events for each monitored individual in each phase of the study are explained and illustrated in the "Appendix".

(vi) **Rationale for these Analyses**

The hypothesis tested in this analysis being that night waking episodes are characteristically of long duration and contain high levels of movement reflecting the children's distress during these episodes. A result of successful treatment should result, over the course of the study, in reductions of the frequencies of these waking episodes, reductions in their duration (ie: they should become shorter) and also reduced levels of movement within these episodes. The corollary of this statement would be that an index of treatment failure could be an absence of these expected changes or, indeed, a
paradoxical overall deterioration (more frequent and longer waking episodes containing higher levels of movement).

2.5. DATA ANALYSIS

Apart from the diary and activity monitor data, the analyses of which will be described later, most of the other outcome measures allowed simple comparisons to be drawn between the “before” (Pretreatment), “immediately after” (Treatment), “a while after” (Four-month Follow-up) and the “at a remote time” (the few Eighteen-month Follow-up) results. The later assessments of sleep problem severity and the generalization effect measures (the MI, MSS and BPI scores, for example) are compared to the initial, Pretreatment, scores and the statistical significance of these differences of the rank orders of these scores assessed by means of the non-parametric Friedman’s and Wilcoxon Signed-Ranks Tests (Siegal and Castellan, 1988). However, for the most part the results will be displayed graphically with the mean values of the parameter given as well as its standard error of the mean to show the distribution of the scores for the group. Repeated measures analyses of variance are used with some of the diary data parameters for each child in each phase of the study to illustrate the changes in their mean values and their score ranges over its course. The power of these analyses is necessarily quite low given the small number of children enrolled into the trial; it is the general directions of change across the principal outcome measures within the group over the study which will dictate whether the treatment intervention shows clinical potential or not.

The parental diary reports of sleep problem parameters provide fourteen data points for
each child in each main study phase and the activity monitors several thousand which allow more sophisticated analyses of variance using the SAS statistical software package (repeated measures analysis of variance). Nevertheless, it was intended that the visual inspection of the graphical presentations of night-by-night changes of the children's various sleep problem parameter mean scores over the course of the study, and particularly over the periods when the behavioural advice was implemented (from night fifteen), would reveal any treatment influences.

The statistical software packages used in this study were the "Minitab" (Minitab Inc., 1989) and "SAS" (SAS Institute, 1989) systems. The textbook of nonparametric statistics by Siegal and Castellan (1988) informed both the choice and also the interpretation of the non-parametric statistics employed in this study.
3: RESULTS
Section 3: RESULTS

3.0.0 Overview of Results Section

This section of the study will begin with a description of the study sample as they presented before the intervention commenced. Thereafter, the results are structured around the principal hypotheses which underpin the study which are described at the end of the "Introduction" section; each of which will be dealt with in turn. This will be followed by three case vignettes and, finally, there will be an overall summary of the principal findings from these results.

3.0.1 RESULTS PART 1: Sample Characteristics

Fifteen severely learning disabled children and their families were recruited into the study; all of the children had severe NS and NW sleep problems which had been present continuously throughout their lives. None of the families approached to participate in the study declined to do so therefore this group represents a continuous series of referrals to the treatment project. Most of the children (11/15) were referred by Specialist Health Visitors the rest by paediatricians and a child psychiatrist colleague.

The principal characteristics of the sample are described below. These consist of: (i) their demographic data (age, sex, degree of learning disability; aetiological factors; comorbidity); (ii) form and quality of any previous help received or their sleep problems; (iii) current severity of their sleep problem (parent and investigator assessments); (iv) presence and severity of other behavioural problems and (v) maternal wellbeing (subjective assessments of their sleep quality and overall stress levels).
3.0.2 Demographic Data

The first table (table 1) shows the basic demographic data of the children recruited into the study:

Table 1: Demographic Data

<table>
<thead>
<tr>
<th>Sample size</th>
<th>15</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male : Female</td>
<td>10 : 5</td>
</tr>
<tr>
<td>Mean Age (SD &amp; Range)</td>
<td>7.2 years (2.6; 3.5 to 12.0)</td>
</tr>
<tr>
<td>Mean Developmental Age (SD &amp; Range)*</td>
<td>2.0 years (1.2; 0.73 to 3.85)</td>
</tr>
<tr>
<td>Mean Developmental Quotient (Range)*</td>
<td>27.5; (8.9 to 47.3)</td>
</tr>
</tbody>
</table>

(* derived from Vineland ABS data)

It can be seen that the mean developmental age of the cohort assessed in terms of their adaptive behaviour is less than a third of its mean chronological age reflecting that all of the children have at least a severe degree of learning disability and all of the children's developmental ages were less than half of their chronological ages. In terms of their developmental quotients (mental age/chronological age x 100), four scored between 35 and 50; seven between 20 and 34; four scored less than 20. Therefore a wide range of ability was discovered within the sample notwithstanding their being categorised as having "severe" degrees of learning disability.
3.0.3 Aetiological Factors for Learning Disability

A specific aetiological factor for the severe learning disability was known for nine (3/5) of the sample children. The commonest was Down's Syndrome (3 children) and the rest were single cases of macrocephaly; Angelmann Syndrome; Smith-Magenis Syndrome; carcinuria; perinatal cerebral anoxia; cerebral leucodystrophy.

3.0.4 Co-morbidity

(a) General Health of the Children

The general health of the group was comparatively good notwithstanding that four of the more severely learning disabled children had marked degrees of cerebral palsy; three of the children were epileptic with generalised seizure patterns, all of whom had not had a nocturnal fit for at least a month prior to entry into the study. Two had a past history of asthma but were not receiving active treatment. One child suffered from recurrent ear infections which affected the control of his epilepsy and also the severity of his nighttime behavioural problems; however, this was quiescent at the point of initial assessment.

(b) Sensory Impairments

One child, a girl with Smith-Magenis syndrome, was partially deaf and wore bilateral hearing aids. Another, a boy with cerebral leucodystrophy, had cortical blindness.
3.0.5 Other Cohort Characteristics

(a) Family Factors

Most of the children (11/15) were living with both natural parents; two lived with their mothers with no contact with their fathers; one lived with his biological mother and a step-father; only one child was in long-term foster care but had regular contact with both natural parents.

3.0.6 Form and Quality of Previous Help for Sleep Problems

(a) Prior Help for Sleep Problems

Two thirds of the families had sought help for their child's sleep problem prior to presentation. Table 2 shows the types of treatment they received:

Table 2: Previous Help

<table>
<thead>
<tr>
<th>Type of Help</th>
<th>Numbers of Families</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sedative medication</td>
<td>7</td>
</tr>
<tr>
<td>Health Visitor behavioural advice</td>
<td>6</td>
</tr>
<tr>
<td>Non-professional advice</td>
<td>2</td>
</tr>
<tr>
<td>Clinical psychologist behavioural advice</td>
<td>1</td>
</tr>
<tr>
<td>Child psychiatrist behavioural advice</td>
<td>1</td>
</tr>
</tbody>
</table>

(some families received more than one form of help)

It can be seen that the commonest form of help that had been received consisted of a
prescription for sedative medication (either chloral hydrate or trimethazine tartrate). Other types of therapy consisted of a variety of behavioural advice approaches among which was limit-setting and ignoring (one child), and also two children received the "graded changes" approach which has been described in the "Introduction" section.

(b) How Helpful was this?

During the initial assessment parents and principal carers were asked to rate the overall effectiveness of previous therapeutic approaches. This was achieved by means of a visual analogue scale whose limits were zero which represented "no help" and ten: "extremely helpful". Table 3 summarises the results of this enquiry.

Table 3: Helpfulness of Prior Intervention

<table>
<thead>
<tr>
<th>Intervention Helpfulness</th>
<th>Parents' Scores</th>
</tr>
</thead>
<tbody>
<tr>
<td>Visual Analogue Scale Score; x/10: (mean; sd; range)</td>
<td>1.4; 2.5; 0 to 7.</td>
</tr>
</tbody>
</table>

These scores reflected generally low levels of satisfaction with previous advice and hence the ongoing nature of the families' current difficulties at presentation. Additional comments obtained from the interviews with the parents emphasised that the prescription of sedative medication, specifically, was universally regarded as being of either no help or even positively damaging; for example, states of severe and extremely distressing paradoxical excitement were reported for three of the children treated in this way.
3.0.7 Current Severity of the Children's Sleep Problems at Initial Assessment

(a) Parental and Investigator's Ratings of NS/NW Problem Severity

The parental and investigator's ratings are summarised in table 4 below:

Table 4: Severity of Children's Sleep Problem

<table>
<thead>
<tr>
<th>Type of Assessment</th>
<th>Result</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parents: Visual Analogue Scale(0=&quot;no problem&quot; to 10=&quot;severe&quot;) (x/10)</td>
<td>Mean: 8.0; sd: 1.3; range: 6-10</td>
</tr>
<tr>
<td>Investigator: HBS Sleep Problem Composite; (x/9)</td>
<td>Mean: 8.2; sd: 1.2; range: 6-9</td>
</tr>
</tbody>
</table>

It can be seen that all of the children referred had severe degrees of sleep disturbance in terms of both of the measures employed thus rendering them eligible for inclusion in the treatment study. All of the children had lifelong severe NS/NW problems. The scores of these two measures show a high concordance with each other which suggests that the visual analogue scale and HBS-derived scale perform similarly to each other and hence would suggest acceptable reliability and validity.

(b) Other Sleep Problems

The modified version of the Simonds and Parraga questionnaire used to assess the subjects (see "Method" section) allowed the detection of any other co-existent sleep problems: Eight of the children were incontinent of urine both by day and by night, a further two
only enuretic at night (all these children wore nappies during the night), two of the children snored habitually and two had marked nocturnal bruxism. None of the children exhibited recurrent nightmares or night-terrors which might be reported for a cohort of non-retarded children of similar ages to the study cohort.

(c) Co-Sleeping

Eleven of the children frequently co-slept with either one or both parents and this practice was described as a "major problem" by most affected parents. (The changes observed in this problem parameter after the intervention will be discussed later in this "Results" section).

3.0.8 Co-existent Behavioural Problems

All of the children's parents and carers reported problems with their children's daytime behaviour in addition to their night-time problems. From the BPI data the mean number of "major" daytime problems per child (ie scoring 2+ for a behavioural problem item) was 4.5 and other minor problems 5.5. In rank order of combined frequency and severity, the specific BPI items most commonly cited within the group were: "dependency", "attention seeking", "bad behaviour" and "management" problems. Equal fifth place in citation by severity rating were: "temper tantrums", "overactivity" and "concentration" problems. These problem items were reported for fourteen out of the fifteen children (93.3%) in each case.

3.0.9 Maternal Wellbeing
This parameter was formally rated in terms of the mothers' subjective Maternal Sleep Scale (MSS) assessments and also their Malaise Inventory (MI) ratings (table 5).

Table 5: Maternal Wellbeing

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Score (mean; sd; range)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Maternal Sleep Scale; x/11</td>
<td>4.6; 2.3; 2-10</td>
</tr>
<tr>
<td>Malaise Inventory; x/24</td>
<td>8.7; 4.5; 2-16</td>
</tr>
</tbody>
</table>

The majority of mothers reported poor quality sleep which they attributed primarily to their children's settling difficulties and their recurrent night-wakings. Their sleep problems being initial insomnia, difficulty in getting back to sleep once disturbed and marked daytime sleepiness. Overall levels of subjective stress were high and two thirds of mothers (10/15) scored above the "severely stressed" threshold score of 7/24 on the MI. From a clinical point of view four of the parents reported psychological symptoms of sufficient severity to allow a diagnosis of mild to moderate clinical depression to be made (however this finding was not addressed clinically in its own right).

3.0.10 Summary of Sample Characteristics Prior to Entry into Study

A continuous series of fifteen severely learning disabled children were recruited who displayed lifelong severe NS/NW problems. The children's demographic and medical details reflected that they represented a recognisably typical cross section of the severely retarded children who present to Leicestershire's Child and Adolescent Psychiatry service. All of the children demonstrated additional daytime behavioural problems and their
mothers were experiencing high levels of both general and sleep-related stress which were primarily attributable, from their mothers' perspective, to their children's sleep problems. The parents reported generally very low satisfaction with previous forms of help especially with sedative treatment. All of these children were deemed eligible for inclusion into the treatment programme and all of their parents (or guardians) consented to this.

HYPOTHESES

(a) Introduction

Over its active course there were no drop-outs and all of the families participated in the final postal follow-up phase, eighteen-months post-treatment. This suggested a high level of commitment on the part of the families to help their children.

The three principal stages of the study were as follows: the Pretreatment (or baseline) phase; the Treatment phase and the Four-month Follow-up phase. Each of which were of two weeks duration; the Pretreatment and Treatment phases representing a continuous four-week period with active treatment commencing on night fifteen. Over these periods the families were asked to keep nightly diary reports of their children's sleep patterns; five of whom were successfully monitored by means of the activity monitors worn continuously over these three phases. The long-term Eighteen-month Follow-up phase which was conducted by post or by telephone, consisted of a simple questionnaire enquiry concerning the children's sleep pattern at that time and provided the opportunity for the parents to request further help from the Child and Adolescent Psychiatry Service.
(b) Quality of Diary Data Over the Course of the Study

Between the initial and follow-up phases the quality of the diary data relating to the children's sleep problems tended to deteriorate and within all of the phases there was some evidence of block entries of data covering several days in most of the children's records, although this was most evident at the Four-month phase. A level of 80% record completeness (or greater) was taken as being sufficient for inclusion into the data analysis. Thirteen of the families completed their diaries sufficiently comprehensibly to allow the data to be analysed over the Pretreatment and also Treatment phases. However, this fell to ten children's data sets at the Four-month Follow-up. The two families who did not provide sufficient information in the first two phases were generally the least organised of the study cohort and low levels of parental literacy contributed to this. During the latter phase another three families did not provide sufficient information and this was probably primarily attributable to reduced motivation to record their children's sleep patterns owing to significant improvements in these following the behavioural modification advice which will be demonstrated below.

The five families whose children were successfully monitored with activity monitors provided analysable diary data for all of the principal phases of the study. Attempts to analyse diary data concerning the day-to-day changes in daytime behaviour problems were thwarted owing to a marked incompleteness of this data set. Analysis of changes in this parameter was only successful terms of the children's Pretreatment and Four-month Follow-up BPI scores.
(c) Testing the Study Hypotheses

At this point each specific principal hypothesis underpinning this study will be tested in turn:

3.1.1 Hypothesis 1: This Treatment Approach is Effective in NS/NW Problems in Severely Learning disabled Children

The treatment outcome results from the activity monitor data obtained from five of the children are presented separately below in the monitor data section. The principal measures employed to test the first hypothesis of this study were the parents' and investigator's general assessments of sleep problem quality before the treatment commenced, two weeks, four months and then, finally for the parents' assessment only, at eighteen months after the initial treatment session. This is followed by the data obtained from the "co-sleeping" item from the BPI questionnaire administered at the Pretreatment screening and also at the beginning of the Four-month Follow-up phase. Night-by-night changes in the sleep problem parameter (problem severity, time asleep, waking time, night-waking frequency) are presented from the continuous diary entry data as well as any changes seen in night-waking frequencies in terms of which night of the week they were occurring.
3.1.1.1 Children's Sleep Problem Severity:

(a) Parent/Carer Scores

The changes in the parental and carer ratings for their children's sleep problem severity scores over the course of the study are shown in the first figure (Figure 1):

This graph shows the high mean subjective score for the group immediately prior to entry into the baseline pre-treatment stage and the significantly reduced scores at the three follow-up points \((F = 28.2; df = 3; p < 0.001; \text{Friedman Test})\). Compared to the Pretreatment mean score, after the Treatment phase and at the beginning of the Four-Month Follow-up phase the mean scores were 72.1\% lower. At Eighteen-month Follow-up this difference had narrowed to 63.4\%.
(b) Degrees of Change in Parent / Carer Ratings of the Children’s NS/NW Problems Over the Course of the Study

The next table (table 6) displays the pattern and degree of improvement of the parental assessments of their children’s sleep problem severity over the three follow-up phases of the study with reference to the Pretreatment, or baseline, assessment point scores. The increments employed are 0% to 33%: "minor" improvement; 34% to 66%: "moderate"; and 67% to 100%: "major improvement" compared to baseline ratings.

Table 6: Degree of Change Over Study

<table>
<thead>
<tr>
<th>Improvement compared to pretreatment ratings</th>
<th>Two-week follow-up: no. children/15 (%)</th>
<th>Four-month follow-up: no. children/15 (%)</th>
<th>Eighteen-month follow-up: no. children/15 (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Minor (0%-33%)</td>
<td>2 (13)</td>
<td>1 (7)</td>
<td>2 (13)</td>
</tr>
<tr>
<td>Moderate (34%-66%)</td>
<td>4 (27)</td>
<td>5 (33)</td>
<td>6 (40)</td>
</tr>
<tr>
<td>Major (67%-100%)</td>
<td>9 (60)</td>
<td>9 (60)</td>
<td>7 (47)</td>
</tr>
</tbody>
</table>

These data reveal that the majority of the children improved to a "major" degree even at the two-week follow-up stage. This pattern was substantially maintained at the Four-month stage with a slight deterioration at eighteen months attributable to two children dropping from the "major" improvement group into the "moderate" improvement group and one from the "moderate" group into the "minor" group. Thus it was demonstrated that most of the children maintained their improvement over the course of the study.
None of the children's problems deteriorated to their pretreatment levels. Intercurrent family crises and medical problems (deteriorated epilepsy control and a chronic suppurative ear infection - see case vignette #3 below) were identified as the primary reasons for the slight relapse seen in three children. Another child had developed a sleepwalking disorder which disturbed the family and merited treatment in its own right. Two other families requested further help for their children's sleep problems, specifically. Three other families requested help for other behavioural problems; nevertheless, nearly two thirds of the families appeared to be coping sufficiently well with their children to not require further help from the child and adolescent psychiatry service at the end of the study period.

