SLEEP PROBLEMS IN CHILDREN WITH DISABILITIES:
BEHAVIOURAL FAMILY INTERVENTIONS

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ABSTRACT

Sleep problems are frequently reported in children and studies indicate that approximately 15 to 30% of children experience some form of sleep difficulty (Partinen & Hublin, 2000; Richman, 1981; Zuckerman, Stevenson, & Bailey, 1987). Children’s sleep problems often impact on family members, in particular parents, causing a considerable amount of stress and frustration. Difficulties with sleep are a common problem for typically developing children; however, research indicates that the incidence of sleep problems is even higher in the disabilities population (Didden, Korzilius, van Aperlo, Overloop, & de Vries, 2002; Espie & Tweedie, 1991; Richdale, Francis, Gavidia-Payne, & Cotton, 2000; Saxby & Morgan, 1983). This has implications for families already under considerable amounts of stress and pressure of having a child with a disability.

The present study aimed to treat persistent sleep problems in children with disabilities using family behavioural intervention methods. A range of behavioural strategies was utilised to reduce sleep problems such as bed refusal, sleep onset delay, night waking, co-sleeping, and nightmares. Techniques such as a positive bedtime routine, reward systems, the parental presence programme, standard and modified extinction were used. In one case, a short-term decremental dose of a mild sedative (trimeprazine tartrate) was used in the initial stages of implementing a behavioural intervention to reduce child and parent distress. A “fear busting and monster taming” programme (White, 1985) was employed in conjunction with other behavioural techniques to reduce the occurrence of nightmares in another child. The results indicate that behavioural family interventions are effective in treating sleep problems in children with disabilities. The majority of the sleep behaviours targeted for intervention were eliminated or reduced to low levels of occurrence with 9 out of 11 target behaviours rated as showing a substantial improvement. These positives changes were maintained at follow-up with the exception of co-sleeping in Case Study Two. The social validity for the programmes was high and caregivers reported satisfaction.
Chapter 1. Introduction and Review of the Literature

Sleep disturbance is a commonly reported phenomenon in children. Childhood sleep problems impact heavily on family members, in particular the parents, adversely affecting family functioning and relationships (Richdale et al., 2000). Furthermore, sleep deprivation has been associated with behavioural problems (Zuckerman et al., 1987) and impaired intellectual and academic functioning (Gozal, 1998; Meijer, Habekotie, & Van Den Wittenboer, 2000; Wolfson & Carskadon, 1998). Children with disabilities are at greater risk of developing sleep problems than typically developing children (Didden et al., 2002; Saxby & Morgan, 1983) and are more likely to have sleep problems that are persistent in nature (Bartlett, Rooney, & Spedding, 1985; Quine, 1991). This highlights the need for interventions for sleep problems in children with disabilities.

Behavioural interventions have been successful in the long-term amelioration of sleep problems in the general paediatric population and there is a growing body of research supporting its use in children with disabilities. This present study aims to demonstrate the use of behavioural strategies in the treatment of sleep problems in four case studies of children with varying disabilities.

Overview of Sleep Development and Organisation in Children

To understand how sleep problems manifest in children, it is important to first be familiar with typical sleep organisation and development in children. Historically, sleep has been perceived as a period of mental and physical inactivity. This, however, is not an accurate description in that sleep is characterised by a complex architecture of sleep stages, periods of increased cognitive activity and arousal, and physiological changes. The scientific analysis of sleep is a relatively new science, only made possible by technological developments in the last 50 years. Dement and Kleitman (1957) used electroencephalogram (EEG, a technique using multiple electrodes on the scalp to gather electrical signals from the brain), to show the relationship of rapid eye movement and dream activity. Other methods of analysing sleep have since been developed. Ferber (1995a) provides an in-depth review of assessment methods in children such as behavioural observation, polygraphy (a diagnostic tool measuring multiple physiological variables), time-lapse video recording, and actigraphic monitoring (a small movement detector).

Mindell, Owens, and Carskadon (1999) present a comprehensive overview of sleep organisation and development. Anders, Sadeh, and Appareddy (1995) also summarise literature
on sleep state organisation in children in a chapter entitled *Normal Sleep in Neonates and Children*. Information presented in this section draws predominantly from these two sources.

Sleep is in fact a complex process involving two distinct states: rapid eye movement (REM) sleep and non-REM sleep. During REM sleep, cortical brain function is extremely active and levels of blood pressure, heart rate, and respiration are extremely variable. REM episodes increase in duration throughout the night, and the longest episodes occur in the early morning. Dreams and nightmares occur in this sleep state and therefore it is more likely for dreams and nightmares to occur in the second half of the night because of this cycle. Short periods of arousal also take place throughout the sleep cycle and people typically experience several episodes of wakefulness during a night of normal sleep. Most of the time however, an individual would return quickly to sleep and would not recall the waking. Non-REM sleep is divided into 4 stages, with stage 4 representing the deepest level of sleep. Stages 3 and 4 develop in the first year and are referred to as delta sleep or slow-wave sleep. It is believed that most of the restorative function of sleep occurs in the non-REM state.

It is important to note that sleep organisation and sleep requirements are different in infants, children, adolescents, and adults. The REM/non-REM cycle lasts approximately 50 minutes in infants, while in adults the cycle is longer, approximately 90 minutes duration. Infant sleep states are immature forms of REM and non-REM sleep states and are referred to as active and quiet sleep. Active (REM) sleep accounts for 50% of infant sleep and gradually reduces over time to adult levels of 25 to 30%. Children have higher amounts of delta sleep than adults. The amount of delta sleep also decreases over time as children mature, with a 40% decrease during adolescence and then a more gradual decrease throughout adulthood. Delta and REM sleep states are gradually replaced by intermediate non-REM stages of sleep. This is the reason why many children grow out of certain sleep problems associated with delta sleep such as sleepwalking and sleep terrors following puberty. Sleep requirements also change with age. Newborns typically sleep a total of 16 to 18 hours a day in 1- to 4-hour episodes (Ferber, 1985). As children develop, sleep becomes more consolidated to night-time and the need for daytime naps diminishes. By 3 years of age, children require on average 10.5 hours of sleep during the night with a 1.5-hour nap during the day. By 4 to 5 years of age, most children have given up daytime naps, requiring on average 11 hours sleep, and by 18 years of age, the average sleep requirement is 7 to 8 hours at night (Ferber, 1985). Developmental norms for sleep should be seen as a guideline as individual differences in sleep requirements occur.

Stores (2001a) summarised current knowledge of sleep-wake rhythms. He wrote that the timing of sleep is regulated by a *circadian clock* in the suprachiasmatic nucleus of the hypothalamus. Environmental cues or “Zeitgebers” (translates as “time givers”) such as light,
meal times, and social activities, as well as internal body signals such as hunger, temperature, and hormonal changes, help to synchronise the body’s circadian rhythm. It is commonly believed that without these Zeitgebers, the free-running sleep-wake cycle in humans is closer to 25 hours, causing the sleep-wake cycle to drift to increasingly later times. However, a study by Czeisler, Duffy, and Shanahan (1999) contests this theory and suggests that the intrinsic circadian cycle is close to 24 hours, consistent with other species (cited in Stores, 2001a).

Although much is known about the development and physiology of sleep, little is known about the actual function of sleep. Horne (1998) and Adam and Oswald (1984) proposed that sleep has a restorative function, while Berger and Phillips (1995) suggested that sleep has an energy conservation function. It has also been hypothesised that sleep contributes to brain organisation by maintaining and stimulating synapses that were under utilised during wakefulness (see Krueger, Obal, Kapas, & Fang, 1995 for a review) as well as by processing emotions and memory (Cai, 1995). It is likely that sleep serves multiple purposes (Rechtschaffen, 1998). A lack of sleep can significantly affect the functioning of an individual, and although the function of sleep has not been clarified, it is well understood that sleep is essential for a person’s physical and psychological wellbeing and is a basic need of humans (Ferber, 1985; Stores, 2001a).

DEFINITION OF CHILDREN

As previously discussed, sleep organisation and sleep requirements differ in infants, children, adolescents, and adults. This has implications for sleep research as the manifestation of sleep problems in an individual may vary according to his or her developmental age. There are other developmental issues that need to be considered and incorporated into the assessment and management of sleep problems. In infancy, there are feeding and care requirements that are specific to the developmental period. Infants are also highly dependent on caregivers in comparison to children, adolescents, and adults. Typically, developing children over the age of 2 years have increased verbal and cognitive ability and this can distinguish childhood from the period of infancy. Older children also have increased independent mobility as they develop gross motor skills such as crawling, walking, and running. These factors need to be taken into consideration during the assessment of sleep problems in children and in the development of an appropriate intervention plan (Ferber, 1985; France, 1989). France (1994) emphasised that higher verbal and cognitive abilities in children allow for more intervention options in the management of sleep disturbance. Adolescents and adults need to be differentiated from children because of factors such as increased independence and lack of dependence on parents, as well as increased cognitive ability. There are other issues for example, social, occupational, and
educational demands that are unique to adolescence and adulthood that may impact on sleep patterns. Therefore, sleep problems in adolescents and adults should be considered separately from those in children.