(c) *Handicaps and Behaviour Scale (HBS)-Derived Ratings*

The principal investigator's (DB's) ratings of the children's sleep problem severity are shown in figure 2 below:

These scores were derived from the two HBS-derived night-settling and night-waking items which were included in the Sleep Problem Questionnaire screening instrument and
the two subsequent follow-up questionnaires. The maximum score for the children's sleep problem severity being *nine*. It can be seen that the changes in severity observed over the three study phases shown in this graph are very similar to those reported by the parents (see above) and the follow-up scores differed from those of the *Pretreatment* phase by a similar degree ($F= 22.8; df= 2; p<0.001; Friedman Test$).

*(d) Children Sleeping in Parents' Bed (Co-Sleeping)*

Figure 3 shows the group patterns for the children who slept habitually in the parental bed at the outset of the study and also at the *Four-month Follow-up* stage. The item of the BPI covering this aspect of behaviour defines a "major" co-sleeping problem as: "a frequent problem occurring all or most of the night and at least twice a week" and a "minor" problem: "occurs all night or for two or more hours a night once a week or less" (a "no problem" category is also provided).

The data reveal that before the treatment intervention the majority of children slept regularly in their parents' beds and this represented a "major" problem. At the follow-up
stage only one child continued to exhibit this bedtime behaviour to this degree, in the majority of cases (7/11) the problem was extinguished entirely; the remainder continuing to co-sleep to a lesser degree.

Summary

From these combined results it can be concluded that significant changes in the children's overall sleep problem severity were observed at discrete points following the intervention. Next, the data relating to sleep problem parameters derived from the children's sleep diaries will be presented.

3.1.1.2 Parental Diary Ratings of Sleep Problem Severity

At this stage the analysis of the diary data will concentrate specifically on the night-by-night assessments of sleep problem severity, settling (in bedroom) time, night waking frequency and duration, definitive waking time, total sleep duration, night of the week effects upon night-waking frequencies. Each parameter will now be presented in turn.

(a) Daily Diary Assessments of Sleep Problem Severity by Principal Carers.

The night-by-night assessments of sleep problem severity were rated on a visual analogue scale. The scores for the majority of the study cohort of children (converted to percentages) are shown in figure 4:
In the Pretreatment (baseline) phase a mean score for the 14 nights of 42.8% (s.d. 5.5%) was obtained. The overall mean score in the Treatment phase is 21.1% (s.d. 6.0%) which represents an overall 50.7% reduction which differs significantly from the baseline scores and this is confirmed when a repeated measures analysis of variance is conducted for each child's data between the Pretreatment and Treatment phases (df = 1; F = 98.6; p < 0.001; ANOVA). It is evident from the graph that the trend towards improvement starts as soon as the behaviour modification treatment commences (night # 15). The steepest fall in the graph is seen to occur between this point and night 18 which corresponds exactly to the brief period where there was therapist contact (treatment session during day 14 (night 15) plus telephone contacts during the ensuing three days). Thereafter the trend is towards further improvement reaching a mean end score for the group on night #28 of 11.0%. The results for the Four-month Follow-up stage reveal a similar pattern to that seen in the Treatment phase data with a mean sleep problem score for the phase of 22.5% (s.d. 6.4%). Over the three phases a highly significant trend towards improvement was observed (df = 2; F = 57.33; p < 0.0001; ANOVA). This represented an overall sleep problem reduction of 47.4% compared to the baseline. The rank order of changes in sleep problem severity for the ten children represented in each of the three diary-monitored phases changed
significantly (Fr= 27.3; df= 2; p < 0.001).

It should be noted that these data reflect the specific nightly sleep problem scores and are not directly comparable with the data displayed in Figure 1 which show the parental assessments of their children's overall sleep problem severity, scores which are influenced by several factors the most important of which being marked night-by-night variations in NS and NW severity between and within the children (this unpredictability of many of the children's sleep problems was another source of family stress from parent reports).

(b) Total Sleep Period

The Total Sleep Period, that is the estimated period between falling asleep and waking definitively in the morning, was calculated from the 13 diary data sets which included those of the five patients who were also monitored electronically. The following graph (figure 5) shows the night-by-night changes in total sleep period over the three study periods:-

![Total Sleep Period: Diary Data](image)

(*df= 1; F= 12.8; p < 0.001. **df=1; F=46.8; p< 0.0001, Repeated Measures ANOVA)

Once again, a marked treatment effect can be observed over the treatment contact time in that the mean duration of the children's sleep periods consistently increased from night
This trend is sustained throughout the rest of the Treatment phase and maintained at the Four-month Follow-up phase. The precise values of the differences between the baseline and the two successive phases 42 and 30 minutes, respectively. There were significant differences between the means of both the treatment and 4/12 follow-up stages compared to the pretreatment baselines. Despite a mean 12-minute fallback towards the baseline mean this was not a significant reduction in reference to the treatment period mean (df=1; F=3.1; p< 0.1) and even if it was, it was not clinically significant. The overall trend observed in terms of the total sleep period was towards significant improvement (df=1; F=17.45; p< 0.001).

(c) Night Settling Times and Time to Settle

The diary data relating to the time taken for each child to settle following the fixed bedtime was insufficiently complete to allow a meaningful analysis of how long it took the children as a group to actually get to sleep after settling in the bedroom. The actual sleep onset times of the five children monitored with the AM technique were established from their monitor data and their parents' reports of sleep onset estimations were sufficiently comprehensive to allow analysis of how long it took for the to get to sleep after having settled in their beds. (This is shown below in section 3.1.4 (c))

Figure 6 shows the night-by-night changes in settling (settling in the bedroom) times for the children derived from the parent/carer sleep diary entries:
The night-settling graph shows a marked change from the baseline mean settling time of 9:34 pm (s.d. 24 minutes) to the Treatment phase mean of 8:40 pm (s.d. 21 minutes): an average of 54 minutes. There is most change in the slope of the graph over the four-day treatment contact period (nights 15 to 18) suggesting a treatment effect. At the Four-month Follow-up stage there is a fall-back towards the Pretreatment value (mean settling time: 9:10 pm; s.d. 15 minutes) although this is not complete there being sustained 20 minute difference between these scores. When analysis of variance is performed for each child in each study period an overall significant reduction in settling time occurs (repeated measures ANOVA: df=2; F=23.65; p< 0.001). The rank order of changes between the subjects' scores for the three phases also achieves statistical significance (Fr= 20; df= 2; p< 0.01).

(d) Morning Rising Times

The times of definitive rising were also recorded for each child however, the changes in the waking times for the children over the study are not as pronounced, indeed the pretreatment and treatment stages results show no obvious major variations although the
distribution of scores around the mean score for the group is narrower in the Treatment phase data set compared to the baseline data (means and standard deviations for these phases are 7.12 hours (s.d. 0.21) and 7.19 hours (s.d. 0.27) respectively). The mean value for the Four-month Follow-up phase is 7.37 hours (sd: 0.32) which is significantly later than the treatment phase value (p< 0.05; ANOVA). This later mean rising time for the group is possibly another index of treatment effectiveness although it was not a primary focus of the intervention. Also, when this later rising time is seen in the context of the later settling time at the Four-Month Phase there is a marked overall consistency between the total sleep period (total time from time settled to ultimate morning rising time) at this stage and that see during the Treatment Phase.

(e) Night-Waking Patterns: Diary Records of Night Wakings

Parents and carers were encouraged to keep records of the children's night-waking habits and these data are shown in figure 7:

![Night Waking Frequencies: Diary Data](image-url)
The Pretreatment phase data reveal a marked night-by-night variation in NW frequency and this variable pattern was reported to be a factor contributing to the overall assessment of severity of the children's sleep problems. Neither the duration nor the severity of these phenomena, factors which also contribute significantly to their overall impact on families, are examined in this analysis because of insufficient diary data relating to these parameters. There is a reduction both in respect of this variability and also the overall frequency over the treatment contact period, patterns which are sustained over the rest of the Treatment phase. Over the Four-Month Follow-up phase a trend reflecting a further reduction in frequency of reported night-wakings is observed. It should be noted that the waking patterns of the three children whose reports were insufficiently complete to include in this analysis may have altered this. The comparison between the diary reports and activity monitor derived waking data further enhance interpretation of these findings (see below). When the mean values and their standard errors are plotted for each study phase (figure 8) the underlying trend is for the children to wake sequentially less frequently in the later stages of the study compared to the baseline. The overall reduction in reported night-waking frequency was 59.8%.

These results are statistically significant when the NW data for each child in each phase
is compared by Repeated Measures Analysis of Variance (df = 2; F = 22.3; p < 0.001).

The changes in the rank order of changes in their NW frequencies also vary significantly
(Friedman statistic = 24.1; df = 2; p < 0.001).

(3) Day of Week Variations of Night-Waking Frequencies

In order to discover whether the reported night-waking frequencies are influenced by
whatever night of the week they occur, the total diary-recorded wakings for 13 children
in the Pretreatment and Treatment phases and for the 10 at the Four-month Follow-up
phase are plotted for each night of the week (figure 9). These data reflect the wakings
derived from a total of 72 weeks-worth of parent reports and are plotted for each phase
of the study in terms of the overall total waking frequencies for each day of the week.
This was easily achieved because each main phase of the study began on the same day
of the week (Wednesdays).

**FIGURE 9**
Night Wakings by Night of the Week
(78 Weeks of Diary Data combined)

In the first two phases of the study weekend troughs and midweek peaks are seen. These
troughs are 30% lower than the peak levels. This overall trend is not seen at the Four-
month Follow-up phase because of the low rates of reported night-wakings during this 
phase and baseline effects.

The plot for the total scores across the study reveals an overall trend of higher levels 
midweek (Tuesdays and Wednesdays) and a trough on Saturdays. Whether or not this 
trend can be confirmed by the results obtained from the objective monitor data is shown 
later in the activity monitor data section of the results (see below).

3.1.2 Hypothesis 2: The Treatment Approach will produce Generalization Effects

This hypothesis was tested in terms of the detection of changes in various child and 
maternal parameters which were not explicitly addressed by the treatment programme. 
Changes in the children's daytime behavioural parameters were assessed using the BPI 
scale (x/44); overall adaptive behaviour: the Vineland ABS; maternal stress: the MI 
(x/24); maternal sleep quality: the MSS (x/11). These measures are now presented in turn:

3.1.2.1 Changes in Other Problem Behaviours

(a) Overall BPI Scores

The scores for the children's behavioural (and emotional) problems were derived from the 
Behavioural Problem Index (BPI) ratings which was obtained at the start of the study and 
also at the beginning Four-month Follow-up phase. The items relating to night-time 
behaviour were included and represented 4/64 (6.25%) of the total score. The following
The overall mean scores for the group as well as the standard errors of their means:

**Figure 1**: Behaviour Problem Index (BPI) Scores, Before, and Four Months After Treatment (n = 15)

The rank order of changes in these overall scores differ significantly (p < 0.001; Wilcoxon Signed Rank Test) reflecting a general improvement in the children's behaviour. This degree of change remains the same when the three sleep problem item scores are omitted from the analysis. When the scores for each item for each child in the two study phases were compared in terms of number of children demonstrating a specific problem item at initial assessment multiplied by the mean percentage score change for that item for each child, those showing most improvement at the Four-month phase were, in rank order: "sociability", "temper tantrums", "attention-seeking", "overactivity" and "worries". Other specific items showing general improvement included "bad behaviour" and "eating problems" whereas other common problems at initial assessment such as "soiling" showed no improvement at all. No patterns of significant deterioration in any of the behavioural or emotional parameters were detected within the cohort.
Another way of presenting these data is to compare the mean rates of discrete daytime problems per child revealed by the BPI in the following categories: "major" and "minor" problems at the Pretreatment stage with those reported at the Four month Follow-up stage.

(For the purposes of this analysis the two sleep problem items were omitted.) The results are presented in figure 11 below:

This analysis reveals that the majority of children had both multiple and severe behavioural problems other than their sleep problems before the intervention. At the Four-month Follow-up stage there were significant improvements in these rates: most of the children exhibited fewer and less severe problems after the intervention. When the scores for these data are combined ("major problem" scoring two and "minor problem" scoring one on the BPI) the combined scores for these are: Mean Pretreatment rating: 14.4 (sd: 4.7) and Mean Four-month Follow-up rating: 10.87 (sd: 4.0). These are significantly different from one another (p< 0.01;WSRT). These scores are very similar
to those of Quine and Wade's (1991) intervention study which also assessed behaviour with the BPI. They obtained mean scores of 13.0 (s.d. 4.6) and 9.7 (s.d. 4.3) at the pretreatment and their cohort's follow-up stages, respectively, suggesting a similar effect on daytime behaviour despite differing treatment approaches (see the Introduction).

(c) Vineland Adaptive Behaviour Scales Scores.

The children in the study were assessed from a developmental perspective in terms of their levels of adaptive behaviour by means of the "Vineland Adaptive Behavioural Scales" (second edition, survey form). As was the case for the BPI scores the two assessment points were at the Pretreatment assessment and at the Four-month Follow-up stage. The results are shown in the following graph (figure 12):

<table>
<thead>
<tr>
<th>Ability Domains of Vineland ABS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Communication</td>
</tr>
<tr>
<td>Daily Living Skills</td>
</tr>
<tr>
<td>Socialisation</td>
</tr>
<tr>
<td>Motor Skills</td>
</tr>
</tbody>
</table>

The mean ratings across the four major domains of the instrument are shown as well as overall developmental scores (expressed in months of development). It can be seen that
as a group the children were relatively impaired in terms of their socialisation and communication scores compared to their motor and daily living skills scores and this relative deficiencies may have some relevance to the aetiology or the maintenance of the sleep problems in some of the children (see the introductory discussion of aetiological theory). At the Four-month Follow-up stage this differential between the domains remains however the overall mean developmental rating for the group is six months higher than the pre-treatment level (the actual time elapsed between the rating stages was approximately five months) (Wilcoxon statistic = 95; p= 0.008).

Parents reported that many of their children continued to develop new skills over the study period and there were some unsolicited reports of specific skill improvements (improved powers of concentration and becoming developing an interest in picture books- see Case Vignette 1, for example). One child's scores decreased; however, this child's condition (cerebral leucodystrophy) is a progressive cerebral degenerative disorder therefore this was a predictable finding. This child's sleep problem resolved quickly and this was maintained at four and eighteen months.

3.1.2.2 Maternal Wellbeing

(a) Changes in Maternal Sleep Quality Scores

Maternal assessments of their own sleep quality using an amended version of the DeDiana sleep rating scale, the Maternal Sleep Scale, over the course of the study are shown below in figure 13:
Maternal Sleep Scale Scores

Good Quality sleep

Mean Scores (+ SEM)

Poor Quality sleep

This shows significant improvements in the rank order of MSS scores over the study (Fr=17; df=2; p<0.001). As in the case of the children's sleep problem scores (figure 3) there is a tendency towards further improvement between the two follow-up stages despite their being no further clinical input.

(b) Malaise Inventory Scores

These are shown in the following graph (figure 14):

A significant reduction in stress assessed by this measure is shown at both follow-up stages (Fr=87.03; df=2; p<0.001. Again, there is an additional improvement between the two follow-up scores although these are not as marked as that obtained between the
3.1.3 Hypothesis 3: The Treatment Approach will Prove to be Acceptable to Parents

3.1.3.1 Parent/Carer Satisfaction with Treatment

(a) Acceptability of General Approach

Upon systematic consumer audit both during and also after the study the families were generally well pleased by the improvements in their children's sleep pattern and, the majority were also of the opinion that the treatment approach was "just right" for their children (figure 15). The following graph shows the categorical response pattern:

Only a minority considered it "rather tough" but were nonetheless willing to continue with the treatment programme.
(b) Overall Levels of Satisfaction with Treatment

Parents and principal carers were asked to rate the overall helpfulness of the treatment approach at two stages: two weeks after the treatment started and at the beginning of the four-month follow-up stage. A visual analogue scale was employed, the limits being "zero" representing "no help at all" and "ten": "extremely helpful".

The results of this enquiry are shown in table 7:

Table 7: Parents' Satisfaction With Treatment

<table>
<thead>
<tr>
<th>Assessment Stage</th>
<th>Satisfaction Score: mean &amp; s.d. (N=15)</th>
</tr>
</thead>
<tbody>
<tr>
<td>End of Treatment Phase</td>
<td>8.6; 1.6</td>
</tr>
<tr>
<td>Start of Four-Month Follow-Up Phase</td>
<td>8.9; 1.9</td>
</tr>
</tbody>
</table>

Thus there was a high overall satisfaction with the results of the intervention from the carers' points of view and, what's more, the levels of satisfaction tended to further increase slightly over time. No adverse events probably or possibly attributable to the intervention were reported over the entire course of the study.

(c) What Components of the Advice Package Worked?