It is thus important to distinguish childhood as a separate developmental period from infancy, adolescence, and adulthood. For purposes of this research, children are defined as being between the ages of 2 and 12 years.

DEFINITION OF DISABILITY

A disability is a physical or mental impairment that may prevent, impede, or limit normal development; affecting an individual’s ability to learn, care for oneself, or complete tasks or activities that would be expected of his or her age. For this research, a disability was defined as an impairment that was physical or developmental in nature, whereby the parent(s) had to alter their parenting to cater for the child’s special needs. Mental health problems such as anxiety or depression were not included in this definition.

DEFINITION OF SLEEP PROBLEMS

A sleep problem can be defined as any pattern of sleep that is unsatisfactory to the child, his or her parents, or physician, that causes concern about the child’s well-being or the parents’ loss of sleep (Ferber, 1996). Parents may not only differ in what they regard as a sleep problem, they may also differ in their tolerance of bedtime behaviours (Ferber 1985; Stores, 2001a).

Stores (2001a) commented that what is considered a sleep problem is variable from one family to another, influenced by their cultural practices and belief systems surrounding children’s sleep requirements. Parental attitudes towards their children’s sleep are influenced considerably by cultural ideals and expectations (Lozoff, 1995). Cross-cultural studies have documented several cultural differences surrounding sleep. This is illustrated with the use of transitional objects across cultures. Although some practitioners view transitional object attachment to be a part of normal emotional development, studies have indicated that there is considerable cross-cultural variation. For example, Hong and Townes (1976) found that Korean children were less likely to be attached to a transitional object compared with American children (18% and 64% respectively). Cultural differences have also been noted in thumbsucking, breastfeeding, and in the patterns of parental interactions at bedtime (Lozoff, 1995). Co-sleeping is a normal part of sleep practices in many cultures. It is only in recent times that Western cultures have encouraged children to sleep alone (Brazelton, 1969). Studies in the United States have noted differences in co-sleeping between ethnic groups with approximately 50% of African Americans, 21% of Hispanics, and 10% of Caucasians co-sleeping with their children (Lozoff,
Wolf, & Davis, 1984; Mandansky & Edelbrock, 1990). Lower socio-economic status also reflected higher rates of co-sleeping, regardless of ethnicity (Lozoff et al., 1984). It is evident that cultural factors strongly impact on sleep practices and patterns and some authorities hold the viewpoint that sleep disturbance is a culturally constructed phenomenon (Bhavnagri & Gonzalez-Mena, 1997). Because of differing cultural beliefs and practices surrounding children’s sleep, it is important for clinicians not to impose their own cultural values upon a family (France & Blampied, 1999). Careful consideration of each family’s needs should be made.

There are developmental differences with sleep-associated behaviour, and what is normal behaviour in one age group may be concerning for another age group. For example, newborns under the age of 6 months require feeding during the night, necessitating night waking (Ferber, 1985). From 3 to 4 months of age, healthy full-term infants start to develop the ability to sleep through the night and no longer physically require a night feeding. If the child continues to have trouble sleeping through the night by 6 months of age, this suggests that something may be interfering with his or her sleeping (Ferber, 1985, 1996). To summarise, night waking is a necessary and normal sleep pattern for infants under the age of 6 months. Beyond this age in a healthy child, night waking can become problematic for the parents and/or child and may therefore be identified as a sleep problem. This highlights the importance of a developmental approach in the assessment of sleep problems in children.

According to the International Classification of Sleep Disorders: Diagnostic and Coding Manual, Revised (ICSD-R: American Sleep Disorders Association, 1997) sleep problems can be classified into two distinct categories: dyssomnias and parasomnias. Dyssomnias are sleep problems that relate to initiating or maintaining sleep, or excessive daytime sleepiness, whereas, parasomnias occur during sleep, and are behaviours that disrupt sleep after it has been initiated. Parasomnias common in childhood include sleepwalking, sleep talking, nocturnal enuresis, sleep terrors, confusional arousals, bruxism (teeth grinding), body rocking, and nightmares. Ferber (1995b) emphasised the importance of distinguishing between dyssomnias and parasomnias as they have different causes, frequencies, and treatments. The main focus of this current research is on dyssomnias often associated with inappropriate sleep onset associations and parental limit setting, namely bed refusal, sleep-onset delay, night waking, co-sleeping, and early morning waking. Definitions are presented below:

- **Bed Refusal**: difficulty getting the child to bed or to sleep at a regular time or when instructed (Blampied & France, 1993). This is also referred to as “bed refusal”, (Sanders, Markie-Dadds, & Turner, 1997), “bedtime delay” (Lawton, France, & Blampied, 1991), and “bedtime struggles” (Kataria, Swanson, & Trevathan, 1987). The terms “bedtime
difficulties” and “settling difficulties” have also been used to describe both bed refusal and sleep onset delay.

- **Sleep Onset Delay**: the length of time the child takes to fall asleep when first placed in bed at night-time; this may be the result of the child refusing to go to sleep or inability to fall asleep. This is often accompanied by demands and/or tantrums for the parents’ attention or desired rituals (e.g. bedtime story, food, drink, T.V.). Other terms such as “sleep refusal” (Kataria et al., 1987), “night settling problem” and “sleep onset latency” (Matthesuis, 2000; Weiskop, 2001) have been used interchangeably to describe this sleep problem.

- **Night Waking**: a waking during the night that occurs after the initial onset of sleep and before the normal time of waking in the morning, whereby the child signals to the parent as opposed to settling himself or herself to sleep (Adair & Bauchner, 1993). The child may signal using behaviours such as crying, calling out to the parent, or getting out of bed. Night waking often becomes a problem if it is persistent and constantly requires parental attention.

- **Co-Sleeping**: when a child sleeps and shares a bed with the parents or another family member for part of the night or the entire night. Co-sleeping is defined as a sleep problem if it is not part of the family’s cultural practices, but rather, utilised as a method to prevent disrupted sleep for the child and/or parent (Blampied & France, 1993).

- **Early Waking**: a time when the child wakes for the day that is considered too early and inappropriate by the parents. This is strongly influenced by social norms and expectations, such as school or work hours and meal times.

**FACTORS THAT MAY CONTRIBUTE TO THE DEVELOPMENT OF SLEEP PROBLEMS IN CHILDREN**

Armstrong, Quinn, and Dadds (1994) emphasised that learning to go back to sleep from regular wakings is an important developmental milestone. They stated that most children obtained their milestone of sleeping through the night before 4 months of age. An interesting finding was that a large proportion of children soon reverted to night waking. There are many factors that contribute to the development of sleep problems in children. These have been summarised into three broad categories for purposes of discussion: child characteristics, parent characteristics and parenting practices, and parent-child interaction variables. Additionally, a section on children with disabilities has been included to address factors specific to this population that may place them at greater risk of developing sleep problems.
Child Characteristics

Sleep State Organisation

Infants in particular are vulnerable to developing sleep disturbance because of the developmental characteristics of sleep organisation. Approximately 50% of infant sleep is spent in REM states. Furthermore, REM and non-REM cycles are shorter and more frequent in infants. This gives the infant more opportunities to wake as periods of arousal often occur after REM sleep. Short periods of arousal during the sleep cycle are common in people of all ages (see Anders et al., 1995 and Mindell et al., 1999 for comprehensive reviews). These arousals are a normal part of the sleep cycle; however, they may become a problem when the infant or child signals to the parent rather than soothing himself or herself back to sleep (Blampied & France, 1993; France & Blampied, 1999).

Child Temperament

Research has indicated that infants and children with sleep problems are more likely to have a difficult temperament (France and Blampied, 1999; Minde, Popiel, Leos, Falkner, Parker, Handley-Derry, 1993; Sadeh, Lavie, & Scher, 1994). Temperamental features include high activity scores, increased levels of crying, low malleability and rhythmicity scores, high irritability, and low adaptability and mood (Sadeh et al., 1994; Richman, 1981). Shaefer (1990) found a relationship between sleep problems and difficult temperament in his study of young children referred to a Crying Baby Clinic. Hayes, Parker, Sallinen, and Davare (2001) reported that children with sleep problems exhibited less adaptability and rhythmicity. Carey (1974) proposed that irritable children are less likely to be able to settle themselves back to sleep, because of high levels of cortical arousal. A study by Owens-Stively and colleagues (1997) also found that negative temperament characteristics were associated with clinically significant behavioural sleep disturbances in a study of 52 children. It is difficult to determine whether difficult temperament contributes to sleep difficulties, or whether these characteristics are caused by sleep disturbance (Sadeh et al., 1994).