Parents and principal carers indicated from a list consisting of the key components of the advice package those which they felt helped their children achieve a better sleep pattern (see the "Advice Audit Questionnaire" in the appendix). Table 8 shows the reported rank order of helpfulness of these components (the figures in brackets represent the mean score for the item; range: +2.0 to -2.0).
<table>
<thead>
<tr>
<th>Rank</th>
<th>Mean</th>
<th>Advice Item</th>
</tr>
</thead>
<tbody>
<tr>
<td>1st.</td>
<td>1.64</td>
<td>Carers agreeing to back one another up</td>
</tr>
<tr>
<td>2nd.</td>
<td>1.4</td>
<td>Winding the child down pre-bedtime</td>
</tr>
<tr>
<td>3rd.</td>
<td>1.34</td>
<td>Setting a regular bedtime and sticking to it</td>
</tr>
<tr>
<td>4th.</td>
<td>1.27</td>
<td>Ignoring child's demands for attention after settling (unless unwell)</td>
</tr>
<tr>
<td>5th.</td>
<td>1.13</td>
<td>One hour pre-bedtime settling routine</td>
</tr>
<tr>
<td>6th.</td>
<td>1.07</td>
<td>Making bedroom safe and secure</td>
</tr>
<tr>
<td>7th.</td>
<td>1.00</td>
<td>No-fuss prompt put-back if child awakens and roams</td>
</tr>
<tr>
<td>8th.</td>
<td>0.93</td>
<td>Light off and bedroom door closed</td>
</tr>
<tr>
<td>9th.</td>
<td>0.86</td>
<td>Short settling time and use of settling phrase</td>
</tr>
<tr>
<td>10th.</td>
<td>0.67</td>
<td>Removing sources of stimulation (toys, TVs, etc.) the from bedroom</td>
</tr>
<tr>
<td>11th.</td>
<td>0.28</td>
<td>Warning the neighbours of the possibility of temporary noisiness</td>
</tr>
</tbody>
</table>
(d) How Soon Were Changes Seen?

At the end of the Treatment phase parents were asked how soon any improvements in their children's sleep patterns were seen in terms of the number of nights following the initial treatment session on day fifteen. All of the parents gave an estimation of this parameter and the mean value was 3.6 nights (sd: 1.9; range: 1 to 7 nights) reflecting that the treatment worked quickly and this is consistent with the changes seen in the graphs relating to changes in the parents ratings of the night-by-night sleep problem parameters (figure 3.1.1.2 (a)) and also the monitor-derived data.

3.1.4 Hypothesis 4: Electronic Ambulatory Activity Monitors

Would Provide Helpful Data

3.1.4.1 Overview

(a) Introduction

Five of the children enrolled into the study were also monitored over the three main study periods by means of the Gaehwiler Ambulatory Electronic Activity Monitor (see Method section). These children were representative of the larger group in terms of the general selection criteria and also their results obtained from the principal outcome measures. Out of the nine (60% of cohort) children where AM was attempted only four patients were successfully monitored in that day and night continuous activity data for the three fourteen-day blocks of the study were obtained for these patients. Additionally, one further patient was monitored successfully for the first seven days of each block. The data so obtained yielded information concerning several quantitative parameters of these patients' sleep patterns (settling time, night waking frequencies, total sleep time and also
the distribution of activity during the nights). The monitors also provided daytime activity data but this was contaminated by too many movement artifacts (for example, the monitor being removed for bathing purposes) to allow meaningful analysis.

(b) Influence of Monitoring Technique on Results

There were no major differences in terms of treatment outcomes between the children who were monitored by AM and those who were not. This can be demonstrated by the similar trends revealed in the various outcome measures for group as a whole (see above) are compared to those for the monitored children specifically (see below) which strongly suggests that the technique in itself does not significantly influence the therapeutic process.

(c) Robustness and Mechanical Reliability of the Activity Monitors

An important aspect of employing activity monitors in the study was to assess their robustness as "real time" data measures. The next table (table 9) shows how successful this initiative proved to be:

<table>
<thead>
<tr>
<th>Table 9: Monitoring Reliability; N=9</th>
</tr>
</thead>
<tbody>
<tr>
<td>MONITORED SUCCESSFULLY:</td>
</tr>
<tr>
<td>PARTIAL SUCCESS*:</td>
</tr>
<tr>
<td>MONITORING FAILURE:</td>
</tr>
</tbody>
</table>

(*investigator calibrated the monitor incorrectly to sample movement over only one rather than two-week periods)
Monitoring failed in four children completely; the next table (table 10) shows why this was so:

Table 10: Reasons for Monitoring Failure; N=4

<table>
<thead>
<tr>
<th>Reason</th>
<th>Count</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child refused to wear monitor</td>
<td>1</td>
</tr>
<tr>
<td>Monitor malfunction</td>
<td>3</td>
</tr>
</tbody>
</table>

It can be seen that successful monitoring was achieved in only five out of nine cases however, the data obtained from the child monitored for only one week out of each study stage provided analyzable data. Generally, the monitors were well tolerated: the child that refused made her feelings about the monitor very plain to the investigator by pulling it off and throwing it to the floor! In all cases the monitor malfunction resulted from mechanical damage to the instruments through being either dropped (two cases) or struck against a solid object (one case). Specifically, the damage took the form of traumatic shearing of the accelerometer balance arm rendering the instrument functionally useless.

The reasonably fast repair time for the monitors (about a month) avoided major delays in the project.

Given these problems the aim of monitoring at least nine of the children was thwarted. The results derived from the activity monitor data therefore represent the activity patterns of five children and one of these provided a halved data set.

(4) Monitor Placement Issues

Several factors influenced the choice of where to wear the monitor: age and size of the child, degree of over- or under-activity and, most importantly, the child’s preference.
In this study the eldest three subjects wore their monitors on their wrists and the youngest (and most active and disabled) on their upper arms. Once placement site was decided all of the children continued to wear the monitors in the same position throughout the programme in order to obviate the confounding variable of changing monitor position.

3.1.4.2 Using the Monitor Data to Assess Sleep Parameters

In this section the data presented are those obtained from investigator Sj's dataset unless otherwise stated.

(a) Percentage of Movement During Sleep

When percentage of time windows during each night which contain 1+ movement (the "Movement Index" - MI) is used as an index of quality of sleep (higher percentage movement corresponding to a poorer quality of sleep and vice versa) the night-by-night changes in this parameter for the group is expressed graphically in figure 16:-
It can be seen that there are no major differences in the percentage mean percentage movement scores in the three stages of the study (horizontal lines): Pretreatment: 11.84% (s.d. 4.38); Treatment: 10.78 (s.d. 3.05); Four-Month Follow-Up: 10.44 (s.d. 2.38), however the mean value of the last phase is marginally significantly less than that for the initial phase (p<0.05; ANOVA). These figures are very similar to those obtained by Middelkoop and Kerkhof (1990) who used the same type of monitor to evaluate sleep quality in cohorts of young and older adults. These data are further analysed below in order to allow more meaningful interpretation of movement pattern changes during the study for each monitored child (figure 17):

Marked inter-subject variation is seen and these figures do not correspond directly to their overall treatment outcome. The rank order of these changes does not vary significantly. These data were influenced by variable total sleep period lengths (the children were tending to be in bed for less time before the intervention) and also the durations of their night-waking episodes in each phase which will be described below. Furthermore, the data figures do not reveal how much movement per "active" sampling period (time-window) was occurring. This lack of clinical relevance of the "movement index" when used by itself as a measure of sleep quality supports Middelkoop and Kerkhof’s (1990)
(b) Activity Within and Duration of Night-Waking Episodes

Analysis of the information provided by visual inspection of the activity monitor data allowed the calculation of mean values for the duration of night-waking episodes and also the mean activity levels per minute during these episodes (a general assumption made in deriving the latter data was that the great majority of movements were assumed to have occurred in the periods of discrete night-waking - a separate analysis of the data revealed that at least 95% of activity did indeed occur during these periods. The results of these analyses are displayed graphically in figures 18 and 19:

**FIGURE 18**
Mean Duration of Night Waking Episodes (Minutes)

**FIGURE 19**
Movements Per Minute of Night-Wakefulness
The mean waking episode duration for the group in the pretreatment phase is 55 minutes and this tends to increase slightly during the treatment phase but to reduce markedly by the four-month follow-up stage by an average of 15 minutes which, when compared to the pretreatment figure, represents a reduction of nearly a third.

For the activity levels per minute of night-waking the trend revealed is an initial fall of 32% compared to the Pretreatment level is seen over the Treatment period. However, at the Four-month follow-up phase this falls back to a 10% difference.

Summary: The trends seen within the data are for the waking episodes to be less frequent, of shorter duration and to contain slightly less activity levels when compared to the pretreatment pattern.

(c) Time Spent Awake per Night

The average time the five monitored children were spending awake per night in discrete night-waking episodes was calculated by multiplying the mean duration of night waking data with their corresponding night waking frequencies in the three phases of the study. This analysis revealed that the children were waking for 51.5, 21.8 and 17.0 minutes, on average, in the pretreatment, treatment and four-month follow-up phases, respectively. This represents an approximate reduction of waking time of 60%. These results compare to Durrand and Mindell's findings (1990) when they treated a repeatedly night-waking fourteen-month-old girl with behaviour modification and analysed time-lapse videotape records of night time activity for objective evidence of treatment effects. These workers observed a 63% reduction in the child's night-waking frequency at their one-month follow up and this was sustained at later stages.
(d) Night Waking Patterns After Initial Sleep Onset

The "ACTPLAN" analytical software allowed accurate timing of night-waking episodes. The three graphs displayed in figure 20 show the waking patterns of the five monitored children in the three monitored phases of the study. The wakings were timed using the reference point of initial sleep onset. The rates shown are expressed in terms of the number of wakings per 15 minute period after initial sleep onset.

**FIGURE 20** Monitor-Derived Waking Patterns During the Night After Initial Sleep Onset: 5 Children; 189 Nights in the Three Study Periods

![Graph showing waking patterns](image)

The *Pretreatment* phase graph shows an initial peak after ninety minutes with successive peaks at around the same frequency. Relatively fewer wakings occur in the second quarter of the night but a steady increase is seen during the third quarter diminishing again in the final quarter. During the *Treatment* phase a marked overall reduction of wakings occurred compared to the baseline levels (79.8%). There is relatively less activity in the first and last quarters of the night however the same 90-minute recurring pattern of waking peaks is seen. In the *Four-month Follow-up* data phase there is an overall slip-back
towards the Pretreatment levels however the overall reduction is 48.9% of the pretreatment level. Compared to the previous graphs relatively more activity is seen during the second quarter of the night with lower levels in the second.

The next graph (figure 21) shows the pattern obtained when all of the night-wakings are combined.

![Figure 21: Night Waking Patterns; All wakings over study period Monitor Data: n=5 (189 nights combined)](image)

The peaks diminish in size as the night progresses and there is a corresponding increase in the trough levels thus there is a reduced amplitude of night-waking frequency. The cyclical pattern of night-wakings is more obvious in the combined data graph. Autocorrelation analysis confirmed the ninety-minute (i.e. six times fifteen-minute) frequency of these phasic changes. This pattern corresponds to the deep/light sleep change transitions which occur during normal sleep and the great majority of the children's waking episodes tended to occur around these points. This trend became less marked towards the end of the sleep period where sleep tends to be normally generally "lighter"
and during which spontaneous waking is more likely. This sleep rest/activity pattern periodicity was also revealed in Middlekoop and Kerckhof’s (1990) AM monitored cohort of non-retarded adult subjects who were not demonstrating frequent night wakings but who, nonetheless, did exhibit most of their sleep-related movement around these temporal points throughout their sleep periods.

3.1.4.3 Comparisons Between the Diary-Derived and Monitor-Derived Data for Certain Sleep Problem Parameters

This section of the results will relate specifically to three principal parameters of the children’s sleep problems: night-waking frequencies, night of the week effects on night-waking frequencies and night-settling times. Correlational data for these and two other parameters (morning rising and total sleep time estimations) will then be presented. This will be followed by a final section which shows the results of a correlational analysis between the two independent raters (DB and SJ) of the monitor data for specific sleep problem parameters: night-by-night movement indices and night-waking frequencies.

(a) Monitor and Diary Recordings of Night-Waking Patterns

Night waking frequencies were assessed by means of parental reports in the sleep diaries and, in five subjects, the activity monitors. The night time activity monitor data were inspected visually by DB for periods of activity occurring after periods of sustained inactivity (reflecting sleep). The definition or algorithm employed to characterise a period of active wakefulness employed in the study was: "a discrete period of sustained activity
of at least five minutes duration and registering activity of at least 50-unit intensity in at least one time window (of 36.5 seconds) during this period". The following graphs show the plots for both the monitored and sleep diary records over the three main stages of the study on a night-by-night basis (figure 22) and the mean overall values (+ SEM) for the night-waking frequencies for each phase (figure 23):

(NB: one patient was activity monitored for only the first week of each period therefore the activity means for the 8th. to 14th. days of each period represent those for four patients only). The graphs show very obvious negative steep slopes around the active
treatment period suggesting a treatment effect detectable by both monitoring methods. Despite a trend for the monitor data to show slightly fewer wakings in the first two phases and slightly more at the final phase compared to the parental diary reports, analyses of variance did not reveal significant differences in the rates between both of the methods except for those of the Four-month Follow-up phase (see figure 23) where the monitor data shows significantly more wakings compared to their equivalent diary entries (p< 0.05; ANOVA). The diary data reveals an overall reduction of night-wakings of 60% and the AM data, 49%; the changes in the rank order of night-wakings for each child in each condition were significant in both modalities (Diary data: Fr= 8.4, p< 0.01 and AM data: Fr=6.4; p< 0.05). The correlational data concerning these two techniques are shown below.

(b) Day of the Week Effects

When the wakings from the three study phases are combined and plotted for the two assessment modalities (figure 24), the diary data shows the Saturday "dip" similar to that seen in the graph for the larger group in figure 15 (an observation which supports the view that the monitored group were similar to the rest of the group).
The totals in the monitor-derived wakings data is far less clear cut although there is a loose trend for waking levels to be higher in the first half (Sunday to Wednesday) compared to the latter half (Thursday to Saturday).

Thus the monitor data shows partial confirmation of a trend detected in the larger-group diary night-waking data for an overall reduction of night-waking over the latter part of the week reaching a nadir on Saturdays rising sharply on Sundays to a midweek peak on Tuesdays and Wednesdays.

(c) Diary versus Monitor Estimation of Time Taken for Children to Fall Asleep from Bedroom Settling Time (Sleep Latency)

The parents of the monitored children estimated the time their children took to fall asleep after settling in their bedrooms. The mean values (plus standard errors) for the diary and monitor data concerning this parameter are shown in figure 25:
These data reveal that these five children were taking an average of over an hour to get to sleep every night confirming their parents' claims at the initial assessment session. However, when these data are compared to the corresponding diary data they reflect that the children were settling ten minutes earlier. A pattern of shorter sleep latencies compared to the Pretreatment (baseline) values are seen in the Treatment phase data in both assessment modalities. At the Four-month Follow-up phase the monitor data show a trend for the children to take longer to fall asleep compared to parent's diary reports however this does not reach statistical significance ($p < 0.07$; ANOVA). Over the course of the study there were reductions of the children's sleep latencies of 75% and 43% from the diary and monitor data sets, respectively. The rank orders of changes in the sleep latency data over the three study phases were the same for both assessment measures ($F_r = 6.6$, $p < 0.05$).

(d) Correlations Between Diary and Monitor Data

Data were derived for the night-settling times, total sleep period and also the night waking frequencies from the sleep diary entries and the activity monitor data for the three principal periods of the study on a night-by-night basis. Table 12 shows the correlational data between these two measures:
Table 11: Diary and Monitor Data Correlational Data

<table>
<thead>
<tr>
<th>Sleep Parameter</th>
<th>Pretreatment Phase*</th>
<th>Treatment Phase*</th>
<th>4/12 Follow-up Phase*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Night-Settling Time:</td>
<td>0.83</td>
<td>0.87</td>
<td>0.98</td>
</tr>
<tr>
<td>Total Sleep Period:</td>
<td>0.84</td>
<td>0.46</td>
<td>0.93</td>
</tr>
<tr>
<td>Night-Waking Episode:</td>
<td>0.02</td>
<td>0.28</td>
<td>0.50</td>
</tr>
</tbody>
</table>

* Spearman's Correlation Coefficient

Night-settling time information is used to derive the total sleep period therefore it is not surprising that they correlate very closely; they are also derived from data which occur when the parents are most alert to their children. The accuracy of parental reports of night-waking episodes is compromised by several factors such as parental sleepiness and a tendency to enter NW data in multiple-night blocks, for example, which might explain the poor correlation for night-by-night assessments of this parameter when these are compared to the monitor-derived data. Nevertheless, a systematic error in their reporting patterns probably explains the close comparability between the mean scores of night-waking frequencies seen in figure 19 for the pretreatment and treatment rates. At the four-month stage the closest but only moderate correlation between these two measures of night-waking episodes is seen yet this is the period where the mean scores are marginally significantly different from one another.

(e) Monitor Data Inter Rater Reliability

In order to establish whether the various monitor data algorithms were sufficiently robust to support the reliability of the monitor-derived data analyses and also the applicability
for further study, the principal investigator's (DB's) data were compared to those of an independent investigator (SJ) who analyzed the data using the same parameter algorithms but without the case notes and the parents' diary data. The principal monitor data parameters compared were: (i) Nightly Movement Indices (that is the percentage of time windows containing 1+ movement per night) and also (ii) Night-Waking Frequencies. These results are shown in table 13.

Table 12: Correlational Analyses Between Raters DB and SJ

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Pre-treatment Phase*</th>
<th>Treatment Phase*</th>
<th>4/12 Follow-up Phase*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Movement Indices: % movement/night</td>
<td>0.74</td>
<td>0.62</td>
<td>0.76</td>
</tr>
<tr>
<td>Night-Waking Frequencies</td>
<td>0.61</td>
<td>0.66</td>
<td>0.60</td>
</tr>
</tbody>
</table>

* Spearman's Correlation Coefficient

This analysis suggests a moderate inter-rater reliability for these parameters of the monitor data and thus supports the robustness of the simple algorithms employed to derive them (all of the other parameters investigated in this study either depend directly on these factors or are themselves used to derive these specific parameters). SJ's interpretation of the monitor data was carried out without reference to the relevant diary data or patient notes therefore monitor artifacts may have compromised the reliability of her findings however this possibility was not pursued further.