Child Health

Numerous medical factors have been implicated to contribute to the development of sleep difficulties in children. Medical issues such perinatal complications (Minde et al., 1993), otitis media, teething, effects of medication (Ferber, 1995b), asthma (Stores, Ellis, Wiggs, Crawford & Thomson, 1998), and epilepsy (Cortesi, Giannotti, & Ottaviano, 1999) have been associated with sleep problems. Ghaem et al. (1998) found a clear association between infant sleep disturbance and gastro-oesophageal problems. Hagemann (1981) in a study of children aged 3 to 8 years
found that hospitalisations due to chronic illness disrupts children’s sleep resulting in sleep loss of up to 25%. Secondary sleep disturbance may thus be precipitated by a disruptive event such as child illness and hospitalisations, as interactions between parent and child may alter (France & Blampied, 1999). Medical problems may trigger the development of a transient sleep problem, and in some cases, this may mark the beginning of a more chronic sleep problem, in which “dysfunctional” interactions continue and maintains the sleep problem (Benhamou, 2000). Adult studies have shown that chronic pain may also impact on sleep quality (Atkinson, Ancoli-Israel, Slater, Garfin, & Gillin, 1988; Smith, Perlis, Smith, Giles, & Carmody, 2000). Smith et al. (2000) found that sleep complaints were reported by 88% of adults in their study who had chronic pain.

Fears and Anxiety

Stressful and traumatic experiences, such as war and natural disasters as well as more common events such as changing school and separation from siblings and parents, can have significant effects on the sleep patterns of children (Sadeh, 1996). Developmental factors such as the fears or anxieties in older children may also contribute to sleep problems (Ferber, 1996). Mindell et al. (1999) described how anxiety and fears relating to bedtime could be learned through simple conditioning. For example, if the child is sent to the bedroom as punishment, the bedroom may become associated with anxious feelings. Another example given by the authors is when the child awakes distressed after a nightmare the parent may attend to the child and turn the light on. The child may learn to associate the dark with distress and nightmares, and associate light with comfort.

Parental Characteristics and Parenting Practices

Parenting Practices in Relation to their Child’s Sleep

There is a growing body of research that suggests that sleep-state organisation is influenced by differing styles of care, and evidence from studies with pre-term infants indicates that co-ordinated, low-stimulating, and rhythmic care are associated with positive sleep patterns (Gabriel, Grote, & Jonas, 1981). Weisbluth (1987) commented that over-attentive parents may stimulate an infant excessively, inadvertently depriving the infant of the opportunity to learn how to fall asleep unassisted. France and Blampied (1999) described parental behaviours such as putting the child to bed asleep, being present when the child falls asleep, and co-sleeping, as techniques which block the development of self-soothing behaviours. It has been suggested that parents inadvertently contribute to sleep problems when they rock, hold, or feed their infants at
bedtime (Ferber, 1985). Infants and children may therefore, learn inappropriate sleep onset associations.

According to a review of literature by Ferber (1995), providing food and/or drink during the night are associated with increased night wakings in infants and toddlers. Infants given a bottle or breastfed at bedtime may associate this condition with sleep onset, thus requiring feedings during the night to reinitiate sleep after a waking. If an infant is fed regularly at night, they may also become conditioned to waking during the night because of hunger or because of soiled nappies (Stores, 2001a). It is important to remember that healthy infants over the age of 3 to 4 months no longer physically require night feeding (Ferber, 1985).

Research by Adair, Bauchner, Phillipp, Levenson, and Zuckerman (1991) found that infants whose parents were present at bedtime were significantly more likely to wake at night than infants whose parents were not present (40% vs. 22%). The authors suggested that infants were likely to have associated parental presence with sleep onset, so when they woke during the night parental presence was required to re-establish sleep. A study by Johnson (1991) found that 80% of infants who fell asleep on their own were able to sleep through the night, compared to less than a third of infants who were soothed (nursed, rocked, or comforted) to sleep by their parents at bedtime. This furthermore supports the notion of sleep onset conditioning of parental presence, the consequence of inadequate stimulus control.

Henderson (2001) demonstrated that there were several variables at 1 month of age that strongly predicted the emergence of sleep disturbance at 6 and 12 months of age. Her study employing discriminant function analysis with 52 infants, found that sleep state organisation in infants (i.e. frequent night awakenings) and two parent behaviours (being present at sleep onset and co-sleeping) at age 1 month, predicted group membership to the emerging sleep-disturbed group or non sleep-disturbed group in 90.4% of infants at age 6 months. Infant sleep state organisation and parental presence at sleep onset were two variables found to strongly predict group membership to the sleep-disturbed or non sleep-disturbed groups at 12 months of age in 86.5% of infants. The study found that parents of sleep-disturbed infants were more likely to employ different strategies to manage infant sleep compared to parents of non sleep-disturbed infants, such as being present at sleep-onset, placing the infant in the cot asleep, and co-sleeping. These parents were also more likely to intervene during a night awakening. This study supports the association between parental practices and sleep disturbance, but also acknowledges the contribution made by individual child characteristics.

Night-time difficulties may stem from inadequate limit setting by the parents. Ferber (1995b) described the transition from a crib to a bed as an opportunity for young children to test this new found freedom. No longer restricted physically by the crib, it is important for parents to
establish other methods of setting limits. Parents may also give in to their child’s request for extra stories, drinks, and so forth, and not set appropriate limits. Bedtime struggles, the child’s reluctance to adhere to parental requests to go to bed, is often a by-product of the parents’ inability to place boundaries on behaviour. Inappropriate parental expectations may also be a factor contributing to night-time difficulties. A child’s inability to go to sleep because of an inappropriate bedtime (e.g. too early) may be misinterpreted as naughtiness and non-compliance. Expectations around sleep therefore need to be developmentally appropriate (Stores, 2001a). Stores (2001a) suggested that children in disorganised families with unstructured lifestyles are likely to develop sleep irregularity, increasing the likelihood of sleep problems. Ferber (1985) and Stores (2001a) emphasised the importance of a regular bedtime routine and bed time, as well as “sleep hygiene”; practices that promote sleep for example, minimising or eliminating caffeine intake before night-time and engaging in quiet activities before bedtime.

Parental Cognitions and Perceptions

As discussed earlier, it is normal for infants and children to wake briefly a number of times during sleep. Generally, the infant or child is unaware of this arousal and returns to sleep promptly. Difficulties occur when the child awakens and signals to the parents. Ferber (1995b) suggested that when parents become aware of the night waking they might incorrectly conclude that wakings are abnormal. As a result, they may assume that help is required and become involved in the sleep transition process by assisting the child to sleep. The child as a result will learn this help process and become unable to make this transition back to sleep alone. Armstrong et al. (1994) found that 50% of parents stayed with their child as a strategy to settle him or her to sleep during the night. Ironically, this form of parental response during night wakings may in fact reinforce the problematic sleep behaviour in children.

Ferber and Boyle (1983) noted that parents often believed that sleep problems in children were characterised by frequent night wakings. The authors however made an important distinction and commented that the child’s inability to re-initiate sleep without parental intervention constituted the sleep disturbance, not the number of night wakings, as night wakings are a normal part of sleep state transitions. It is probable that this parental belief may elicit a parent to target night waking as a “problem” by using strategies such as feeding, staying with the child, and co-sleeping.

Maternal Depression

Mothers of sleep disturbed infants and children have higher levels of malaise or depression (Matthesius, 2000; Minde et al., 1993; Richman, 1981; Zuckerman et al., 1987).
Armstrong et al. (1994) found that parents of poor sleepers tended to be over-permissive, chronically anxious, or depressed. Although there is a strong association between children’s sleep problems and maternal factors such as depression, stress, and anxiety, the causal direction of this relationship is not clear (Armstrong et al., 1994). Furthermore, Armstrong and colleagues (1994) suggested that mothers may in fact be experiencing sleep deprivation guised as depression. It is valuable to note that research has indicated improvements in maternal affect as a result of effective treatment of sleep problems in children (Minde et al., 1993).

**Parent-Child Interaction Variables**

**Attachment**

Bedtime may be viewed as a time of separation for the child from his or her parents (Ferber, 1985). Settling difficulties such as bed refusal may therefore stem from separation anxiety or separation difficulties. Benoit, Zeanah, Boucher, and Minde (1992) found that 100% of mothers of toddlers with sleep problems were insecurely attached in comparison to 57% of mothers in a control group. Although maternal attachment may be a contributing factor, the fact that over half the control group were also insecurely attached implies that attachment is not the only influence in problematic sleep behaviour.

**Signallers versus Self-Soothers**

The distinction between signallers and self-soothers is important, as this may contribute to the development of problematic parent-child interactions. Infants or children who cry or request parental attention when they awake are commonly referred to as “signallers”, while infants who are able to resume sleep without arousing parents are referred to as “self-soothers” (Anders et al., 1995; Stores, 2001a). Anders et al. (1995) remarked that the problem of night waking is more appropriately redefined as a problem with the infant’s response to the awakening and not the actual event of awakening per se. In a review of literature, Anders et al. (1995) concluded that infants who were put to bed awake were more likely to return to sleep on their own. This suggests that the ability to return to sleep following a night waking resembles the pattern of falling asleep at bedtime. Children who do not have the ability to self-sooth are more likely to signal to parents, and are thus more susceptible to developing sleep problems. Additionally, parents who comfort and assist their child to sleep may in fact be denying him or her opportunity to develop self-soothing behaviours. This interaction between parent and child of signalling and responding is part of the coercive behaviour process.
Behaviour Traps

The development of many sleep problems in children can be explained from a social learning perspective (Blampied & France, 1993; France & Blampied, 1999). Sleep can be viewed as a biological process that is integrated with behavioural components. Interactions between children and parents around sleep can influence whether the child develops appropriate sleep behaviours or not. France, Blampied, and Henderson (2003) emphasised that behavioural aspects of sleep are vulnerable to learning influences similarly to other aspects of children’s behaviour. Sleep has a reinforcing function for children and their parents, increasing the probability of the behaviour that precedes it (Blampied & France, 1993; France & Blampied, 1999). France and Blampied (1999) described this learning process of sleep in infants as follows. In the development of normal sleep, the infant will be reinforced for quiet and calm behaviours and self-soothing strategies, while the parents will be reinforced for providing appropriate proximal cues for sleep onset such as regular time cues, a simple and non-stimulating bedtime routine, and quiet time to allow sleep onset. During night awakenings, the child is reinforced for engaging in self-soothing behaviours by the resumption of sleep while parents are reinforced for providing attention that is non-reinforcing for the infant in its nature and intensity. The process is different for infants with sleep disturbance. In the case of the sleep-disturbed infant, the infant’s behaviour of signalling to the parents is reinforced by parental attendance and stimulation. The infant learns to gain parental attention by signalling and develops an association between the stimulating parental presence and the onset of sleep, increasing the likelihood of signalling to the parent upon awakening during the night. Furthermore, the infant will be reinforced for distress behaviours such as crying, behaviours that essentially compete with sleep. In this model, parents are reinforced for engaging in inappropriate behaviour and providing inappropriate proximal cues for sleep onset, including intensive stimulation such as rocking, feeding, or co-sleeping.

The reinforcement mechanism underlying these inappropriate parent-child interactions is referred to as a behaviour trap, namely a coercion trap (Patterson, 1982). Within this cycle, both the child and his or her parents learn to avoid aversive stimuli by engaging in certain behavioural repertoires. The child learns to avoid the negative and distress-provoking circumstances of falling asleep alone, while the parent learns to avoid the child’s distressed behaviour. Parents in Richman’s study (1981) reported attending immediately to their child when they heard them cry to prevent further disturbances. This may be an immediate resolution however the interaction would feed into the coercive cycle.

Behavioural traps often pose difficulties for parents when they attempt to withdraw their attention (the reinforcement) for bedtime demands and signalling. The parents change of repertoire and withdrawal of reinforcement, results initially in an increase in crying and other
distress behaviours known as the “post-extinction response burst” (PERB), (Kazdin, 1984; Lerman & Iwata, 1995). The parents may misinterpret this response and assume that they are making the problem worse, and resume the reinforcement, the attending behaviours. Unfortunately, resumption of attention during the PERB reinforces the child’s signalling and distress behaviours at a more intense level, resulting in behaviours that are also more resistant to change. Furthermore, the parents’ experience of the PERB is likely to cause avoidance of further attempts to withdraw reinforcement, thereby strengthening the coercion trap (France & Blampied, 1999).

Factors Associated with Sleep Problems in Children with Disabilities

The literature indicates that children with disabilities are more susceptible to developing sleep problems. There are several factors that may predispose children with disabilities to develop sleep difficulties. These factors could be physical or psychological in nature, or a combination of the two.

It is suggested that children with disabilities are at greater risk of developing sleep problems due to physiological abnormalities. It is suspected, for example, that underlying brain maldevelopment and damage in children with severe intellectual disability may disrupt or alter sleep physiology and structure causing sleep problems. Okawa and Sasaki (1987) proposed that children with intellectual disabilities might have impaired perception of Zeitgebers, impacting on the establishment of the circadian rhythm. They further hypothesised that children with intellectual disability are possibly more susceptible to sleep problems because of neurological malfunctions or brain damage in areas responsible for sleep. In addition, children with intellectual disabilities are at increased risk of other conditions such as epilepsy, which is known to disrupt sleep (Okawa & Sasaki, 1987).

Children with physical disabilities such as cerebral palsy may have difficulty getting comfortable during the night due to reduced mobility and may also have sleep disrupted by discomfort and pain. Pain may prolong sleep onset and may interfere with the quality and quantity of sleep in children (Lewin & Dahl, 1999). Children with Down syndrome were shown to have significantly higher rates of sleep related breathing problems and symptomology associated with sleep apnoea, suggesting that the physical characteristics of Down syndrome places children at risk of developing a sleep problem (Stores, Stores, & Buckley, 1996).

Intellectual, learning, or communicative impairments may interfere with the development of good sleep habits. Teaching a child with an intellectual disability may be more difficult because of problems with communication and understanding. Johnson (1996) emphasised in her review of sleep problems and children with intellectual disability and autism that children’s
ability to learn self-soothing skills are likely to be delayed or markedly impaired. Quine (1991) reported that incontinence and adaptive skill deficits, such as communication, academic and self-help skills, were strongly associated with sleep problems in children with learning disabilities. Parental perception of their child’s disability may also contribute to the development and maintenance of sleep problems. Parents of children with intellectual disabilities may perceive their child’s sleep problem as an inevitable and untreatable part of his or her disability (Stores & Wiggs, 2001; Wiggs & Stores, 1998). Didden et al. (2002) found that 25% of parents attributed their child’s intellectual disability as the reason for the development of sleep difficulty. Studies have shown that parents of children with a disability who have sleep problems are more responsive to their child at night (Quine, 1991, 1992). Didden et al. (2002) furthermore found that parents of children with intellectual disability who had sleep problems used strategies such as co-sleeping (26.3%), providing food or drink (11.4%), and offering a night-time activity (8%) when their child woke during the night. These interactions are likely to maintain persistent night waking in children.

Stores and Wiggs (2001) commented that parents’ attitudes, parenting ability, and wellbeing, are influenced and affected by their child’s condition, resulting in some parents becoming overly permissive and inconsistent in their parenting of their child. Some parents may feel unable to discipline or impose boundaries on their child’s behaviour because of their disability (Wiggs & Stores, 2001b). Additionally, the stress of parenting a child with a disability may impact on family functioning and thus increase the likelihood of falling into behaviour traps, such as those described by France and Blampied (1999). Blum (1999) found that mothers’ general over-concern about their child’s needs and health was associated with night waking in typically developing infants. This finding could be applied in area of children with disabilities, as disabilities in children are likely to elicit concern from parents about health and wellbeing of their child, creating susceptibility for parents to become over-responsive to the needs of their child at night. Furthermore, medical conditions such as asthma and epilepsy may necessitate the parent to attend to the child at night and limit the parents’ ability to ignore their child because of concerns about safety. This may place children who are less able to communicate their needs at greater risk. It is also important to note that parents who are fatigued and under considerable amount of stress would be less able to be consistent in their parenting.

MEASURES OF SLEEP IN CHILDREN

Parental report is one of the most common and cost-effective measures used to assess sleep behaviours in infants and children. The types of parental report include: questionnaires, interviews, sleep logs, charts and records, and sleep diaries. Questionnaires and interviews are
used frequently in the assessment of paediatric sleep patterns to gather information on the sleep patterns and behaviours of children (Armstrong et. al., 1994; Richman, 1981; Van Tassel, 1985). These however, collect information that is retrospective in nature. Sleep logs, charts and records are used to gather prospective information on the length and frequency of sleep periods of children, and are often presented as a form or chart for parents to code. Sleep diaries also record the length and frequency of sleep patterns, but have the added benefit of being able to gather a variety of information relating to parent and child behaviours, such as time in bed, duration to sleep onset, frequency and duration of night awakenings, and the parental behaviours at bedtime and on night awakening (France & Hudson, 1990; Richman, 1981; Minde et al., 1993). France (1989) concluded that the combination of parental diaries and interview are the most common measures used by studies.