3.1.4.3 Summary

The results obtained from the data collected from the five monitored children show trends
towards improved sleep patterns similar to those revealed by their diary data in terms of reduced night-waking frequencies, improved sleep efficiency and less night-by-night variation.

3.2 THREE CASE VIGNETTES FROM THE STUDY

3.2.0 Introduction

Up to this point the results have been presented in terms of trends observed within the whole group or among those whose movement patterns were successfully monitored. However, this approach, of necessity, obscures the differences in response of the individual children and the challenges to the therapist encountered when attempting to implement the treatment programme with a heterogeneous sample of children and their families. In order to illustrate this clinical process three of the cases involved in the study were selected at random and will now be presented in a concise form.

3.2.1 Case I: Mark

At the start of his treatment Mark, whose learning disability was the result of Down's syndrome, was 3.5 years old. He was referred by the family's Specialist Health Visitor because of his life-long refractory NS/NW problems which were causing his middle-aged parents extreme stress. Previous help included behavioural advice and also a prescription for sedative medication which had not helped at all. Most recently, his father who was a self-employed ice cream vendor, was so sleep-deprived as a consequence of Mark's nocturnal demands for attention that he fell asleep at the wheel of his van and collided
with a tree which effectively put him out of business for a week. This situation was made worse by the necessity for Mark having to sleep in his parents' bedroom owing to overcrowding which provided him with ready access to his parents' bed which he managed to obtain after long battles most nights of the week. He would also take up to two and a half hours to settle in the evenings. At the initial assessment his parents rated his sleep problem at: 8.5/10 and the HBS rating was 9/9. His mother's sleep disturbance was 5/11 and her MI score: 12/24. Mark had several daytime behavioural problems scoring 35/64 on the BPI which included a dangerous tendency to run away from his parents when their vigilance lapsed and he also was wont to take all of his clothes off when thwarted at home and at his day nursery. His parents accepted the terms of the study and also agreed for him to wear a monitor during the second week of his Pretreatment phase two events occurred: the family's pet dog, whom Mark was very fond of, died suddenly; additionally, Mark developed chickenpox. Nevertheless at the beginning of the Treatment phase he had recovered from this and his parents agreed to start the programme. There was nowhere else for him to sleep in the house other than his parents' bedroom therefore emphatic limit-setting was of particular importance for his parents. A suspended heavy blanket was used to screen him from his parents so as to limit his awareness of them and also provide a physical barrier to reinforce their limit-setting. The family did not require more than the usual three additional telephone contacts and implemented the programme with alacrity. At the two-week post-treatment phase the scores of his various outcome measures were as follows: parent assessment of sleep problem 0/10; HBS sleep-item score: 4/10; BPI: 21/64. In addition to these improvements his parents noted that he appeared generally calmer and self-controlled: for the first time he was observed to pick up one of his picture books and become engrossed in it without being prompted to do so. His temper outbursts had reduced in frequency and he had not
run away from his parents at the supermarket and elsewhere. His mother's "stress" indices fell (MI: 1/24 at two weeks and 2/24 at four months) and her sleep quality improved more slowly (6/11 at two weeks and 11/11 at four months). Also his father reported improved sleep and feeling more alert during the day. At both the Four- and Eighteen-month Follow-up phases, Mark's sleep pattern and other behavioural indices remained satisfactory and no additional input from the child and adolescent psychiatry service was required. His parents rated their satisfaction with the treatment approach as being 10/10 and considered it to be "just right" for their son.

(Mark's activity monitor data is shown in the Appendix, pages 199 to 202)

3.2.2 Case 2. Sophie.

This girl who suffers from Smith-Magenis syndrome, was also referred by her Specialist Health Visitor as having severe and refractory of NS/NW problems. The referral letter also expressed concern about her mother's ability to cope with her child's demands as a consequence of her having a cardiac pacemaker. Additionally the mother was due to appear in court on cheque fraud charges. Sophie presented as a strong-willed, overactive and demanding girl who required bilateral hearing aids to enable her to hear and understand the simple demands made of her. Her mother appeared desperate to receive any help admitting that she often experienced the urge to physically chastise her daughter but had, to date, resisted this. She was experiencing particular difficulties during the summer holidays which was the time of this initial assessment. She was supported by her husband however it was readily apparent that there were very evident inconsistencies between their styles of parenting: her father readily gave into her frequent and unpredictable demands for attention in the evenings and during the night which were
usually accompanied by shrill screaming fits. The pretreatment assessments of this girl's parameters were as follows: sleep problem score: 7/10 (parents); 8/9 (HBS). Maternal stress: 8/24 ("very stressed" on the MI) and maternal sleep quality: 6/11. Sophie scored 26/64 on the BPI scale. The household routines were generally chaotic and the family's dog (a mature mastiff) was also ill-disciplined: a picture which is often encountered in such families.

Sophie's parents were prepared to accept the terms of the study and during the Treatment phase required two extra telephone contacts in order to emphasise the importance of the parents supporting one another and to resist their daughter's, sometimes extreme, nocturnal demands. After the treatment had commenced some general improvements were noted in that the night-settling time had reduced to around a quarter of an hour compared to over an hour before treatment and Sophie's waking frequency had almost halved. Her parents rated her sleep problem as 5/10 however on the HBS she scored 2/9. Her mother's MI score had fallen to 1/24 ("non-stressed") which supported the investigator's impression also her subjective appraisal of her sleep quality had improved to 10/11. Sophie's daytime behaviour had improved slightly and this was particularly the case for her demeanour in the mornings. Over the ensuing four months her mother's general health improved following the recalibration of her pacemaker, she was declared innocent of the fraud charge and Sophie had returned to school. However at the Four-month follow-up stage her daughter's sleep pattern had deteriorated slightly (parent assessment: 6/10; HBS: 3/9) which merited a review of their management which merely involved a restating of the original principles of the programme. Her daytime behaviour had continued to improve however her parents attributed this as being primarily the result of her being back at school and her reduced BPI score of 20/64 reflected this. At the Eighteen-month Follow-up stage they rated her sleep problem as 4.5/10 and were moderately satisfied with
the treatment programme (satisfaction score: 7/10) stating that agreeing to back one another up at bedtime was the most important therapeutic factor. They requested further help with Sophie's troublesome daytime behaviour which had deteriorated again over the previous six months.

3.23 Case 3: Ahmed

Ahmed's mother had asked the family's paediatrician to be referred to the treatment programme having heard about it from her Health Visitor. Ahmed, an eight year-old, mixed-race, Anglo-Arabic boy was one of a pair of dizygotic twins. His profound learning disability and severe cerebral palsy resulted from anoxic brain damage at birth (his female twin was unharmed). His British mother had returned from Saudi Arabia some three years previously following the irretrievable breakdown of a chronically abusive marriage to an Arabic businessman. She had sought previous help for Ahmed's life-long severe NS/NW difficulties from a clinical psychologist whilst living in Bristol having previously explored other treatment approaches which included night-sedation which had not helped at all (he developed an acute paradoxical reaction to this medication). The psychologist commenced a "graded-changes" approach (see the 'Introduction') which achieved little success over the nine months it was tried. After this the family, (Ahmed, his mother, his twin and his two older sisters) moved to Leicestershire in order to further distance them from their allegedly extremely vengeful father. At initial assessment his mother scored his sleep problem at 9/10 and the corresponding HBS score was 9/9; Ahmed slept on a mattress in the lounge and required children's videos to soothe him into sleep at night a process which would take up to several hours and he would commonly wake several times during the night and scream.
for his mother's attention (which he invariably received). He suffered from reasonably well-controlled generalised epilepsy, his last seizure being six weeks before presentation. Despite his limited mobility he scored very highly on the BPI (42/64) particularly in the areas of aggressiveness, tempers and poor habits. His mother appeared both chronically exhausted and clinically depressed scoring 13/24 ("very stressed") on the MI describing an unsatisfactory sleep quality (5/11) with daytime sleepiness being a particular problem. She wrote a helpful account of her son's sleep problems and their effects on her and this included the following highly poignant passage:

"...what happens when a child doesn't sleep? When night after night, year after year, a child wakes, cries and demands attention? That's when exhaustion takes over and life becomes a grey limbo, all thoughts of the future forgotten, our only concern to survive another day. It erodes the fabric of life, causing depression and resentment both of which are closely followed by overwhelming guilt, especially when the child is [so disabled - DB]. Most parents who suffer this type of long term sleep deprivation must do so in silence, their optimism replaced by resignation”.

There were also some concerns about her eldest daughter's behaviour and mood which were a consequence of alleged racially motivated bullying at school which her mother was taking up with the school. Notwithstanding these problems, the whole family appeared keen to help Ahmed learn a more acceptable sleep habit and the programme was started. After the two-week Treatment phase his sleep problem had virtually disappeared (mother's rating: 1/10; HAS: 0/9); his daytime behaviour had improved and his mother's mental state had also improved considerably (MI score: 2/24) and her sleep pattern virtually normalised for the first time since Ahmed's birth (9/11). She described the most
important factors underpinning these changes as being her learning to ignore her son's cries for attention and to "wind him down" before bedtime. These changes were substantially maintained at the Four-month Follow-up phase and this was particularly the case for his daytime behaviour (BPI score: 25/64). However, at the Eighteen-month review stage his sleep pattern had deteriorated over a short period prior to the assessment owing to a deterioration in Ahmed's epilepsy control secondary to mastoiditis for which he had commenced antimicrobial therapy. All of the other principal outcome measures were satisfactory from his mother's point of view and this temporary exacerbation did not warrant further psychiatric intervention. She believed that the style of intervention was "just right" for her son's erstwhile sleep problem and rated her overall satisfaction with the treatment's outcome as 10/10.

3.2.4 Comment

These three cases show that the chronically sleep disordered severely learning disabled child can be helped to achieve a more acceptable sleep pattern by means of the behavioural modification package described previously notwithstanding a range of other co-existing individual and family problems and also the wide range of adaptive ability deficits demonstrated within the affected group. The core approach employed in this study appeared to have helped all of these diverse families to a significant degree without requiring that any significant alterations or amendments be made to it. Furthermore, no negative attitudes towards the treatment approach on the part of the children's parents were obtained.
3.3 SUMMARY OF PRINCIPAL RESULTS

Table 13 provides an overview of the study's principal results:

Table 13: Summary of Overall Results

<table>
<thead>
<tr>
<th>PARAMETER</th>
<th>RESULT</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Sleep Problem Severity:</strong></td>
<td>1. Marked reductions by both parental and investigator assessments.</td>
</tr>
<tr>
<td></td>
<td>2. Night-by-night ratings show maximum reduction over treatment contact period.</td>
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<tr>
<td></td>
<td>3. Results generally sustained at both long-term follow-up points despite no further therapist input.</td>
</tr>
<tr>
<td></td>
<td>4. Only one child demonstrating a &quot;major&quot; co-sleeping problem at four-months compared to eleven prior to treatment.</td>
</tr>
<tr>
<td><strong>Generalisation Effects:</strong></td>
<td>1. Reduction in daytime behaviour problem rates and also individual problem severity.</td>
</tr>
<tr>
<td></td>
<td>2. Improved maternal wellbeing (stress and sleep quality); improvements occurring generally less quickly than those of their children's sleep problems.</td>
</tr>
<tr>
<td></td>
<td>3. Individual reports of improvements in cognitive skill parameters but no specific skill domain improvements demonstrated in the group.</td>
</tr>
<tr>
<td></td>
<td>4. No adverse effects reported.</td>
</tr>
<tr>
<td><strong>Consumer Satisfaction:</strong></td>
<td>1. High levels of satisfaction expressed at both initial and four-month follow-up points.</td>
</tr>
<tr>
<td></td>
<td>2. Treatment approach generally helpful and acceptable.</td>
</tr>
<tr>
<td></td>
<td>3. A minority of families (three) requested further child psychiatric help at eighteen months.</td>
</tr>
</tbody>
</table>
### Activity Monitor Data:

1. Confirmed general trends seen in the diary data and the positive influence of the treatment contact period.

2. Provided novel information about the changes in duration and temporal distribution of night-waking events, activity levels during these episodes and other quantitative measures of sleep problem parameters which diary data did not reveal.

3. Generally moderate inter-rater reliability of monitor data parameters but more variable for monitor/diary data.

4. High rate of monitor failure resulting in loss of valuable data in three children.
4: DISCUSSION
Section 4: DISCUSSION SECTION

4.0.1 Overview of Discussion Section

The discussion of the various findings of this thesis will be presented in five major sections:-

(1) Effectiveness of the treatment
-what appeared to be happening; other possible explanations and influences for the changes observed; patterns of change and possible mechanisms of change.

(2) The satisfaction of the caregivers with the treatment approach
-why they were satisfied and how this came about with particular emphasis on the style and safety of the intervention tested.

(3) Generalization effects of the treatment
-how and why these happened in terms of the children's and also their parent's reactions to the intervention.

(4) The utility of the activity monitors in the study
-where monitors can add to the understanding of the processes of sleep problems and their resolution; the failure of monitoring attempts in several children and the practical implications of this; directions of further research using monitors.

(5) Implications for further work
-resource and clinical service implications; comparisons with other techniques; working with older sleep-disordered children and adults; directions for further clinical research; final conclusions.
4.1. EFFECTIVENESS OF TREATMENT

4.1.0 Introduction

In small cohort studies which employ multiple outcome measures statistically "significant" changes in one or indeed several of these might be expected by chance; however, over the course of this study directions of change in all of the principal measures were consistently towards improvement: sleep problem severity, night-settling and waking patterns, indices of generalization effects and, also, consumer satisfaction. The results are very similar to those of the few studies which have addressed NS/NW problems in developmentally delayed children as well as those conducted with non-retarded children (Hewitt, 1985; Richman et al., 1985; Quine & Wade, 1991) Activity monitoring provided confirmatory evidence of some of the treatment effects that were observed by the investigator and also the children's parents.

In order to ascertain whether these effects were principally attributable to the treatment approach itself or explicable through other possible processes, these questions will now be examined.

4.1.1. Alternative Explanations for the Changes Observed.

(a) Regression Effects: This is a statistical issue; when a target parameter is rated initially and if it is extreme a later rating is likely to show spurious improvement purely as a natural consequence of the variability within the ratings. This is also referred to as regression to the mean. Such inaccuracies can result from limitations in the reliability and
validity of the test instruments themselves. In this study the cohort was selected on clinical grounds in addition to instrumentally-derived criteria. The results obtained from the diary data do not suggest that this process was operating to a significant degree because the trends observed in the results derived from more objective outcome measures (particularly the monitor data), were very similar to those seen in the diary data. Also, when observable changes were detected in the various outcome measures assessed on a time series basis, they tended to correspond to when the intervention was introduced and closely matched clinical assessments of improvement. Furthermore, the high levels of parental satisfaction with the results of the intervention reject the notion that only spurious improvement had occurred.

(b) "Hawthorne" Effects: These are said to occur when there is a tendency for raters reliably to reflect accurately what is actually occurring owing to a desire to see improvements. In the case of this study, the principal raters (DB and the children's parents) were not unaware of the aims of the treatment and at what point in the course of the study it was supposed to start. Practice effects could also influence this. While it is possible that these influences may have occurred the consistent trends seen in all of the major outcome measures including the objective monitor data would suggest that additional real effects were occurring. Furthermore, these changes were substantially maintained at follow-up. The only means of testing the power of these influences would have been to employ a control group of untreated subjects who would be rated in the same way as the treated children. Independent raters who were blind to the status of the children would also have to be used.

(c) Natural Trend Towards Improvement: At the time of initial assessment and induction
into the treatment programme all of the families could have been described as being in a state of crisis and the severities of their children's sleep problems were at their peak; indeed, this factor was undoubtedly the principal reason for enrolling their affected children into the programme. The only direction of change in the sleep problems available to these families would have been towards improvement and, as has been shown, this occurred in all of the children albeit to varying degrees; As such a "ceiling effect" could also have been operating, however, it is highly unlikely that this improvement would have occurred without the specific treatment intervention. The large follow-up studies of sleep disordered retarded children which have been discussed previously demonstrated that severe NS/NW problems, if left untreated, tend to run a chronic course, usually over several years and sometimes continuing into adulthood (Hogg & Lambe, 1988; Quine & Pahl, 1989); therefore, it would be highly unlikely that an heterogeneous cohort of fifteen of such children who all demonstrated lifelong and severe forms of such a disorder would improve spontaneously over a period of just a few weeks and at the precise time of the intervention. The various parameters of the sleep problem which were assessed on a night-by-night basis by the children's parents and, additionally, in a third of the children, by means of the activity monitors, showed an observable trend towards improvement during the Treatment phase of the study compared to their baseline values. Throughout the Pretreatment baseline no general trends towards improvement were seen in any of the sleep problem parameters. However, from the general patterns observed in the results, trends towards improvement in these sleep problem parameters for the group commenced over the periods when there was direct therapeutic contact between the therapist (DB) and the affected children's parents or principal carers. These corresponded to the nights following the initial treatment sessions and their subsequent daily telephone follow-ups (nights 15 to 18). After this period the trends were generally sustained. Nevertheless, it
must be acknowledged that a minority of children (four) showed some degrees of improvement in some problem parameters before this stage during their Pretreatment, "baseline", assessments; however, they too showed further improvement during the active Treatment phases. (The recognised phenomenon of improvement in target behaviour by merely keeping a diary record of it is discussed in the "Introduction" of this thesis.)

(d) Influence of Non-Specific Therapeutic Effects: In addition to the factors influencing treatment outcome which were intrinsic to the treatment approach and specifically evaluated during the study, it is also necessary to acknowledge the influence of several non-specific factors which are known to operate in therapeutic interactions. These include: (i) an expectation on the part of the children's parents of successful resolution of what hitherto had been a intractable problem by attending a "specialist" treatment clinic; (ii) a congruence of the therapeutic approach to the children's parents' views of the presenting problem; (iii) explicit discussion of what can be expected in terms of the children's reactions to the treatment; (iv) a communication of the therapist's enthusiasm to help the families with what he believed to be an effective treatment; (v) the therapist's capacity to "sell" the intervention successfully to the families; and (vi) the ability of the therapist to both acknowledge and also contain carers' anxieties over the course of the study and particularly at the beginning of the Treatment phase in this specific study. While it is entirely probable that all of these factors were operating during the study albeit to varying degrees in each case it was perhaps surprising that none of the carers or parents mentioned any of them specifically when given the opportunity to describe what they felt were the important components of the therapeutic approach (see "Results" section). Perhaps they were operating too subtly to be cited or, alternatively, such considerations were eclipsed by the perceived importance of more concrete factors. Frank and Frank
(1991) discuss how these non-specific, essentially placebo-effect enhancement, factors operate in all successful psychotherapeutic situations. Despite their apparent importance they are often overlooked when trying to explain how treatment "works" yet they appear to provide the necessary conditions for therapy to be effective within, providing essentially a favourable therapeutic context or therapeutic alliance between service-users and therapists. Nevertheless, in this study it is highly improbable that these influences, in themselves, were responsible for the treatment effects. There may be various reasons why this might be so but probably the most powerful argument against this proposition is that ten of the affected families had already received prior professional help in which, presumably, some or, indeed, many of these important non-specific factors would have operated yet this help had proved to be generally unsuccessful up to the point of enrolment into this study. However, it must be conceded that actively fostering these non-specific treatment influences is an important part of everyday clinical practice when working with affected families.

4.1.2. Patterns of Improvement

Night-setting patterns tended to improve to a greater degree than those for night-waiting when these parameters were assessed objectively and this may reflect an intrinsic tendency for these children to wake during the night which is only partially amenable to the simple behavioural modification approach employed in this study. As has been discussed previously children vary greatly in their waking frequencies. It is what they do when they awaken (talk, scream, self-injure, etc.) which defines the presence or absence of a problem (Scott & Richards; 1993). Nevertheless, despite the finding that some of the children continued to awaken regularly during the Four-month Follow-up phase there is some
evidence that they were more able to settle themselves without requiring their parents' presence or other, less direct, forms of attention. In support of this proposition is the monitor data relating to night-waking which shows that although the frequency of waking tended to fall back somewhat to the Pretreatment phase level when compared to the Treatment levels, the overall levels of activity during these episodes remained lower, however, in order to have assessed accurately what the children were actually doing when they awoke would have required that they were videotaped throughout the night. Furthermore, at the Four-month follow-up the mean duration of the discrete NW episodes tended to be nearly a third shorter and the average time spent awake per night reduced by nearly two thirds. The monitor data reflects that the parents themselves probably tended to under-report these episodes which might reflect either an enhanced capacity for them to ignore their children's cries or, alternatively, that the children did not need to cry out when they awoke and managed to settle themselves. The first proposition is supported by the fact that the main thrust of the intervention was to encourage parents to ignore and thus not respond to their children's unreasonable nocturnal demands; their reports of reductions in night-wakings could be a measure of their ability to manage to do this. Also, over the latter phases of the study the children's night-waking episodes tended to occur in the middle of their sleep period which would probably correspond to the times when their parents were most deeply asleep themselves and thus least likely to be responsive to their children's attention-seeking ploys. The waking patterns seen before the intervention period suggested that the children's wakings were more likely to occur when their parents were either still awake in the late evening or else during the latter part of the night when the parents were in "lighter" stages of sleep (stages 1, 2 and REM) when they were more likely to be awakened by their children's nocturnal disturbances. The changes in the children's physical proximity to their parents also helped in these respects:
before the intervention the majority eventually managed to co-sleep with their parents whereas after the treatment only one child did so and even she spent most nights of the week in her own bed only gaining access near her parents waking time. Thus, a successful physical night-time separation occurred between most of the children and their parents during the "active" treatment stage which probably in itself accounts for much of the reduction in the parents' responsiveness to their children (and, presumably, vice-versa). The role of parental proximity in maintaining children's sleep problems has been discussed previously in the "Introduction", the success of the intervention would appear to hinge primarily upon whether this factor can be removed or not. The children's parents themselves nominated the "ignoring" of their children's demands as being the fourth most important specific item in the advice audit exercise ("Results": table 8).

4.1.3. Conclusion

In summary, it is proposed that there were many changes occurring both within the children and their parents themselves and also within their mutual relationships in and around bedtime and these can help to explain the improvements in the families' sleep patterns. It is probable that these various explanations may all have some relevance however, as was the case for the non-specific influences, the degrees by which they operate varied on a case-by-case basis. Nevertheless, whichever specific or dynamic changes occurred they resulted in parental reports of a general improvement in their children's sleep problems. It is well recognised in the treatment of many adult sleep disorders and particularly the psychophysiological forms of insomnia that an improvement in the complainant's subjective appraisal of the problem is crucial for treatment success and this can often occur in the absence of any objective evidence (Espie, 1991).
4.2. SATISFACTION OF PARENTS AND CARERS WITH THE TREATMENT APPROACH

4.2.1. Parent's Views

The results show that although ten of the families received help for their children's sleep problems prior to the study, they reported generally low levels of satisfaction with this. Given that this study involved a rather small and selected cohort and also, by definition, in being so would not be attracting subjects who had received previously successful treatment, caution is necessary when drawing any firm conclusions about the quality of the previous interventions. Nevertheless, this finding does support the author's clinical experience (and also those of other interested clinicians) that such severe sleep problems are either not treated at all prior to initial presentation to child psychiatry clinics - as was the case in five of the families involved in the study or, if they are, they are very likely to have received only sedative medication. Other workers have commented on this unsatisfactory situation (Bartlett et al., 1986; Quine & Wade, 1992). In this study the children's parents universally condemned this practice and generally rated the effectiveness of this form of treatment as very low indeed, a finding which supports Bartlett et al's (1986) conclusion that sedatives tended to "...make matters worse" rather than help the sleep disordered learning disabled child. The parents of three of the children reported induced states of paradoxical excitability in their children resulting from sedative medication. As has been discussed elsewhere (see the discussion of drug treatment in the "Introduction"), this reaction has also been described by other commentators (Valman,
Psychological and, more specifically, behavioural modification assistance was reported to be both more helpful and also better tolerated by the children when compared to sedative medication. Despite this, a common criticism described by the carers who had received prior behaviour management advice was that it was often obtained from several different sources and, as a result, such advice was often contradictory. This lack of consistency between the advice offered particularly by general practitioners, health visitors and paediatricians engendered feelings of confusion and anxiety for the children’s parents. Another particular problem with the behavioural advice previously received by the families was that it was usually given without any firm follow-up arrangements having been made in the short-term which would allow the therapists and the families involved to review progress and, if necessary, adapt the treatment. This would also provide the opportunity to support the carers’ resolve to continue with treatment. This common omission probably rendered some of the prior therapeutic approaches more likely to fail. On the other hand, one parent had tried a particularly intense and detailed graded-changes approach (see case vignette #3 in the “Results” section) assisted by a clinical psychologist over several months. Unfortunately, few positive changes in her child’s sleep pattern were observed and a subsequent family relocation to another part of the country resulted in these minor gains being lost. The disappointment of parents facing continuing severe sleep disturbance after failure of professional help can only serve to further magnify their general despair and, perhaps in some cases, make them reluctant to seek further help in order to avoid further potential disappointments. The five families in the study which had not sought advice prior to presentation had been led to believe that no help was available for their children’s particular problems and that sleep disturbance was an inevitable consequence of caring for severely learning disabled children. This form of therapeutic nihilism was encountered by
several of the families in the study even from health care professionals. Comments such as "...well, what do you expect, your child's mentally handicapped?", for example, were reported and these specific attitudes were described by families as being particularly demoralising. Given that all of these families were in contact with a range of health, care and welfare agencies whilst they were experiencing such major specific difficulties with their children it is particularly regrettable that these problems were only presented for treatment when this study was initiated, following several years of distress for all of the families concerned. This situation reveals serious general deficiencies in the current levels of provision of effective psychiatric services for these families and also very particular deficiencies of health and welfare professionals' knowledge of the management of common sleep problems (see below).

4.2.2. Safety Considerations

Another reason for the high degree of reported overall satisfaction with the intervention technique tested in this study could be that there were no reported incidents of any significant sustained deteriorations in the children's behaviour and demeanour over its course. Indeed, quite the opposite effects were reported. It is well recognised that an initial deterioration in target behaviour is commonly encountered in behavioural modification programmes with children (Skiimer, 1953) and this specific "extinction burst" effect particularly when using rapid extinction approaches (White et al, 1972). In this study parents were explicitly warned of this possibility (see "Method"), although only minor and transient exacerbations were reported and these did not influence the trends seen for the children as a group. More seriously, behaviour modification interventions are known to run the risk of inducing negative changes in children's behaviour which may be
sustained and require further treatment in their own right (Graziano, 1977), such developments can result in parents opting out of treatment, and, what's more, become wary of soliciting help at a later stage. A minority of the parents and carers involved in this study had experienced this through previous therapy however the paradoxical reactions to drug treatment rather than to failed prior behavioural interventions that three of the children had experienced previously (see above) appeared to be the most potent determinants of induced negative reactions on the parts of parents manifesting in terms of delays in requesting further help with their children. In the sparse literature concerning rapidly acting behavioural modification approaches to night-waking and night-settling problems which are similar to that used in this study, no significant negative effects have been reported (Sanders, Bor & Dadds; 1984). Nevertheless, in clinical practice the author is aware of a severely sleep disordered child with profound physical and learning disabilities whose parents ignored her nocturnal distress and breathing difficulties during treatment who suddenly died during the course of treatment although the treatment itself was not regarded as having contributed to this. Therefore it is salutary that extreme care is taken when using this technique to ensure that parents understand the advice and, most importantly, when it is appropriate to respond to their sleep disordered children's cries. A written advice sheet is now used by the author in his clinical work as an aide memoire for the parents which strongly emphasises the need for parents to attend to their children should they suspect the emergence of medical problems during the course of treatment and to only recommence the programme when their child recovers. Before any such treatment commences with such families, they have to demonstrate that they know the difference between their children's distress resulting from physical illness and that shown when they are demanding parental attention inappropriately. The author would also commend this practice on medico-legal grounds.
4.2.3. Treatment Style

The style of the treatment approach employed in this study falls midway on a continuum between the two major commonly employed therapeutic strategies for night-settling and night-waking problems. On the one hand, sedative drugs represent a very restricted interventional approach which does not allow for any individual variation other than adjustment of dosage: either the patient is given the medication or is not. On the other hand, the "graded changes" behaviour modification approach (see the "Introduction") is far more flexible and open-ended. It can be tailored to suit the individual needs of a wide range of families and allows the parents and, in some cases of older and more able children, to have an important influence upon the both the form and size of these changes and also with the choice of rewards which may be built into such a programme. In the latter case the therapist and family work together on a more democratic basis whereas the former approach effectively ignores the intrinsic capacities of families to work constructively towards a resolution of the sleep problems. The rapid-settling and extinction techniques employed in this study necessarily require a close working relationship with affected families and can tolerate some individual tailoring of the approach. However, the core principles of establishing bedtime routines, rapidly settling the children and, thereafter, ignoring their demands for attention (unless unwell) are, of necessity, non-negotiable. In the opinion of the author this style represents an economical happy therapeutic medium and is best suited as a first-line approach to these types of problem. Certainly the results of this study support the contention that it is at least as effective as the "graded changes" approach has been shown to be (see "Introduction"). Also, this technique has been demonstrated to be highly acceptable to parents and
principal carers upon direct enquiry (see "Results"): the majority believed the treatment approach to be "just right" for their children. Additionally, none of the families reported any feelings of disempowerment through this essentially more directive approach. On the contrary, although the parents acted as closely directed co-therapists they generally reported high levels of personal satisfaction with the results of their own efforts in changing their children’s sleep habits by implementing the programme although this factor was not explored in any systematic manner. What’s more, they were also happy to discover that their enhanced limit-setting skills at bedtimes usually generalised to several daytime settings (see above) and this probably contributed considerably to the improved daytime behaviour observed in most of the children after the intervention. Untoward effects were not detected, which is not the case with the use of sedative medication, and the benefits of the treatment, for the most part, were sustained at long-term follow-up. Thus it appears that the therapeutic approach which is the focus of this thesis has several advantages over the those that are currently most commonly used.

4.3 GENERALISATION EFFECTS OF TREATMENT

4.3.1. Introduction

It is well-recognised that generalization effects (i.e. changes of other parameters of psychological functioning within the subject, his family or wider social group which are not specifically addressed) can occur in psychotherapeutic interventions which may be attributable to the focus of treatment. This section will explore to what degree and through which processes generalization effects occurred as a result of the intervention employed in this study.
4.3.2 Effects Upon the Children

Given the necessarily focused nature of the treatment intervention, no other aspect of the children's or their parents' difficulties were addressed directly during the treatment phase of the study yet, as has been seen, significant positive changes in many parameters other than those relating to the children's sleep problems were observed. In the case of the children's improved general daytime behaviour this is possibly attributable to several factors:

(a) Enhanced Sleep Consolidation. It is postulated in this study that an improved sleep pattern resulting in greater consolidation of sleep (sleep efficiency) primarily results from the children learning to resist the urge to solicit inappropriate parental attention at bedtime and also during the night thus reducing the likelihood of becoming uncontrollably distressed and physiologically aroused, states which preclude both sleep onset and also its maintenance. They learn this primarily through their parents' non-reinforcement of their demands for attention: the only easy option left for them is to fall asleep again. The specific BPI daytime "attention-seeking" item showed the third highest improvement at follow-up which supports this proposition.

(b) Reduced Daytime Sleepiness for the Children. It is well understood that both adults and children require a relatively undisturbed night's sleep in order to function optimally during the following day (Horne, 1988). Sleepy children particularly tend to be sub-vigilant, irritable and, paradoxically, somewhat over-active and less amenable to limit-setting measures. However children who manage to obtain regular stress-free refreshing night's are thus more likely to be responsive to their carer's authority. The BPI daytime
"temper tantrums" and "overactivity" items score improvements support this proposition.

(c) Enhanced Self-Control. Another possible explanation of the improvements in the children's day-time behaviour is that their enhanced self-control during the night could also be operating during the day. Perhaps a necessary precursor of self-restraint and deferment of immediate gratification is the acquisition of a capacity to tolerate the temporary distress of separation from the mother during the night. Daws (1988) discusses the importance of carers and their children achieving a mutually satisfying sleep habit in terms of the theoretical principles of object relations theory and children's development of a sense of self; these themes are further explored in the "Introduction". Interestingly, four of the subjects were profoundly retarded with a developmental age of less than six months - a level below that considered necessary for the concept of object permanence to have developed which allows an infant to know that an object continues to exist despite being out of sensory contact. Whilst caution is necessary when interpreting the results obtained from the BPI, it is interesting to see that both the quality and quantity of "bad behaviour" items improved significantly as well as that for the "temper tantrum" item. Both of these results suggest enhanced self-control on the part of the children. The improvements in the "habits" and "sociability" items suggest a reduction in self-stimulatory activities such as rocking, using objects for twiddles and thumb or finger sucking in the former case and more interest in others and the outside world generally in the latter.

(d) Enhanced Sociability. The effects of chronic sleep disruption manifesting as daytime sleepiness, irritability and overactivity are probable important factors contributing to and also, perhaps, maintaining the relatively high levels of social impairment discovered in this group revealed in the children's "Vineland ABS" scores. Thus it seems plausible that the improved sleep quality per se can lead directly to improved day-time sociability although
the changes in this specific domain assessed by means of their "Vineland" scores did not reflect this: improvements occurred to a similar degree across all of the instruments' domains. In addition to this result the scores for the specific BPI "sociability" item concerning relationships with siblings and peers showed the highest overall item change per child at follow-up.

4.3.3 Effects Upon Children's Parents

The changes in maternal well-being can also be explained, at least partially, by their enhanced sleep patterns. The analysis of the Malaise Inventory data reveal that the items that changed most significantly were those most closely related to the consequences of chronically disturbed sleep namely difficulty in getting and staying asleep, feeling tired or depressed, waking unnecessarily early in the mornings, being easily upset or irritated, headaches and having a racing heart. These results are very similar in substance to those reported in Quine and Wade's (1991) intervention study. The chronic stress of repeatedly disturbed nights operating over several years was very apparent from the mothers' accounts of their problems. The 24-hour demands of their children became the focus for their families' routines and even when professional help was sought the results were, at best, short-lived which added further to their difficulties and sense of despair (see above). Following treatment, in addition to the results seen in the mother's sleep quality scores, most parents reported a lag in the improvement in their sleep pattern compared to the few days it took their children's patterns to improve. Several parents noted that they were continuing to wake up at the times their children were want to do prior to treatment which reflects the reciprocal interaction operating between the children and their parents' sleep habits. This tended to continue for a week or so. Once their patterns had improved (and
this took several weeks in some cases compared to their children's response of only a few days) a common report from these parents was that they felt generally more alert during the day and thus more able to meet their children's demands. Many of them also commented that their confidence in being generally firmer with their children was enhanced by their success with the bedtime limit-setting problems, problems hitherto regarded as being untreatable by most of them. It might be construed that the effects of improved and consequently more restorative sleep coupled with the parents' success in what was regarded previously as an entrenched set of problems resulted in both improved parental morale and also enhanced parental effectiveness. In addition to this, it is also probable that the improvements in their children's sleep quality made them calmer and thus more responsive to their parents' limit-setting during the day-time. Previously held "myths" involving the parents equating a firm approach to their children's behaviour with unfair punishment, for example, noticeably changed after the treatment phase of the study although this was not formally assessed.