Direct observation methods such as infrared time-lapse video recording (TLVR) and actigraphic home monitoring have been used to assess infant sleep. The TLVR was originally used by Anders (1979) to record infant sleep-wake behaviour. The video equipment and microphone is placed by the infant’s cot to record behaviour throughout the night. This method has been increasingly used to research infant sleep-state development as well as to check the reliability of parental report measures (France & Blampied, 1999; Minde et al., 1993). Actigraphic home monitoring employs a small-computerised movement detector that is worn on the child’s arm or leg. This correlates highly with Non-REM, REM, and wake states (Sadeh, Lavie, Scher, Tirosh, & Epstein, 1991). Actigraphy alone however does not record important parent and child behaviours or interactions.

Polysomnography, a method that records brain electrical activity (EEG), motor activity, heart rate, respiration, and eye movements can provide a large amount of information about the child’s sleep, however is very costly to implement. Furthermore, it is typically conducted in a laboratory setting and requires the child to stay overnight. Because this method has economic and practical limitations it is often reserved to assess sleep problems with a suspected physiological cause such as sleep apnoea or epilepsy (Durand, Mindell, Mapstone, & Gernert-Dott, 1998; Kuhn, Mayfield, & Kuhn, 1999).

Henderson (2001) proposed a hierarchy of measuring infant sleep based on France’s hierarchy (1989). The measures ranked from most to least desirable are: i) direct measurement of both infant and parental behaviour as used by Anders (1979); ii) prospective parental measures such as daily diaries; iii) sleep questionnaires; and iv) time specified interviews.

Parental diaries have been shown to have high validity. Anders (1979) compared TLVR recordings with parental daily diaries and reported a high correlation between measures. Other studies have similarly found a high correlation between TLVR and parental sleep diary records.
(France & Blampied, 2001; Minde et al., 1993). It is recommended that daily diaries be completed consecutively for more than 3 days (France & Hudson, 1990).

Sleep diaries are a practical and cost-effect measure of sleep and has proven validity. France (1989) and Henderson (2001) advocate the direct measurement of infant and parent behaviour as the most desirable measure. The TLVR method however, has been implemented in studies of infant sleep and not with older, mobile children. Therefore, direct measurements with TLVR may have practical limitations with children.

PREVALENCE OF SLEEP PROBLEMS IN TYPICALLY DEVELOPING CHILDREN

It is well established that sleep difficulties are a common problem associated with childhood and there have been several studies evaluating the prevalence of sleep problems in the general childhood population. Studies report that between 15% and 37% of children are described by their parents as problem sleepers (Owens Spirito, McGuinn, & Noblie, 2000; Richman, 1981; Salzarulo & Chevalier, 1983).

Sleep problems are especially common in infants and young children. Armstrong et al. (1994) assessed a range of sleep behaviours in their Queensland survey of 3269 typically developing children (from 1 month to 38 months of age) and found that 28.6% of children had problematic sleep behaviour, including settling difficulty and night waking. Richman (1981) conducted a community survey of 1- to 2-year-old children and estimated from her findings that 13 to 20% of 1- to 2-year-olds woke regularly (more than 3 times a week) while 6 to 10% had severe sleep disruptions. Zuckerman et al. (1987) completed a 3-year longitudinal survey and assessed sleep problems in children at 8 months of age and then at 3 years of age. The results showed that 22% of children aged 8 months had sleep problems, waking every night. At 3 years of age, 29% of the children had sleep problems. They reported that 41% of children who had a sleep problem at 8 months of age continued to have sleep problems at 3 years of age. This suggests that many sleep problems are persistent.

Sleep problems are not isolated to infants and young children and although sleep problems are reported to have the highest prevalence in the preschool years, parents frequently report sleep problems in older children. Salzarulo and Chevalier (1983) in their study of 2- to 15-year-old children reported that 32% of children sleep talked, 31% had nightmares, 28% experienced night wakings, 23% had trouble falling asleep, and 17% had nocturnal enuresis. In the renowned Isle of Wight study, Rutter and colleagues (1970) found that 20% of 10- to 12-year-olds were regarded as having sleep problems. In a more recent study, Owens et al. (2000) indicated that 37% of children between the ages of 4 and 11 years had significant sleep problems, the most common problem being bedtime resistance (15.1%). Although the prevalence of sleep
problems decreases with age, there is evidence to suggest that many children do not simply
"outgrow" sleep problems as commonly perceived. Quine (2001) found in her sample of children
from mainstream schools that 37% of 4- to 5-year-old children had settling problems. Although
the proportion of children with settling problems reduced with age, settling difficulty still
affected 25% of children 9 to 12 years of age. A longitudinal study by Richman, Stevenson, and
Graham (1982) found that sleep problems that persisted at 3 years of age were more likely to
become chronic. Their study showed that two-thirds of 3-year olds described as problem sleepers,
continued to be described as such at a 5-year follow-up. Pollock (1994) similarly found that
children with sleep problems at 5 years of age were likely to continue to have sleep related
difficulty at 10 years of age.

PREVALENCE OF SLEEP PROBLEMS IN CHILDREN WITH DISABILITIES

Research suggests that there is a higher rate of sleep problems in children with
disabilities than in typically developing children. Some studies report prevalence to be as high as
80 to 87% in this population (Bartlett et al., 1985; Quine, 1992). Prevalence studies of sleep
problems in children with disabilities have produced varied results. Didden, Curfs, van Driel, and
de Moor (2002) conducted a study of 286 children with mild to profound intellectual disability
aged 1 to 19 years in the Netherlands and found that 23.7% had a sleep problem, such as night
waking and severe settling difficulty. An interesting finding from this study was that the
prevalence of sleep problems increased with the severity of the children’s intellectual disabilities.
Sleep problems affected 8.6% of children with mild, 14.8% of children with moderate, 27.9% of
children with severe, and 35.3% of children with profound intellectual disability. The authors
also noted that sleep problems were common in children with epilepsy (31%) and cerebral palsy
(33.3%). Furthermore, they found that most parents attended to their child when they woke
during the night and although most children took only a few minutes to return to sleep, 28.7%
took around 30 minutes to settle after a night waking, 7.2% took up to an hour, and 4.3% took on
average 1 to 2 hours to settle. This indicates the severity of night waking for some children in the
study, and the impact of sleep problems on family members especially the parents.

Clements, Wing, and Dunn (1986) completed a survey of 163 children with severe
intellectual disability aged 15 years and younger and found that 34.2% had sleep difficulties that
were regarded as problematic by their parents. Sleep problems that were addressed in the study
were night waking and limited hours of sleep per night. Hayashi and Katada (2002) used a
parental questionnaire to evaluate sleep disturbance in 670 children and adults (aged 1 to 43
years) with intellectual disability. The study reported that 75% of children and adults
experienced night waking while 15% had sleep onset disturbance.
Bartlett et al. (1985) used a questionnaire to assess sleep problems in 214 children with an intellectual disability and found that 80% of children in the study were described as having a mild to severe sleep problem by their parents. Additionally, the results showed that a large proportion of older children experienced sleep problems, suggesting that sleep problems were more persistent in the population with disabilities. The reports of sleep problems were high in all age groups with 86% of children under 6 years of age, 81% of 6- to 10-year-olds, and 77% of 11- to 16-year-olds.

Wiggs and Stores (1996a) found that 44% of children with intellectual disability aged 5 to 16 years had a current sleep problem, the most common being settling difficulty and night waking. A concerning finding from this study was that the average duration of sleep problems was 7.13 years. This suggests that children with an intellectual disability are not only are more susceptible to sleep problems, they are at greater risk of developing a persistent sleep problem. Quine (1991) completed a longitudinal study with children with severe intellectual disability and reported that 51% had settling problems and 67% had night waking. A large number of children who had sleep problems were reported to have epilepsy (33%): There were also a significant proportion of children with cerebral palsy with sleep problems (71%). The study found that two-thirds of children who had sleep problems continued to exhibit sleep problems at a 3-year follow-up, which is suggestive that sleep problems are very persistent in this population.

Another study by Quine (1992) reported that 65% of children (aged 5 to 20 years) with severe learning difficulties had settling problems, while 87% had night waking problems. Out of those who had settling difficulty, 61% took over 2 hours to sleep most nights. A third of children with night waking problems woke 3 or more times a night. The study also found that 32% of children slept regularly in their parent’s bed and 11% slept in a sibling’s bed. In a more recent study, Quine (2001) compared 576 children from mainstream schools with 182 children with learning disabilities in special schools aged 4 to 12 years. The results showed that sleep problems occurred more frequently in children with learning disabilities. Settling problems presented in 41% of children from special schools compared to 27% of children from mainstream schools, night waking 45% compared to 13%, and early waking 14% compared to 5%. An important finding from the study was that the frequency of sleep problems in children with learning disabilities remained high in older groups of children. In children with learning disabilities aged 9 to 12 years, settling problems affected 41% compared to 25% of children in mainstream schools, and night waking affected 38% compared to only 6% of children in mainstream schools. Quine (2001) suggested that children with learning disabilities were less likely to outgrow problems without intervention.
A study by Richdale et al. (2000) assessed the prevalence of sleep problems in children with an intellectual disability and children who were typically developing between 2 and 17 years of age. Sleep problems were reported in 57.7% of children with an intellectual disability compared to only 16% of controls. Sleep problems in children with intellectual disability were also more persistent, with over half of the children having a sleep problem for 2 or more years. Children with an intellectual disability also had a higher rate of night waking, with 68.9% described as frequent night wakers compared to only 17.4% of children in the control group. Co-sleeping was also found to be a common issue for children with intellectual disability (42.3%). Saxby and Morgan (1983) completed a survey of behaviour problems in children with learning disabilities and found that sleep problems were the highest reported behaviour problem with 72% of children described by their parents as having night waking and settling difficulty. Sadly, 19% of parents stated that they felt they could not cope with their child’s sleep problem.