The small cohort size used in this study precluded the use a formal statistical pathway analysis which might have revealed whichever factors at presentation predicted specific subsequent changes. Nevertheless, the results strongly suggest that it would be highly improbable that any such marked changes in the children's daytime behaviour, maternal well-being and, specifically, maternal sleep quality would have occurred were it not for the significant improvements in the children's sleep problems. Larger studies and especially that of Quine and Pahl (1989) have established the complex interplay of factors responsible for both the aetiology and maintenance of sleep problems in learning disabled children. A much larger study than that currently being discussed would have to be required to firmly establish what are important predictors of treatment response when
using the approach described in this thesis.

4.4 THE UTILITY OF THE ACTIVITY MONITORS

4.4.1 Limited Success with Monitoring

It was unfortunate that only five patients were successfully monitored over the three major study stages and one of these only partially so. The main reason for monitor failure was mechanical damage to the accelerometer resulting from the monitor being stuck against a solid object which caused shearing of the balance arm in all cases of monitor damage. It was known that the monitors were likely to sustain damage when exposed to forces in excess of 5kg. This degree of force can occur if the monitor is dropped onto a hard surface and would certainly be exceeded were it to be actively struck against such a surface. Therefore it would be wise when using these devices in any future studies which involve either very young or severely learning disabled children, that they are worn enclosed in a padded protective arm or wrist band in order to reduce the risk of such damage. The author would now recommend that a more robust device such as the water and shock resistant "Motionlogger" actigraph (Ambulatory Monitoring Instruments Inc., USA) should be used in further work with children’s sleep problems.

4.4.2 Monitor-Derived Data

The results of the study yielded interesting differences and similarities between the various
sleep parameters when they were assessed by means of both the parents’ diary reports and the activity monitors. The author assumes that the monitor data is intrinsically more accurate at assessing these parameters based upon the conclusions concerning the use of activity monitor data drawn from the review of the available literature presented in the “Introduction” section, however it must be acknowledged that certain important assumptions were made when interpreting the monitor data. For example: a major assumption employed when calculating the quantitative changes in movement within night-waking episodes over the course of the study was that almost all of the children's measurable motor activity during the night occurred within discrete night-waking episodes. Another assumption concerned the validity of the night-waking algorithm; however, this assumption was lent some support by the similarities between the frequencies of night-waking frequencies derived from the monitor data and those reported by the parents. Nevertheless, these assumptions operated throughout the study period and yielded data which was consistent with the changes reported by the parents and observed by the investigator.

The monitor data also yielded information concerning aspects of the quality of night period activity patterns which simple retrospective diary data would not allow, for example, the detection of real-time activity patterns during the nights and records of night-waking episodes occurring while the parents were asleep. Changes in these patterns appear to have been influenced, at least in part, by the treatment intervention.

The night-waking frequency data reveal a slipping back towards the Pretreatment baseline value after an initial improvement over the Treatment phase. This trend is not seen in the comparative diary data and may represent intrinsic tendencies to night-wake operating in these children however they are probably modified by the treatment intervention in terms of what the children actually do when they awaken (see above). Marked changes were
discovered in terms of precisely when the children were tending to wake in reference to their initial sleep onset times: before treatment they were tending to wake principally around the first NREM to REM transitional point. Following treatment this pattern shifted towards the middle of the night and this was sustained at Four month Follow-up. In all of the monitored phases the ninety minute periodicity of night-waking frequency peaks was observed, a finding which suggests that despite their severe to profound degrees of learning disability, the children's NREM-REM sleep biorhythms appeared to be very similar to those reported for non-retarded subjects (Kerkoff & Middlekoop, 1991) which supports the contention that this component of their sleep processes are essentially unaffected by their intellectual disability or any underlying brain impairment.

Because the children were monitored over several weeks, the night-of-the-week influences upon the night-waking frequencies over the course of the study were tested in relation to the marked middle-of-the-week excess of night-wakings compared to that seen over the weekend reported by the parents. This comparison reveals similar trends in both the monitor and diary-derived data, however, more night-to-night variation is seen in the monitor data.

4.4.3 The Case for Using Monitors

From the point of view of obtaining objective evidence of treatment effects the monitors have been shown by this study to provide convincing evidence of improved settling time, improved "sleep efficiency" (that is more time spent asleep during the children's sleep period) and overall reductions in discrete night-waking episodes similarly to how Sadeh, et al (1992) using a different type of activity monitor demonstrated with their cohort of non-retarded infants. Furthermore, for the first time using this method, a trend towards
shorter waking episodes and reduced levels of activity within these episodes after the intervention were revealed. These changes may be interpreted in terms of the children becoming better able to soothe themselves when they awake during the night without the same intense need for parental attention (see above). Also for the first time, monitored severely retarded children were shown to wake more frequently around the times associated with the normal transitions from "deep" to "light" sleep. As has been discussed previously, the revealed periodicity of these recurrent points of high waking probability of approximately ninety minutes did not differ from that reported for non-retarded individuals. Therefore from an empirical research point of view the monitors provided good evidence of altered movement patterns over the course of the study which were easily interpretable as improvements in two of the key specific sleep problem parameters (night-settling times and frequencies of night-waking). However, from a clinical perspective, whether the children were monitored or not did not appear to make any difference in terms of treatment outcome from either the carers' or the investigator's points of view nor did the monitor data influence the investigator's assessment of the degrees of improvement. Additionally, the general directions of change in these parameters were very similar to those seen in the children's diary reports. Therefore a case for using monitors in ordinary clinical practice with children who have NS/NW problems has not been supported by the results of this study: diary monitoring appears to yield sufficiently robust practical data for this purpose. The high mechanical failure rate of the monitors resulting from their susceptibility to damage at the hands of children renders them impractical for these purposes.

4.4.4 Conclusion
The results of this study demonstrate that ambulatory electronic activity monitors do appear to provide a novel and potentially important means of assessing sleep problems and their treatment from a more objective standpoint within the context of the sufferers' normal environment. However, their utility in ordinary clinical practice has not yet been properly established. Whether or not they will eventually prove to be the most popular means of investigating several types of sleep disorders by clinicians as Sadeh et al (1992) predict, remains to be seen.

4.5. IMPLICATIONS FOR FURTHER WORK

4.5.1. Resource Implications

A major advantage of the "rapid" treatment approach tested in this study is that it is far less expensive in terms of therapist contact time: "graded changes" techniques usually require at least half a dozen contacts between affected families and their therapists. These usually involve home consultations in addition to clinic sessions, each session lasting up to an hour and a half (Quine, personal communication) with additional travelling time. Also, current practices often require a baseline period of several weeks' worth of sleep diary data which is collected and analysed before the actual treatment is started in addition to at least one session devoted to negotiating the type and speed of the desired changes. This can add considerably to the resource implications. Even in the form employed in this study the "rapid" approach employed less contact time than that commonly required for "graded changes". In clinical practice the contact time required is even less than this. Prior to the study the author's usual schedule for an uncomplicated case was as follows:

(i) A combined assessment and treatment session usually taking place in a clinic setting...
but sometimes occurring at home, this lasts between an hour and a half; (ii) This is followed by three or four telephone contacts on successive days after the initial session which take between five to ten minutes per call until the treatment starts working and, thereafter, further telephone contact with the therapist as required. (iii) A brief one-month follow-up appointment at the home or in the clinic; alternatively, this may be another telephone consultation. By using this protocol the average therapeutic contact per case is about three hours compared to an estimated six to nine hours, at least (not including travel time), with the longer approach. Put another way, in theory for every case dealt with by means of the "graded changes" approach, at least two or three cases could be treated by the "rapid" approach. Given that these types of sleep problem are commonly encountered in primary care and child health practices there appears to be a strong economic argument for therapists to consider using the "rapid" approach as a first-line treatment for night-settling and night-waking problems. In the personal experience of the author of over a hundred cases of severely sleep disordered children only two cases required a "graded changes" approach after the "rapid" approach proved ineffective. What's more rapid failure of the initial approach provided important information relating to the affected families' functional dynamics which were not originally apparent (extreme separational anxieties on the part of one mother and undeclared marital dysharmony in the other case). Thus this technique can occasionally prove useful from an assessment or diagnostic point of view in addition to its usual therapeutic efficacy.

4.5.2. Implications of the Study Results for Clinical Practice

In the light of the results of this study and from an appraisal of the relevant literature, the
author's clinical practice relating to this particular clinical problem has been modified in several ways:

(a) Two weeks before the initial appointment with affected families whose primary stated problem upon referral is a severe NS and / or NW disorder, they are now sent routinely copies of the standard sleep diaries (see "Appendix") with information concerning to how to use them. As has already been discussed, this activity by itself can have therapeutic value in that it can serve to distance the parents from their children's problems which is also a probable prerequisite for treatment success. The quality of the record keeping and the comments added to the sheets can also reveal important information concerning families' dynamic functioning and their resources which might prove helpful in the initial assessment interview. Additionally, diaries are now employed routinely by the author in order to monitor treatment outcome which has the benefits of giving the parents and carers rapid feedback of the effectiveness (or otherwise) of their efforts and gives the clinician important data with which to alter or reinforce the advice accordingly.

(b) Additional post-treatment brief telephone contacts are employed in order to ensure that progress is being made or that any impending relapse is attended to quickly (as might be the case with older and more able children who, clinical experience suggests, commonly "test" their parents' resolve to be firm with them at bedtimes after the first week of treatment).

(c) In order to further improve the cost-effectiveness of the treatment the author is currently devising a means of delivering this treatment to a group of half a dozen affected families at a time - a group size which has been shown to be an efficient means
of delivering behavioural modification instruction to parents (O'Dell, 1985). Unfortunately in delivering therapy in such a way one important factor which might determine treatment effectiveness ultimately is the loss of the opportunity to act decisively close to the time of the initial referral when the family are still in crisis. This effect could be reduced by contacting regular referral sources and requesting suitable subjects for a group which is set to start imminently. However, given the high prevalence of these sleep problems it is likely that sufficient numbers of subjects could be recruited quite quickly and easily.

4.5.3. Training Other Health Workers

An even more efficient means of addressing this problem would be to encourage child health and welfare professionals who are employed to screen vulnerable children to ask about sleep problems (and other common behavioural problems of early childhood) when they review such families routinely. Early detection could lead to early rapid treatment before the problems become ingrained and begin to distort the children's and their families' development. Health visitors, general practitioners and paediatric staff should also be prepared to instruct parents, and particularly the parents of severely retarded infants, about their responsibility to train their children to establish good sleep habits by the end of their first year of life thereby preventing NS/NW problems from emerging in the first place. If significant sleep problems are detected then these same professionals are best placed to initiate therapy along the straightforward lines employed by this study. Health visitors particularly have been shown to be effective behaviour therapists for sleep and other common behavioural problems of childhood after short training courses. Furthermore, health visitors who develop these skills report enhanced levels of job satisfaction (Quine & Wade, 1991). If this system could be established then it is highly likely that the rates
of common behavioural problems should fall and the abilities of parents to cope with the everyday demands of dependent and handicapped children will be enhanced. Furthermore, the currently high prevalence of behavioural and emotional problems seen in severely learning disabled adults, problems which in many cases probably arise in early childhood, could also fall in the future and, consequently, reduce the high costs of caring for such individuals. In such a system child or learning disability specialist psychiatrists would have more freedom to train primary care staff in the necessary skills of behaviour management and act in a consultative capacity to these trained professionals. Only very severe, refractory cases or cases complicated by factors such as severe medical factors or issues of abuse or neglect where straightforward solutions are contraindicated would require direct specialist involvement with such children.

4.5.4. Other Suggestions

Other approaches might include the establishment of special children's sleep clinics which appear to yield good results (see "Introduction") and the use of advice sheets which can be freely available at primary care practices. A health visitor colleague who developed such advice sheets which included the proviso that parents who get "stuck" with the advice or require further help with their children's sleep problem specifically, could contact him at the practice and a home appointment arranged. Through this approach he managed to virtually eradicate early childhood NS/NW problems in a large urban group practice in less than a year (although upon enquiry six months after he moved away to another job, the rates slowly returned to those seen before his initiative started).

4.5.5. Implications for Working with Older Learning Disabled Children and Adults
It is highly likely that there is continuity between childhood and adult sleep problems throughout the population. In the field of learning disability the strongest continuity is probably for NS/NW problems; however, as has been discussed previously, further long-term prospective follow-up studies are required to establish this proposition satisfactorily. Nevertheless severe and chronic NS/NW problems are seen in teenage and adults with severe learning disabilities.

The clinical experiences of the author and several colleagues suggest that such cases respond to a regimen based upon the core principles of the approach examined in this study. However, certain adaptations are required in order to cater for the older, and often stronger, patient:

1. When rapid put-back is impractical then the house has to be made safe and secure requiring that potentially dangerous areas such as the kitchen, stairs and house exits are blocked or locked. Ideally, the patient should only be given access to the landing or hallway and special gates can further restrict access. The availability of toys, TVs or other sources of pleasant recreation in the bedroom should be removed. The parents' bedroom door, particularly, usually requires locking if the patient is ambulant.

2. Customised adaptation of the bedroom environment which make fixtures and fittings safe and fitting particularly robust grilles for windows and restraining brackets for heavy furniture is helpful. Heating pipes and radiators can also be covered with protective grilles to prevent accidental scalding.

3. Protected one-way mirrors in the bedroom doors can allow parents to check their children without disturbing them and a dim red bedroom light controlled from outside the patient's bedroom can be used in conjunction with this.

4. Heavy curtains can be attached to the patient's bedroom curtain rail by "Velcro" fastening tapes in order to prevent the patient repeatedly pulling down the curtain.
rail during the night. Particularly in the summertime, the bedroom windows may be painted with non-permanent non-toxic window paint or, alternatively, be blocked with cardboard and tape in order to cut down the level of external light in the bedroom and thus promote sleep.

(5) Some older individuals can benefit from a short (one or two-week) low dose of neuroleptic medication (10-20 mg of thioridazine, for example) given two hours before bedtime. If they are already receiving such medication then an increased early evening dose may help to promote the relaxed frame of mind necessary for sleep onset. This would be in addition to the usual "winding down" processes described in the "Method" section of this thesis. Alternative forms of medication are the newer night sedatives, such as temazepam, zolpidem and zopiclone, for example; however, these agents may interfere with the learning and adaptational processes involved and would require close monitoring (they may also induce "paradoxical" reactions - see above). In certain circumstances sedative hypnotic medication may also be prescribed for the parents. For example: when the patient's parents demonstrate severe psychophysiological insomnia resulting from anxieties about being able to tolerate separation from their children. Again, the close monitoring proviso applies in these circumstances.

(6) With patients who have a mental age of at least three years, in addition to the usual morning praise from the parents should the patient sleep well, "star charts" and other material rewards can be built into the programme as long as they are demonstrated to be meaningful for the individual concerned.

Summary

Many of these initiatives may prove to be helpful over and beyond the effects of any "sleep programme" when trying to establish good bedtime habits for patients and their families. The District Occupational Therapist can offer invaluable advice in these
circumstances and also carries statutory responsibilities to disabled individuals and their families for the delivery of therapeutic aids and domestic adaptations. Sometimes in sleep and generally behaviourally disturbed patients this can be as substantial as planning and funding special bedroom extensions.

4.5.6. Directions for Further Research

(a) Sleep Studies

As has been stated previously, this project has provided the opportunity for an exploration of the subject of sleep disorder medicine in the field of learning disability with a particular emphasis on the commonest sleep problems encountered in childhood namely night-settling and night-waking difficulties. The results of the intervention study suggest that a behaviour modification approach aimed at the rapid extinction of these distressing problems when they present in a severe and chronic manner can be very successful and thus confirming the clinical experience which initially prompted this study. However, the small scale, selective, uncontrolled and "open" nature of this clinical enquiry necessarily requires that a high degree of caution is applied when drawing conclusions about the scientific validity of the results obtained. As has been discussed at present there appears to be a marked reluctance to use this type of treatment in child psychological and psychiatric practice at present and, until the results of this study can be replicated by other workers using the same protocol, many will remain in doubt about its efficacy and acceptability. A major ethical problem with this situation is that the results confirm the treatment's clinical efficacy yet to test this finding to a generally satisfactory manner
would require the use of untreated control groups matched for the cardinal features and severity of their problems, development and disability. As prospective studies have shown, such families would stand a very high chance of having to endure several more months of distress; a proposition which clinicians and, indeed, affected children's families would find ethically unacceptable. One answer to this impasse would be to test the intervention in children demonstrating only moderate degrees of NS/NW problems whose clinical need for treatment would be less pressing and whose families would probably accept some deferment of remedial therapy. Another, and more useful option, would be to conduct a large series of multiple baseline long-term single-case studies applying the intervention at different stages in each subject. The subjects themselves could have a range of degrees of severity of sleep problems. This would allow specialised statistical techniques such as time series analyses to be applied which could ascertain more precisely whether it is the behavioural advice, specifically, or alternatively the influence of other factors which are responsible for any changes in the target behaviours. The "rapid" technique also needs to be directly tested against what appears to be the more commonly applied forms of treatment namely "graded changes" and sedative hypnotic therapy. This would require that children matched with respect to the principal variables previously described would be randomly allocated to the three treatment options. Independent ratings of the outcome measures by investigators who are "blind" to the type of treatment and also to knowledge of when it was applied would also be desirable. Activity monitors could be used to furnish objective data concerning the children's sleep and daytime activity patterns in all of these experimental paradigms.

(b) Activity Monitoring of Childhood Sleep Problem
With children who were monitored in other situations (by DB) monitor data have proved to be helpful in showing parents who report that their children "...never sleep at all" long periods of inactivity during the night. This is readily demonstrable in their children's night-by-night activity printouts (see example given in the "Appendix"). These long periods of no or very little activity can only reflect that their children were indeed sleeping unless it can be shown that the monitor is either not worn or otherwise malfunctioning. It is also possible that the monitors might prove to be helpful when encouraging very disorganised families to stick to the treatment when prior attempts have failed through parental inconsistency, by virtue of the monitor itself having to be closely monitored by the parents; thus putting an immediate level of structure into the treatment which other approaches do not require. Another potentially useful clinical application for them is with more able NS/NW children who have proved refractory to previous treatment. If their enthusiasm to participate in treatment is an identifiable obstacle, the monitor and its data printout facility could prove to be of sufficient novelty value to engage these child effectively. Somnambulistic and nocturnal rhythmic activities (headbanging and body rocking particularly) have also been both assessed and treated with the help of activity monitors by the author although in a non-systematic manner. Again, the graphical visual feedback of their bedtime activity patterns to the children and their parents has, on occasion, proved helpful in both objectifying the problem and also motivating affected children to change their behaviour for the better. Given the increasing popularity of computer video games with children, the use of the activity monitors might prove to be an effective additional means of helping them with their sleep problems. However, the utility of this particular application of AM when applied to sleep disordered individuals who are also severely learning disabled would be doubtful because they would be unable to understand the relevance of the data. Another area where activity monitoring
is being used to both investigate and also monitor treatment effects in a specific sleep
disorder common to a specific handicapping condition is with Down's syndrome children
with obstructive sleep apnoea; however the findings of this enquiry have not yet reached
the stage of publication (Stores, personal communication).

(c) Sleep Disorders of Adult and Elderly Learning Disabled People

As Espie and Tweedie have pointed out (Espie & Tweedie, 1992), there has been very
little epidemiological enquiry in this area. Ideally, health surveys of these groups should
enquire routinely about the cardinal symptoms and signs of sleep disorders (and in their
principal carers if living at home). Given the high rates known to exist in community
cohorts of severely impaired children and their known tendency to persist it is necessary
now to conduct large-scale studies specifically aimed at establishing the patterns of normal
and abnormal sleep existing in heterogeneous groups of learning disable people.

(d) Audit of Current Clinical Practices

Another avenue of investigation would be to conduct a large scale audit of clinicians'
practices in respect of NS/NW problems when they present in learning disabled children
and adults. One way to obtain this would be to provide clinicians with case vignettes and
fixed option alternative management strategies from which they would chose. This would
allow relatively easy interpretation of data collected in this way. By surveying child
psychiatrists, learning disability psychiatrists and paediatricians in this way, firmer
conclusions might be drawn about the prevailing attitudes and practices in this clinical
area. Such information could then inform interested clinicians' and researchers' attempts to characterise and, where necessary, elaborate strategies to improve practices in this area.

4.5.7. Final Recommendations

This study has demonstrated not only the serious effects of poor sleep upon severely learning disabled children and their families but also the possibility of effective rapid treatment. This points to the need for a much more systematic approach to these problems, indeed, the profile of sleep disorder medicine in Britain needs to be raised generally (Stores, 1990) as has been the case in North America and several West European countries (Dement and Mittler, 1993). This specific issue is currently being addressed on a national basis by such professional bodies as the Royal Colleges of Psychiatry and Physicians and also the British Sleep Society. At a local level, interested clinicians and researchers give talks and run workshops for child health, care and welfare professionals on sleep disorder topics and it is the experience of the author and his colleagues that these meetings are generally very well attended and the advice received is usually taken back to workplaces and used in practice. This suggests that there is a growing grass-roots awareness of the clinical importance of sleep disorders, unfortunately this belief has not yet been translated into action by the curriculum managers of our medical and nursing schools. New editions of the standard textbooks of learning disability and child care should include specific chapters devoted to the assessment and treatment of sleep disorders. Further research is required in order to properly evaluate the various treatment approaches to sleep problems used in clinical practice with different groups of people. It is hoped that this thesis has highlighted the particular needs of such a specific
group namely learning disabled children and their families and has also shown that a straightforward treatment approach to this group's commonest sleep problem can be very effective.
5: APPENDIX
Dear Colleague,

As you are probably aware, I am conducting a research project which evaluates a behavioural approach to sleep disordered children who also have severe learning difficulties. It appears from the literature that about 40% of these children have problems related to night time settling and waking which can add considerably to the subjective burden of care of parents. The children themselves suffer from the effects of chronically disturbed sleep; this can take the form of overactivity, irritability and daytime drowsiness. These additional problems can further impair attention and concentration thereby compounding their learning difficulties. Therefore it is important to treat these problems promptly and efficiently. In the past there has been general reticence to attempt to do this and there are various reasons why this has been so. Notwithstanding this, modern clinical practice has shown that simple behavioural techniques can be highly effective with sleep disordered children irrespective of degree of learning difficulty. This now has to be tested properly in order to inform good clinical practice and to also help in planning good psychiatric services for this disadvantaged group.

My investigation will take the form of a series of single case studies involving sleep disordered children (defined as children with pronounced difficulties in settling at night, demonstrating repeated night waking and having limited hours of sleep). The children will be assessed using a range of parameters including the use of wrist-worn activity monitors (which are the size of a large watch). The behavioural treatment will be one that has been shown to be useful in clinical practice and is well accepted by parents and carers. The project has received approval from the Leicestershire Research Ethical Committee.

If you know of any children who have, or might, have this type of problem and who are otherwise reasonably healthy and between the ages of two and ten (or thereabouts), please discuss this project with their parents/careers and, if they are willing to participate, refer them to me at the address above. I enclose an introductory letter which you might find helpful. Depending on demand I can probably see the children within two weeks of referral.

Yours sincerely,

Dr. David Bramble
Lecturer in Child Psychiatry
3 April 1991

Dear

I have received a request for help with your child's sleep problem. If your child has difficulties in settling at night or demands attention once he/she has gone to bed, then the Department of Psychiatry at Leicester University Medical School would be interested in hearing from you because it is currently undertaking research into this type of problem. We believe we have an effective (non-drug) treatment but this impression needs to be tested properly.

The study involves an initial assessment conducted either in the Department or at your home followed by a short treatment phase which will occur at home.

If you are interested in participating in this project I would be obliged if you could complete the enclosed Sleep Problem Questionnaire and a week's Sleep Diary (also enclosed) and return them to me in the FREEPOST envelope provided.

If you are keen to obtain help for your child but not to participate in the research let me know and I will make the necessary arrangements.

Yours sincerely,

Dr. David Bramble
Lecturer in Child Psychiatry and Mental Handicap
I consent to my child/charge participating in Dr. Bramble's research project. Dr. Bramble has explained both the nature and the purpose of the project and I realise that I can withdraw my child/charge at any time as I see fit and expect to receive appropriate alternative treatment for his/her problems.

Signed:_________________________ Parent/Guardian

Witnessed by:_________________________

Status:_________________________
SLEEP PROBLEM QUESTIONNAIRE

Q1 Does your child/charge have problems related to his/her sleeping? (YES/NO). If YES please fill in the questionnaire below:

Q2 Name of Respondent:

Q3 Relationship to Child:

Q4 Name of Child:

Q5 Sex of Child:

Q6 Age of Child:

Q7 Date of birth:

Q8 Address:

Q9 Name of G.P.:

Q10 Name of Health Visitor:

Q11 Hospital Consultant (if any):

Q12 Cause(s) of learning disability (if known):

Q13 Any associated medical conditions? (e.g. epilepsy, deafness, physical handicap):

Q14 Current medication (if applicable):

Q15 What is the height of your Child (in metres)?

Q16 What is the weight of your Child (in kilos)?

Q17 Any excessive weight gain in past 6 months?

Q18 Any excessive weight loss in past 6 months?

Q19 Has your Child had any significant allergies or infection involving ears, nose or throat in past 6 months? (Please specify)

Q20 Do any other children sleep in the same room as this Child? If YES, How many?
SLEEP PROBLEM QUESTIONNAIRE II

PLEASE ANSWER THE FOLLOWING QUESTIONS ON THE BASIS OF THE CHILD'S SLEEP BEHAVIOUR DURING THE PREVIOUS 6 MONTHS.

Q1 How long does it usually take for your Child to fall asleep at night?

Q2 How many hours does your Child sleep per night?

Q3 Does your Child usually awaken during the night?

Q4 If he/she awakens during the night how many times does he/she usually awaken?

Q5 Does your Child generally sleep soundly?

Q6 Does your Child usually sleep lightly? (i.e. any noise will awaken him/her)

Q7 Please indicate (by a tick) from the list of statements below - the statement(s) which best describe your Child's sleep related problem:-

(a) Disturbance of Sleep

i) Sleep disturbed most nights - screams and needs attention.

ii) Sleep disturbed once or twice/week - screams and needs attention.

iii) Sleep disturbed occasionally - screams and needs attention.

iv) Sleep disturbed, wants attention, does not carry on or screams.

v) Sleeps poorly but lies quietly when awake.

vi) Usually sleeps well.

(b) Limited Hours of Sleep

i) Usually goes to bed very late and wakes very early.

ii) Usually wakes very early but goes to bed at a reasonable hour.

iii) Usually goes to sleep very late but wakes at a reasonable time.

iv) Occasionally late going to sleep or wakes early but usually has no problem.

v) Always, or nearly always, goes to sleep and wakes at a reasonable hour.
Q8 Please describe your Child's pattern in detail below:

Q9 Any other problems related to night-time? (e.g. bedwetting nightmares, epilepsy, snoring, etc)
If YES, please describe below:

Q10 i) Any behavioural problems at other times? (e.g. self-injury hyperactivity, aggression)
If YES, please describe below:

ii) If there are any other problems please list up to two of these here:

Problem 1
Problem 2

Q11 In the general context of your Child's problems please rate the seriousness of his/her sleeping problem on the line below:–

0 No problem 10 Very severe

Q12 i) Have you sought any help for your Child's problems in the past? YES/NO

ii) If YES, who have you approached:– (please tick)

a) Other family members
b) Spouse
c) Member of Care Staff
d) Health Visitor
e) GP
f) Health advice journalism
g) Psychologist
h) Child psychiatrist
i) Mental handicap psychiatrist
j) Teacher
k) Social Worker
l) Other (please state)

iii) What advice did you receive?

Q13 Did you find the advice helpful? (Please mark on the line below a point to indicate how useful it was)

0 Not helpful at all 10 Extremely helpful

Q14 How long, approximately, did your Child have a sleep problem before you received help for it this time? (Years/Months/Weeks)
## Sleep Problem Questionnaire
### Part III

During the past 6 months has your child shown any of the following behaviors? If you respond Yes check the appropriate column for frequency.

<table>
<thead>
<tr>
<th>Description of Behavior</th>
<th>No</th>
<th>Yes</th>
<th>Daily</th>
<th>More than 1X week but not daily</th>
<th>2 to 4X per month</th>
<th>About 1X month</th>
<th>Less than 1X month</th>
</tr>
</thead>
<tbody>
<tr>
<td>Talks in sleep</td>
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<tr>
<td>Walks around while asleep</td>
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<tr>
<td>Grinds teeth during sleep</td>
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<tr>
<td>Bangs head during sleep</td>
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<tr>
<td>Has quick movements of arms or legs during sleep e.g. kicking, jumping, arm flailing</td>
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<tr>
<td>Moves around in bed quite a bit during sleep</td>
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<tr>
<td>Bites tongue during sleep</td>
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<tr>
<td>Nores during sleep</td>
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<tr>
<td>Gags or chokes during sleep</td>
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<tr>
<td>Seems to stop breathing for periods of time lasting up to 30 seconds during sleep</td>
<td></td>
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<tr>
<td>Wets bed during sleep</td>
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<tr>
<td>Awakens during night complaining of nightmare or frightening dream and during this awakening seems moderately anxious</td>
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</table>

Over
Description of Behavior

<table>
<thead>
<tr>
<th>Description of Behavior</th>
<th>No</th>
<th>Yes</th>
<th>Daily</th>
<th>More than 1X week but not daily</th>
<th>2 to 4X per month</th>
<th>About 1X month</th>
<th>Less than 1X month</th>
</tr>
</thead>
<tbody>
<tr>
<td>During the day has irresistible urges to go to sleep</td>
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<tr>
<td>During the day appears drowsy most of time but this is not an irresistible urge to sleep</td>
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<tr>
<td>During the day does he/she appear more active than usual</td>
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</table>

If you have any further comments to make concerning your child or this questionnaire please write them in the space provided below :

Thank you for filling in this questionnaire.

Please return to Dr. David Bramble, Lecturer in Child Psychiatry, Department of Psychiatry, C.S.B., L.R.I., Leicester, LE2 7LX.
SLEEP PROBLEM FOLLOW-UP QUESTIONNAIRE

Introduction

You remember filling in a questionnaire concerning your child's sleep problem when we first met. This one
endeavours to assess any changes in the sleep problem (and other related problems).

Q.1 Please indicate (by a tick) from the list of statements below - the statement(s) which best describe your
child's sleep related problem:-

(a) Disturbance of Sleep

0 Sleep disturbed most nights - screams and needs attention.  
1 Sleep disturbed once or twice/week - screams and needs attention.  
2 Sleep disturbed occasionally - screams and needs attention.  
3 Sleep disturbed, wants attention, but does not carry on or screams.  
4 Sleeps poorly but lies quietly when awake.  
5 Usually sleeps well.

(b) Limited Hours of Sleep

0 Usually goes to bed very late and wakes very early.  
1 Usually wakes very early but goes to bed at a reasonable hour.  
2 Usually goes to sleep very late but wakes at a reasonable time.  
3 Occasionally late going to sleep or wakes early but usually has no problem.  
4 Always, or nearly always, goes to sleep and wakes at a reasonable hour.

Q.2 Please describe your child's current pattern in detail below:-

Q.3 Any other problems related to night-time? (e.g. bedwetting, nightmares, epilepsy, snoring,etc):

YES/NO - Please describe:-

Q.4 Any behavioural problems at other times? (e.g. self-injury, hyperactivity, aggression):

YES/NO - Please describe:-

182
Q.6 Please describe below which (if any) aspect of the advice you received concerning your child's problem(s) you found helpful:

Q.7 Overall, which single aspect of advice (if any) did you find most useful? Please describe below:

Q.8 How helpful was the advice generally?
(Please indicate on the line below a point, which best reflects its usefulness):

0 Not helpful at all

10 Extremely helpful

Q.9 (a) Have there been any other changes in your child's behaviour, mood or abilities since you received help for his/her sleep problems?

Yes No Don't Know

(b) If there have please describe these below:

Q.10 (a) Have you noticed any changes in yourself or the rest of the family during this treatment period? (please tick):

Yes No Don't Know

(b) If you have please describe these below:

Q.11 If there has been a change in your child's sleep problem how soon after the first interview did you notice this change?
(Months/Weeks/Days)

Thank you for your help with this questionnaire.

Please return to:- Dr David Bramble, Lecturer in Child Psychiatry,
Department of Psychiatry, C.S.B., L.R.I., Leicester, LE2 7LX
Dear,

It is now nearly eighteen months since was treated for (his/her) sleep problem. If you can spare the time I would be obliged if you could answer the brief questions listed below and return this to me in the FREEPOST envelope enclosed.

1. (i) Does still have a problem at bedtime?
(ii) If so please describe it below and rate its severity on the line provided:

| 0 (No problem) | 10 (Very Severe Problem) |

(iii) Do you require further help with this?

2. (i) Since the treatment have you noticed any change in which you think might have resulted from the sleep treatment?
(ii) If so please describe below:

3. (i) Have there been other changes in yourself or the rest of the family which you think might be related to sleep improving?
(ii) If so please describe below:

4. Any other comments?

Many thanks

Yours sincerely

[Signature]

Dr David Bramble
Lecturer in Child Psychiatry
Now I would like to ask you some questions about N's day time behaviour.

Interviewer:

When asking about behaviour, if a problem is reported, further questions should cover the following:

What happens, when it happened, severity, frequency, fluctuations, what brings it on, how parents deal with it, what makes it better or worse, when it started, has it ever happened before, is behaviour different with others?

Record ALL information on the schedule and summarise in a report at the end.

1) How about eating? Is N going through the faddy stage? How has his/her eating been in the past 4 weeks? Does N eat everything or is he/she fussy? Do you have to make special meals? Is this a problem?

0. No eating problems (include here the occasional missed meal or fad)

1. Some eating difficulties (has a few fads, or has a poor appetite for one meal a day or less)
   a) Poor appetite

2. Marked difficulties (appetite poor for at least half the meals: diet mainly milk and/or baby foods or sweets and biscuits, or eats only a few articles of food or refuses practically all meat and/or vegetables)
   b) Faddy eater

2) How often has N had an accident - soiled - in the past 4 weeks?

0. None
1. Twice a week or less
2. Three times a week or more
Now I'd like to ask you some questions about N's sleeping habits.

1) Does N have a room of his/her own to sleep in? Does he/she share a room/bed?
   1. Has own room/own bed.
   2. Has shared room/own bed.
   3. Has shared room/shared bed

2) A lot of children don't like going to bed. What about N? How long does it take to settle him/her? What about going to sleep? Does N sleep right through the night? (What happens? How long does it last? How often is this?). Does N seem to need very few hours sleep?
   0. No problem or problems less than once a week
   1. Problem occurs 1-2 times a week only or settles in less than 1 hour
   2. Problem occurs 3 times a week or more and either takes more than 1 hour to get to sleep, or wakes at night for more than a few minutes, or goes into parents room or bed

3) How often do you take N into your bed or sleep in N's bed because N is upset? Do any of N's brothers or sisters take N into bed with them?
   0. Never sleep with child
   1. Occasionally: all night once a week or less, or for a couple of hours only, more often
   2. Frequently: all or most of the night twice a week or more

   a) Going to bed/going to sleep
   b) Waking at night
   c) Needs limited hours sleep

   a) Parents
   b) Brothers/Sisters
3) Is N independent or does he/she cling a lot? Can N be left with people he/she knows? At home does he/she follow you around all day ... even into the bathroom?