Stores et al. (1996) compared the sleep habits of children with Down syndrome, with their siblings, as well as children from the general population, and with children with intellectual disability other than Down syndrome. They found that children with intellectual disability (Down syndrome and other forms of intellectual disability) were more likely to have a sleep problem than the control groups. The authors also reported that children with Down syndrome were more likely to have symptoms that were characteristic of sleep apnoea and sleep related breathing problems, such as snoring, gagging or choking, mouth breathing, and restlessness, whereas children from other groups were more likely to present with sleep problems associated with initiating and maintaining sleep.

Richdale, Cotton, and Hibbit (1999) examined sleep and behavioural disturbances in children with Prader-Willi syndrome. The study compared 29 children with Prader-Willi syndrome (aged 6 months to 46 years) with an age- and gender-matched control group. The results showed that there was a higher rate of sleep problems including excessive daytime sleepiness, snoring, and early waking as well as behavioural disturbance in the group of children with Prader-Willi syndrome. Excessive daytime sleepiness was a distinctive feature of this group and the authors suggested that this might be related to problems with the sleep-wake cycle and hypothalamic dysfunction. Fragile X syndrome is another disability that has been associated with sleep problems. Richdale (2003) reported that 31% of children with fragile X syndrome had a current sleep problem in a study of 13 children aged 3 to 19 years. Sleep problems were associated with clinically significant behavioural problems and parental stress.

A review by Richdale (1999) indicated that children with autism had higher rates of sleep difficulties compared to other groups of children with disabilities and typically developing children. Studies have estimated prevalence as high as 56% (Clements et al., 1986) and 83%
(Richdale & Prior, 1995). Richdale and Prior (1995) compared two groups of children: Group One included 12 children with autism with moderate to severe intellectual disability and Group Two included 27 children with autism and mild intellectual disability to normal intelligence. Both groups were compared with 35 typically developing children. The results indicated higher rates of specific sleep problems in children with autism, such as sleep onset delays, long periods of night waking, short night sleep, and early morning waking, particularly in children aged 8 years and under. Additionally, it was found that the lower functioning group of children with autism had higher rates of daytime napping and slept earlier in the evening, in comparison to the higher functioning group of children with autism.

Schreck and Mulick (2000) compared sleep problems in children with autism with children with other forms of intellectual disability, children in special education, and a control group of typically developing children. The study found that children with autism had significantly higher rates of dyssomnias and parasomnias than the other groups of children.

Most prevalence studies have addressed sleep problems in children with developmental disability and intellectual disability, and research on children with physical disability or chronic illness is sparse. Some studies have indicated that children with chronic physical illness or disabilities are also at higher risk of developing sleep disturbance than typically developing children. Sleep may be affected by night-time discomfort or pain of atopic dermatitis (Stores, Burrows, & Crawford, 1998), juvenile rheumatoid arthritis (Zamir, Press, Tal, & Tarasiuk, 1998), chronic pain (Lewin & Dahl, 1999), and burn injury (Lawrence, Fauerbach, Endell, Ware, & Munster, 1998 cited in Stores, 2001a). Medical conditions have also been implicated to disrupt sleep such as asthma (Loughlin & Carroll, 1995; Stores, Ellis et al., 1998) and epilepsy (Cortesi et al., 1999). This is a very under-researched area.

There are several factors that could explain the variance of prevalence results in this population. A review by Didden and Sigafsoos (2001) suggested that the disparity of prevalence of sleep problems in children with disabilities could be due to the low response rate for questionnaires, as well as the use of subjective measures such as parental sleep logs. Another possible factor is the definitions used in studies to assess sleep problems in children. Individual researchers may use different criteria and cut-offs for sleep problems, as well as determine whether they are mild, moderate, or severe. This may produce variation between studies. Another factor that may impact results is the type of sleep problems investigated in the studies, as well as the number of sleep problems addressed. For example, Bartlett et al. (1985) addressed settling and night waking problems, while Clements et al. (1986) looked at limited hours of sleep and night waking. Quine (2001) studied settling difficulty, night waking, co-sleeping, and early waking, whereas Stores et al. (1995) not only assessed a range of dyssomnias, such as settling
and night waking problems, but also investigated sleep-related breathing problems and sleep apnoea. Most studies however, have focussed on dyssomnias, primarily settling difficulty and night waking (Didden et al., 2002, Hayashi & Katada 2002; Quine 1991, 1992).

The type of disabilities investigated by studies would also affect prevalence results. Many studies have treated children with intellectual disabilities as a group without considering the different aetiologies or disorders underlying the intellectual impairment or learning disabilities of individual subjects (Bartlett et al., 1985; Clements et al., 1986; Hayashi & Katada, 2002; Quine, 1992). This is significant as studies indicate that certain disability subgroups may be more at risk, such as epilepsy (Didden et al., 2002; Quine, 1991), cerebral palsy (Didden et al., 2002; Quine, 1991), autism (Schreck & Mulick, 2000), fragile X syndrome (Richdale, 2003), Prader-Willi syndrome (Richdale et al., 1999), and Down syndrome (Stores et al., 1995). Another consideration is the severity of the disability within the sample. For example, Didden et al. (2002) included children with mild, moderate, and severe intellectual disability whereas other studies (e.g. Quine, 1991) have focused on the severe range of intellectual disability. Other factors that may affect prevalence studies are limited sample size (Richdale et al., 2000; Quine, 1992) and a lack of an experimental control group for comparative measures (Bartlett et al., 1985; Clements et al., 1986; Didden et al., 2002; Hayashi & Katada, 2002; Wiggs & Stores, 1996a; Quine, 1991, 1992).

These factors may contribute to the disparity of prevalence results. Nevertheless, it is clear that children with disabilities are at greater risk of developing sleep problems than their typically developing counterparts. Most studies have presented prevalence above 40% in children with disabilities (Bartlett et al., 1985; Quine, 1992, 2001; Richdale et al., 2000; Saxby & Morgan, 1983; Wiggs & Stores, 1996a). The majority of studies to date have treated children with disabilities as a homogeneous group. Further research is necessary to address sleep problems in various disability subgroups.

IMPACT OF SLEEP PROBLEMS ON THE INDIVIDUAL AND THEIR FAMILY

Cognitive Function and Educational Performance

There is evidence from studies of adults to suggest that sleep disturbance and deprivation can have a significant impact on psychological functioning (Bonnet, 2000). Sustained sleep loss has been shown to negatively affect cognitive functioning, in particular memory, sustained attention, and visual-spatial abilities (Horne, 1988; Pilcher & Huffcutt, 1996). Most studies have been confined to adults due to ethical limitations of experimental research in children of this nature. Although research is limited, there is evidence to suggest that sleep loss has similar effects on children. Wolfson and Carskadon (1998) in their study of adolescents found that sleep
loss was associated with daytime sleepiness and impaired school performance. Gozal (1998) also demonstrated a relationship between sleep disordered breathing and poor academic performance in children. A study by Meijer et al. (2000) furthermore supports the notion that sleep loss in children is associated with daytime sleepiness and impaired performance at school. These findings have significant implications for children with disabilities as their learning is often compromised by their disability.

Mood and Behaviour

Research has indicated that poor sleep is associated with negative mood changes in adults with increased signs of irritability, aggression and depressed mood (Dahl, 1996; Pilcher & Huffcutt, 1996). In children, the relationship between sleep problems and behavioural problems has been supported by several studies. Zuckerman et al. (1987) found that 3-year-old children with persistent sleep problems were more likely to present with behavioural problems such as tantrums. The authors suggested that persistent sleep problems were part of the behavioural difficulties between parents and their children involving issues of limit setting and boundaries. In a study of children 2 to 5 years of age (Lavigne et al., 1999), a strong relationship between the total amount of sleep and daytime behaviour problems was indicated. Children aged 2 to 3 years who had low amounts of sleep (less than 10 hours in a 24 hour period) were more likely to have high levels of externalising behaviour problems as indicated on the Child Behaviour Checklist, such as hyperactivity, oppositional behaviours, non-compliance, and aggression. This relationship was less pronounced in the older age group of 4 to 5 years. The association between sleep problems and behaviour problems appears to be more marked in younger children. Studies such as Gruber, Sadeh, and Raviv (2000) have shown a relationship between sleep difficulty and attention deficit hyperactivity disorder (ADHD). They suggested that the instability of the sleep-wake system is a characteristic of ADHD. Additionally, shortened or disrupted sleep has been associated with ADHD-like symptoms (Dahl, 1996), emphasising the importance of thorough assessment practices and the awareness of differential diagnosis. A study by Kuhn, Lund, and Olesh (1998) compared the sleep disturbance in children with ADHD and children with generalised behaviour problems and concluded that sleep disturbance was more closely related to the severity of the behaviour problem and not attention and arousal. The sleep disturbance between the two groups could not be differentiated.

Studies have also found a close relationship between sleep problems and daytime behaviour problems in children with intellectual disabilities (Didden et al., 2002; Richdale et al., 2000). Wiggs and Stores (1996b) reported that children with severe learning disabilities with sleep problems showed significantly more types of challenging behaviour than children without
sleep problems. The severity of challenging behaviour was also greater in the group with sleep problems. The types of challenging behaviour reported were self-injury, aggression, screaming, temper tantrums, non-compliance, and impulsivity. According to the authors, children with sleep problems were also more likely to show daytime irritability, lethargy, hyperactivity and stereotypic behaviour.

Parents and Family Functioning

Sleep problems in children can have adverse effects on parents and family members as well as on family relationships and functioning (Stores, 1996). Chavin and Tinson (1980) suggested that in some circumstances marital discord and separation could result from children’s sleep problems. Armstrong et al. (1994) found that a significant proportion of parents of children with sleep problems were chronically anxious or depressed. Gelman and King (2001) also demonstrated that mothers of children exhibiting sleep disturbance reported significantly higher levels of overall psychopathology and stress. It was found that mothers had higher levels of stress, anxiety, and depression than controls as indicated by a self-report measure. Research in children with disabilities has found that parents especially mothers are negatively affected by their child’s sleep problem. Richman (1981) for example found that mothers of children with intellectual disabilities who had sleep problems had higher rates of psychiatric disturbance and were more likely to report feeling irritable and out of control. Richdale et al. (2000) also says that children’s sleep problems can strain family functioning.

 Mothers of children with sleep problems in Quine’s (1991) study of children with intellectual disability reported higher levels of stress and irritability than mothers of children without sleep problems. Sleep problems were furthermore perceived to have a significant impact on the family by the mothers. In another study of children with intellectual disability, Didden et al. (2002) showed that parents were adversely affected by their child’s sleep problems. According to the study, parental fatigue was reported in 47%, irritability reported in 28%, and failure to cope reported in 9% of parents. Furthermore, 20% of parents stated that they experienced inadequate sleep because of their child’s sleep problem. In support of this association, Mindell and Durand (1993) noted positive change in the parents’ moods and relationships as a result of successful behavioural intervention in ameliorating children’s sleep problems. Wiggs and Stores (2001a) found that successful behavioural treatment for sleep problems in children with severe intellectual disabilities and daytime behaviour problems had a positive impact on mothers. Mothers in the study reported reduced stress, increased perception of control and ability to cope, and more satisfaction with their own sleep following treatment for their child’s sleep problem.
Parent-Child Relationships

A study by Quine (1992) suggested that mothers of children with intellectual disability and severe sleep problems were more irritable and less affectionate towards their children. Mothers in the study who had a child with a severe sleep problem were also more likely to use more physical punishment. Armstrong et al. (1994) also found that 6% of parents whose children had sleep problems resorted to smacking their child when they woke during the night. Chavin and Tinson (1980) suggested that sleep problems, may in some circumstances trigger child abuse. It is evident that sleep problems can be stressful for the child and parent and has potential to strain relationships between them.

INTERVENTION OPTIONS FOR SLEEP PROBLEMS IN CHILDREN WITH DISABILITIES

The main approaches for the treatment of sleep problems in children with disabilities are pharmacological and behavioural approaches. Treatment options for sleep related breathing disorders such as surgery are not discussed in this review they are beyond the scope of this dissertation. Although a brief overview of pharmacological interventions is presented, the main focus will be on behavioural interventions. There is increasing evidence to suggest that behavioural options used to treat typically developing children are effective in treating children with disabilities. Wiggs and France (2000) reviewed the literature on behavioural treatments for children with physical illness, psychological problems or intellectual disabilities and concluded that behavioural interventions were successful in treating sleep problems. The authors noted that behavioural strategies employing a more gradual approach, such as graduated extinction, may be more suitable for some children with special needs because of safety concerns with planned ignoring methods, such as in the case for some children with epilepsy or asthma. Additionally, graduated strategies may be more acceptable for parents who feel concerned for their child’s wellbeing and unable to ignore the child completely, or for parents who are anxious about starting an intervention.

Behavioural interventions discussed include bedtime fading, chronotherapy, sleep-wake scheduling, positive routines, stimulus control, standard extinction, and modifications of extinction. It is important to note that although behavioural interventions have been divided into categories for discussion, many studies do not use a single technique but rather employ a variety of behavioural strategies (Bartlet & Beaumont, 1998; Bramble, 1997; Durand, Gernert-Dott, & Mapstone, 1996; Weiskop, 2001). These combined approaches have been shown to be successful in ameliorating sleep problems in children with disabilities.
Pharmacological Interventions

Sedative medication is the most common form of intervention for sleep problems in children (Adair & Bauchner, 1993; Chavin & Tinson, 1980). A New Zealand study found that medical practitioners frequently prescribed sedative medication to manage paediatric sleep disturbance, resulting in a third of children being prescribed sedatives by age 5 years (Werry & Charlielle, 1983 cited in France & Hudson 1993). Armstrong et al. (1994) found that 4% of children (38 months and younger) were prescribed sedative medication for their sleep problems in Australia. Disturbingly, the study showed that very young infants were given pharmacological treatment, with 2% of children aged less than 3 months and 7% of children aged 13 to 18 months receiving medication for their sleep difficulties. Quine (1992) reported that 7% of children who had severe learning disability were receiving medication for their sleep problems. Wiggs and Stores (1996a) found that pharmacological treatment was the most common form of intervention offered to children with severe intellectual disabilities, with 36% of children who had sleep problems being prescribed sedative medication. Only 58% of parents reported that medication was a helpful form of treatment for their child. In contrast, 79% of parents who received behavioural advice reported that it was helpful. The most commonly prescribed drugs for sleep disturbance in children include antihistamines such as trimeprazine tartrate (Valergen), benzodiazapines, tricyclic antidepressants, and chloral hydrate (Adair & Bauchner, 1993).

Despite widespread use, there have been very few clinical trials of medication for the treatment of children’s sleep problems. Ramchandani, Wiggs, Webb, and Stores (2000) compared drug treatments with behavioural treatments in a review of randomised controlled trials for settling problems and night waking in young children. The main outcome of this review was that drug treatments were effective in treating night waking in the short-term, but long-term efficacy was questionable. In contrast, behavioural interventions showed both short-term and long-term efficacy. Richman (1985) carried out a double-blind drug trial using trimeprazine tartrate in 22 children (aged 1 and 2 years) with severe night waking problems and found that improvements were only moderate, with frequent night wakings still occurring. A third of the children showed no improvement, even on relatively high doses of trimeprazine. Furthermore, the drug did not produce any long-term effect on sleep patterns, with sleep problems persisting in the majority of the children at a 6-month follow-up. Simonoff and Stores (1987) also examined the efficacy of trimeprazine for night waking in children aged 1 to 3 years. The controlled trial with a sample of 18 children reported that children on treatment with trimeprazine had significantly fewer night wakings, less time awake at night, and more night-time asleep compared to those on treatment with placebo. A 1-month follow-up found no significant difference in sleep from baseline measures. The authors suggested that trimeprazine may be
useful for short-term relief of sleep problems in children or otherwise used as a precursor for further treatment such as a behavioural programme.

France, Blampied, and Wilkinson (1999) investigated the effect of trimeprazine on infant sleep disturbance in a multiple baseline, double-blind study of 12 infants aged 6 to 27 months. The administration of trimeprazine failed to show clinically significant improvements in sleep disturbance. A positive drug-effect was more evident in higher doses of trimeprazine however this was not consistent across all cases. The study found that only one child in the low-dose group and two children from the high-dose group had Sleep Behaviour Scale Scores (Richman, 1985) during the drug treatment phase that were below the criterion for problematic sleep. Drug related improvements were not maintained in the final baseline phase for any of the children. The authors concluded that trimeprazine was not effective as a treatment for infant sleep disturbance. The use of the sedative was not recommended unless in combination with a behavioural intervention.