0. Reasonably independent for age
1. Some dependency; upset if left, takes some time to get over it
2. Marked dependency; cannot be left at all, continually demands to be with mother

Dependency ____ 9

4) Most children are difficult to manage at times - how do you find N? Can you take N shopping ... visiting without trouble? Do you feel N is out of control or difficult to discipline?

0. Easy to manage and discipline
1. Sometimes difficult or out of control or hard to discipline for short periods
2. Long or very frequent periods, nearly every day, when difficult to manage or discipline

Difficult to manage ____ 10

5) Does N have temper tantrums? What happens? Is it a real tantrum with shouting, screaming ... banging ... kicking? How often in the past 4 weeks? How long do they last? When did they start?

0. No tantrums/difficult behaviour
1. Brief tantrums lasting a few minutes: one or two a day
2. Frequent tantrums three a day or more or tantrums lasting more than 15 minutes or frequent difficult behaviour

Tantrums ____ 11

6) Does he/she have moods of being miserable or irritable? Is he/she usually happy? How has N been in the past 4 weeks? How often does this happen? When did it start? How long do the moods last?

0. Usually happy except for brief periods
1. Sometimes miserable/irritable/discontented for periods less than 1 hour on most days and/or for longer periods once or twice a week
2. Frequently miserable/irritable on most days and/or for long periods 3 times a week or more

Mood ____ 13
7) How active is \_N\_? Is he/she the sort of person who doesn't like sitting still even for meals? How many minutes will he/she sit at meal times usually? What about for a story? TV? How long has he/she been like this? Is it a problem? Do you ever feel \_N\_ is too active?

Interviewer: Code '0' for child who is immobile

0. Not markedly active
1. Very active
2. Hyperactive: sits still for meals or on other occasions usually less than 5 minutes
3. Underactive: spends most of time stationary and unoccupied (apart from habits)

Activity

8) Can \_N\_ amuse him/her self or does he/she keep asking you to do things with him/her? What's the longest time \_N\_ will usually stick at one thing indoors, if interested?

0. Concentrates well
1. Concentration variable or very variable
2. Finds it hard to concentrate

Concentration

9) How has \_N\_ got on with his/her brothers/sisters in the past 4 weeks? Do they squabble much? How much is playing together affected? Is he/she jealous at all? What about other friends? Are there any around for \_N\_ to play with? How do they get on?

0. Trivial or no difficulties
1. Some difficulties, play disrupted or prevented most times but only for short periods
2. Marked difficulties: play disrupted or prevented most of the time
3. No brothers/sisters/no opportunity to play with others

a) Siblings

b) Peers

10) Does \_N\_ keep asking for attention, ask to have things done which he/she could do for self, e.g. feeding ... dressing? Will \_N\_ play/occupy him/herself on his/her own or does he/she want you to do things with him/her all the time? Is this a problem? How is he/she with others?

0. Rarely demands undue attention
1. Sometimes demands undue attention
2. Continually asking for attention

Attention seeking
11) Is _N_ a worrier .... gets anxious about something that might happen .... about plans or changes? What about if _N_ loses something? Does he/she brood over things .... like accidents .... falls .... illnesses .... monsters? Does he/she ever keep repeating questions about something" .... like being left .... being loved .... death?

(A WORRY IS APPREHENSION ABOUT SOMETHING THAT MAY HAPPEN; A FEAR IS APPREHENSION ABOUT SOMETHING THOUGHT TO BE PRESENT OR ACTUALLY PRESENT)

0. Never or rarely worries
1. Some worries for brief periods
2. Many different worries, or worries over certain things for long periods

12) Does he/she have any rituals, things being done in a certain order e.g. at bedtime? Gets upset if things are touched .... moved out of place? Are toys/clothes always kept in a special order?

0. None
1. Some, brief
2. Many, prolonged

13) Most young people have some fears. What is _N_ afraid of? What about?

Interviewer: Code each fear separately.

(A FEAR IS APPREHENSION ABOUT SOMETHING THOUGHT TO BE PRESENT OR ACTUALLY PRESENT)

Scale A for individual fears

A. Individual fears

0. Not afraid
1. Somewhat afraid; uncertain about approaching, requires reassurance
2. Marked fear; runs away or avoids, clings to adult, cries, has to be comforted

- Dogs
- Cats
- Other animals/insects
- Thunder/loud noises
- The dark
- Strangers
- Going out
- Car, bus, train, tube
- Lift, escalators
- Water, the bath
- Haircut
- Doctors
- Stories
- TV programmes
- Hoover
- Other
Scale B for overall rating of fears

0. Is somewhat afraid of 1 or 2 things or has no fears

1. Has 1 or 2 marked fears or 3-5 fears altogether

2. Has 3 or more marked fears or 6 or more fears altogether

B. Overall Rating of Fears

14) Does N have any habits before going to sleep or during the day? Something he/she keeps on doing, possibly without being aware of it? When did it start? How often does he/she do it?

Interviewer: Code each habit separately

(A HABIT IS A STEREOTYPED, REPETITIVE ACTION.)

If a habit is mentioned, ask: Is this a severe problem for you? Only a mild one?

0. Never

1. Usually less than 20 minutes a day

2. Usually for 20 minutes a day or more

Headbanging

Flicking, pulling, scratching hair, skin or nails

Other self injurious behaviour

Rocking

Sucking thumb or fingers

Sucking other objects

Making irritating noises, e.g. growling, humming, incessant giggling

Nervous movements like blinking, pulling faces, grinding teeth, licking or biting tips, sucking tongue

Using object for twiddles

Play with self down below

Overall rating of habits

0. No habits

1. 1-3 habits sometimes or 1 frequent habit

2. 2 or more frequent habits or 4 or more habits altogether

Overall Rating of Fears

37

38

39

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48
15) Does \_N\_ do any of the following more than you would normally expect? e.g. more than most young people of same age.

0. Never
1. Sometimes
2. At least once a week

- Run away or attempt to run away
- Spitting, smearing, etc.
- Use toilet inappropriately, e.g. stuffs paper or other objects, drinks from it, spreads water around
- Shout and scream
- Aggressive behaviour
- Hoard or take other people’s belongings (not stealing)
- Take clothes off at awkward or inappropriate times
- Behave inappropriately to people outside the family, e.g. kissing strangers, sucking people’s clothing, smelling people
- Interfere with other’s belongings or activities
- Play with matches, fires or try to light fires
- Destructive behaviour
- Scatter or throw objects round
- Eat things which aren’t food (coal, soil, etc.)

16) Is there any other behaviour of his/hers that you or other people find embarrassing?

RECORD VERBATIM AND CODE
17) Does N ever get into trouble for fighting ..., stealing ..., truanting ..., at school/SEC? How often is this? Does he/she stand up for him/herself? Is it a severe problem?

<table>
<thead>
<tr>
<th>0. No</th>
<th>1. Occasionally/mild problem</th>
<th>2. Regularly/severe problem</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>Taking things he/she shouldn't</td>
<td>6</td>
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<tr>
<td></td>
<td>Getting into fights</td>
<td>7</td>
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<td></td>
<td>Being disruptive in class</td>
<td>8</td>
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<td></td>
<td>Truancy</td>
<td>9</td>
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<td></td>
<td>Bullying</td>
<td>10</td>
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<tr>
<td></td>
<td>Doesn't stand up for self</td>
<td>11</td>
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<tr>
<td></td>
<td>Being teased</td>
<td>12</td>
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<tr>
<td></td>
<td>Swearing/being rude</td>
<td>13</td>
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</tbody>
</table>
## MATERNAL SLEEP SCALE

**SLEEP QUALITY (GENERAL)**

<table>
<thead>
<tr>
<th></th>
<th><strong>AGREE</strong></th>
<th><strong>DISAGREE</strong></th>
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</thead>
<tbody>
<tr>
<td>1</td>
<td>I feel tired after getting up in the morning</td>
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<tr>
<td>2</td>
<td>I usually sleep during the night</td>
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<td>3</td>
<td>I often lie awake for more than half an hour before falling asleep</td>
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<td>4</td>
<td>I often wake up several times during the night</td>
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<td>5</td>
<td>I usually fall asleep easily</td>
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<td>6</td>
<td>I usually sleep quietly</td>
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<td>7</td>
<td>I think that I usually enjoy my sleep</td>
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<tr>
<td>8</td>
<td>I often don't sleep for more than five hours</td>
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<tr>
<td>9</td>
<td>I often get up during the night</td>
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<tr>
<td>10</td>
<td>I take a sleeping drug regularly</td>
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<tr>
<td>11</td>
<td>I am sleepy during the day</td>
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</tbody>
</table>
Please ring the correct answers

<table>
<thead>
<tr>
<th></th>
<th>Question</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Do you often have backache?</td>
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<tr>
<td>2</td>
<td>Do you feel tired most of the time?</td>
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<td>3</td>
<td>Do you often feel miserable or depressed?</td>
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<td>4</td>
<td>Do you often have bad headaches?</td>
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<td>5</td>
<td>Do you often get worried about things?</td>
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<tr>
<td>6</td>
<td>Do you usually have great difficulty in falling asleep or staying asleep?</td>
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<td>7</td>
<td>Do you usually wake unnecessarily early in the mornings?</td>
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<td>8</td>
<td>Do you wear yourself out worrying about your health?</td>
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<tr>
<td>9</td>
<td>Do you often get into a violent rage?</td>
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<td>10</td>
<td>Do people often annoy and irritate you?</td>
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<tr>
<td>11</td>
<td>Have you at times had a twitching of the face, head or shoulders?</td>
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<td>12</td>
<td>Do you often suddenly become scared for no good reason?</td>
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<tr>
<td>13</td>
<td>Are you scared to be alone when there are no friends near you?</td>
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<tr>
<td>14</td>
<td>Are you easily upset or irritated?</td>
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<td>15</td>
<td>Are you frightened of going out alone or of meeting people?</td>
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<td>16</td>
<td>Are you constantly keyed up or jittery?</td>
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<tr>
<td>17</td>
<td>Do you suffer from indigestion?</td>
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<tr>
<td>18</td>
<td>Do you often suffer from an upset stomach?</td>
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<tr>
<td>19</td>
<td>Is your appetite poor?</td>
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<tr>
<td>20</td>
<td>Does every little thing get on your nerves and wear you out?</td>
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<tr>
<td>21</td>
<td>Does your heart often race like mad?</td>
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<tr>
<td>22</td>
<td>Do you often have pains in your eyes?</td>
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<tr>
<td>23</td>
<td>Are you troubled with rheumatism or fibrositis?</td>
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<tr>
<td>24</td>
<td>Have you ever had a nervous breakdown?</td>
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</tbody>
</table>
Here is a list of the main points of the advice you were given to help your child's sleep problems. Please indicate by a tick in the appropriate box how useful you found each item of advice:

<table>
<thead>
<tr>
<th>No.</th>
<th>Advice</th>
<th>Very Helpful</th>
<th>Helpful</th>
<th>Unhelpful</th>
<th>Very Unhelpful</th>
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</thead>
<tbody>
<tr>
<td>1.</td>
<td>Setting a regular bedtime and sticking to it.</td>
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<tr>
<td>2.</td>
<td>Having a pre-bedtime settling routine in the hour before bedtime.</td>
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<tr>
<td>3.</td>
<td>Winding your child down before bedtime.</td>
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<td>4.</td>
<td>Short settling time in the bedroom and the use of a &quot;magic&quot; word.</td>
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<td>5.</td>
<td>Warning the neighbours about the likelihood of temporary noisiness.</td>
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<td>6.</td>
<td>Making sure that your child cannot be harmed in the bedroom by making it safe and secure.</td>
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<td>7.</td>
<td>Removing stimulating toys, t.v.s etc, from bedroom.</td>
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<td>8.</td>
<td>Agreeing to back one another up about the sleep programme.</td>
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<tr>
<td>9.</td>
<td>Ignoring child's demands for attention after settling(unless they are unwell)</td>
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<tr>
<td>10.</td>
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Finally, could you indicate whether my general approach was too tough, rather tough, just right, rather soft or too soft (circle the relevant phrase)
Example of "MINITAB" Database for Study Parameter Scores

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Example of "MINITAB" Database for Study Parameter Scores

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**key:**

'mumpres' : preintervention *maternal sleep scale score*
'mumpos' : 2/52 postintervention
'mumfus' : 4/12 postintervention

'bpipre' : preintervention *behaviour problem index score*
'bpipost' : 4/12 postintervention
'mumpos' : 2/52 postintervention

'sleepre' : preintervention *parental rating of sleep problem*
'sleeppost' : 2/52 postintervention
'sleepful' : 4/12 postintervention
'sleepfu2' : 18/12 postintervention

'matmalpr' : preintervention *maternal malaise inventory score*
'matmalpt' : 2/52 postintervention
'matmalfu' : 4/12 postintervention

'satispo' : 2/52 postintervention *parental rating of intervention helpfulness*
'satisfu' : 4/12 postintervention

'hbspre' : preintervention *hbs rating of sleep problem*
'hbspost' : 2/52 postintervention
'hbsfu' : 4/12 postintervention
Nocturnal Activity Patterns from Activity Monitor - Derived Data

The following three figures illustrate the activity data patterns derived from monitored patient #1 ("Mark") over the first week of each principal phase of the intervention study. The graphical display of this data was obtained by means of the software programme supplied with the Gadhwiler Electronics activity monitor system. Arrows indicate night-waking events confirmed by analysis by the "Actplan" software programme.

ACTIVITY MONITOR DATA SET: 1

Mark: Pre-treatment (one week)

Pre-treatment data supports the parents' description of the sleep problem. Note repeated wakenings (indicated by arrows) and variable night settling times.

Mean sleep period duration: 5.88 hours/night
ACTIVITY MONITOR DATA SET: 2

Mark: Treatment period + Follow up (one week)

Treatment phase data shows an improved night settling pattern (after the agreed 8.00 pm bed time) and reduced frequency of night waking. Note reduced daytime activity levels during this phase.

Mean sleep duration: 8.76 hours/night

ACTIVITY MONITOR DATA SET: 3

Mark: Four month follow up (one week)

The follow-up data shows a sustained improvement of sleep pattern however the amplitude of daytime activity is greater than the pre-treatment phase.

Frequent low-amplitude short duration (ie non-waking) activity during sleep may reflect a degree of respiratory obstruction (Mark snores loudly)

Mean sleep duration: 9.48 hours/night
Example of "ACTPLAN" Sleep Statistics Software Application

Filename: aaal.mon (subject 'a': data for the first "Pretreatment" phase night)

Key:
- **timewindow**: summed activity data for each 18 second sampling time
- **movement index**: total number of timewindows containing +1 movement divided by total number of timewindows from sleep onset time (20:30 Wednesday) to morning waking time (07:00 Thursday)

### SLEEP STATISTICS

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### SLEEP STATISTICS

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<th>Nr of epochs &lt;= 60 sec without movement</th>
<th>Total number of epochs without movement</th>
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*NB: 'Fragmentation index' data not used in study*
Calculation of Duration and Movement Content of Discrete Night-Waking Events From Activity Monitor Data

Step 1: Establish total amount of movement data counts per night and divide by number of nights monitored
Mean movement per time window x total number of time windows in total sleep time

Step 2: Establish total number of "active" timewindows i.e. those containing 1+ movement during the night = total number of timewindows of night / movement index

Step 3: Establish mean movement count per "active" timewindow = total movement count for night / total no. "active" timewindows in night

Step 4: Establish activity mean movement levels for the time spent awake in discrete NW events per night = from activity patterns establish the total time awake in NW events and convert this to number of timewindow equivalents x mean movement level per "active" timewindow (step 3)

Step 5: Establish the mean movement level per discrete NW event = divide the result of Step 4 by the mean NW frequency per night

Step 6: Establish the mean level of movement per minute of night wakefulness = divide the result of step 5 by the mean duration of night-waking episode for the specific study phase expressed in minutes.

Worked Example:

Patient #1 ("Mark"): Pretreatment Phase (Time Window 18.0 seconds duration)

Step 1: (i) TSP expressed as number of time windows (TWs) = 890.13 TWs
(ii) mean movement per TW = 1.56 movements
(iii) total mean movement per night = (i) x (ii) = 1388.6 movements

Step 2: Number of "active" TWs = total TWs x movement index/100 = 890.13 x 13.4/100 = 116.96 TWs per night

Step 3: Activity per "active" timewindow = 1388.6/116.96 = 11.87 movements

Step 4: (i) Mean Total Waking Time per night = 1.67hrs = 159.86 TWs
(ii) mean movement within this time = Step 3 x Step 4 (11.87 x 158.9) = 1977.54 movements while awake during the night

Step 5: Divide Step 4 result by the mean number of waking episodes per night = 1889.54/2.014 = 942.18 movements per mean night-waking episode

Step 6: (i) Mean duration of night-waking episode in minutes = mean total waking time per night / mean night-waking frequency x 60 = 1.67 hrs x 2.014 wakeings per night x 60 = 47.96 minutes
(ii) Divide the result of step 4 (ii) by the duration of night waking episode (in minutes) to give the mean movement level per minute of night-wakefulness = 1897.54 / 47.96 = 39.56 movements per minute of night-wakefulness for the Pretreatment phase of patient #1 (see Figure 19 in the Results section)
6: REFERENCES
Section 6: REFERENCES

6.1 TEXT REFERENCES


6.2 SOFTWARE REFERENCES

"ACTPLAN" C.G.S. Kramer, University Hospital Leiden, Dept. Clinical Neurophysiology, P.O. Box 9600, NL-2300 RC Leiden, The Netherlands.

"MINITAB" Release 7 (1989), Minitab Incorporated, 3081, Enterprise Drive, State College PA 16801, USA.

"SAS" Version 6 (1989), SAS Institute, Cary, NC, USA.