Studies have indicated that medication is not effective in treating sleep disturbances in children in the long-term. Results show that the efficacy of sedative medication is limited and its usefulness restricted to short-term relief. Medication is not a popular choice with parents and it is often met with resistance (Richman, 1985; Simonoff & Stores, 1987). France and Hudson (1993) also comment that little is known about the safety of sedative medication in children, which is concerning because of its common use in the treatment of paediatric sleep problems despite behavioural treatment alternatives. Moreover, there are associated problems with the use of medication such as tolerance, dependence, paradoxical effects, daytime drowsiness, and other side effects (Dahl, 1992; Mindell et al., 1999). Furthermore, the use of medication fails to consider the underlying cause of the sleep problem and is likely to interfere with the child learning to fall asleep (Stores, 2001a). Many prominent authors discourage the use of sedative medication in children (France et al., 1999; Johnson, 1996; Mindell et al., 1999; Stores, 2001a). Behavioural interventions are more favourable and there is a growing body of research demonstrating the efficacy of behavioural treatments in children.

**Behavioural Interventions**

**Bedtime Fading**

Bedtime fading involves shifting the sleep-wake cycle of the individual to an earlier time every night. Baseline data is gathered to determine a time when rapid sleep onset is highly probable. Once an average sleep onset time is established, 30 minutes is initially added to the baseline and the individual is not allowed to fall asleep prior to this time. If the child achieves sleep onset within 15 minutes, then the bedtime is shifted 30 minutes earlier the following night.
If the child does not initiate sleep within 15 minutes then the bedtime is made 30 minutes later on the subsequent night. Piazza and Fisher (1991b) used this procedure to treat 2 children with severe sleep problems: a 6-year-old girl with ADHD with bed refusal, night waking, and nocturnal enuresis, and a 4-year-old girl with profound intellectual disability and tuberous sclerosis who presented with daytime sleepiness and delayed sleep-wake cycle. Several treatments had been attempted previously in both children without success. The 4-year-old girl was treated in an inpatient unit, while the 6-year-old girl was treated by her caregiver on an outpatient basis. The bedtime fading protocol resulted in improved sleep-wake times and amount of sleep in subjects, as well as reduced night waking and nocturnal enuresis. The authors noted that the procedure had the added benefit of eliminating “power struggles” between parent and child surrounding bedtime because the child was instructed to go to bed when the probability of compliance is high. Follow-up data was not obtained in the study.

Bedtime fading with a response cost protocol has also been used to treat multiple sleep problems in children with developmental disabilities. The response cost component consists of removing the child from bed for 1 hour when sleep onset is not initiated within 15 minutes. It is hypothesised that the removal of the child from the bed would act as a contingency, increasing the child’s motivation to fall asleep quickly to avoid the aversive situation of remaining awake for an extra hour. The effects of tiredness would also increase the chances of the child falling asleep quickly. Piazza and Fisher (1991a) used this approach with 4 children who were non-verbal with developmental delay and profound intellectual disability to treat severe sleep problems such as excessive daytime sleepiness, sleep onset delays, and night waking. The majority of children (3 out of 4 children) were treated in an inpatient unit. All children showed significant improvements in the percentage of appropriate sleep, and decreases in night wakings. No follow-up was undertaken. Piazza, Fisher, and Sherer (1997) compared bedtime fading with response cost and bedtime scheduling for sleep problems in 14 children with developmental disabilities who had been admitted to an inpatient unit for severe behaviour problems that posed a danger to self and/or others. Children were randomly assigned to a treatment group and were treated during their stay in the inpatient unit. They found that bedtime fading with response cost improved children’s sleep significantly more than the bedtime scheduling method. The study did not conduct a follow-up and thus the long-term efficacy of the intervention was not ascertained.

Studies of bedtime fading and bedtime fading with response cost have shown significant improvements in severe sleep problems in children with disabilities however research in these procedures is very limited. There have been only a few studies conducted, most of which have been limited with small sample sizes. Another limitation of all three studies addressed in this review was the omission of follow-up data. Long-term efficacy of this intervention therefore has
not been addressed. Additionally, studies have demonstrated the use of bedtime fading (with and without response cost) in an inpatient unit and so further research would be necessary to assess the efficacy of the procedures in the home environment. The systematic and laborious procedure may make it difficult to implement for families. Bedtime fading does, however, show promise in the treatment of severe sleep-wake problems in children with disabilities and may be an appropriate option for when other treatments have been unsuccessful. Despite high practical demands of the intervention, faded bedtime with or without response cost may be appealing for parents as there is no element of ignoring the child.

Chronotherapy

Chronotherapy has been used to treat severe sleep phase delays (Dahl, 1992). Chronotherapy involves systematically delaying the bedtime of the individual each night, shifting the sleep-wake cycle around the clock until the desired bedtime is reached. The therapy is hypothesised to be effective because it takes advantage of the free-running circadian cycle. Piazza, Hagopian; Hughes, and Fisher (1998) demonstrated the use of chronotherapy to treat severe sleep problems (irregular sleep onset times, frequent night and early wakings, and short total sleep times) in an 8-year-old girl with autism and severe intellectual disability. “Jan” was admitted to an inpatient unit where her bedtime was delayed by 2 hours every night. The time she was allowed in bed was set to a total of 10 hours based on developmental norms for sleep. During waking time, an attempt was made to simulate Jan’s normal daily schedule, consisting of therapy sessions, school, leisure activities, and meals. An appropriate bedtime for her age was achieved within 11 days, and 4 months of follow-up indicated that improvements were maintained in the home.

The use of chronotherapy is limited as it is labour intensive and requires constant monitoring, and thus would not be widely applicable in the home. The around-the-clock procedure would make it extremely difficult to implement because of disruptions to the family schedule. Chronotherapy therefore may be a useful intervention for children with severe sleep-wake patterns when other treatments have been unsuccessful. The implementation of chronotherapy however, may be most suited to an inpatient setting because of the high practical demands of the strategy and the stringent monitoring process. Piazza et al. (1998) indicated that chronotherapy may be more effective than faded bedtime procedures in situations where sleep onset had drifted significantly beyond the point where shifting to an earlier time was likely to be successful (according to the authors more than 3 hours). Shifting the bedtime forward in these circumstances may also resolve sleep-wake problems more quickly than faded bedtime.
Research on chronotherapy is very limited and further studies would be helpful to establish efficacy of the treatment.

Sleep-Wake Scheduling

Sleep-wake scheduling involves setting a fixed time schedule for going to bed and for waking. The morning waking time is set, and daytime and evening naps are prevented. Average sleep requirements are taken into account and the individual is sent to bed when readiness for sleep is shown, this is referred to as the “threshold time”. The threshold time is then adjusted until 90% sleep efficiency (the time asleep divided by the time in bed multiplied by 100) is achieved. Espie and Wilson (1993) showed that sleep-wake scheduling was effective in reducing sleep problems in 5 children and adults with intellectual disabilities, however this was a preliminary investigation and more research is needed to determine the usefulness of this intervention. This study did not obtain follow-up information and so long-term effects of treatment were not determined. Other limitations of the study were the lack of a control group and small sample size.

Positive Routines

Positive routines involve the parents establishing a bedtime routine of quiet activities that the child enjoys. Initially the bedtime is set to the naturally occurring bedtime and is gradually shifted to an earlier time until the desired bedtime is achieved. If a tantrum occurs the parents are required to terminate the activities and tell the child it is time for bed (Adams & Rickert, 1989). Mindell (1999) considered positive routines to be a promising intervention for typically developing children with sleep problems, using the Chambless Criteria (Chambless & Hollon, 1998). The Chambless criteria were developed by the Task Force on Promotion and Dissemination of Psychological Procedures in 1995 in order to evaluate treatment efficacy. Treatments can be described as well established, probably efficacious, or a promising intervention based on the amount of between-group designs, multiple baseline or case studies conducted (Chambless & Hollon, 1995). There have been only a few studies addressing the use of positive routines for the treatment of sleep problems in typically developing children, with one larger well-controlled study (Adams & Rickert, 1989) and two small studies employing a within-subject design (Galbraith, Pritchard, & Hewitt, 1993; Milan, Mitchell, Berger, & Pierson, 1981). All three studies demonstrated successful treatment outcome. There has been only one study to date that has used positive routines in the population of children with disabilities. Milan et al. (1981) used positive routines in 3 children with severe intellectual disability aged 2, 4 and 15 years to reduce bedtime tantrums. All 3 children showed significant improvements at bedtime.
Results from these initial studies have been promising and further research would be valuable to examine the efficacy of positive routines in typically developing children as well as in children with disabilities. The use of positive routines would be an appealing intervention option for parents, as it does not involve systematically ignoring their child’s behaviours. Instead, the approach focuses on positive interactions between parent and child, and sets out to make bedtime an enjoyable time instead of a time characterised by child resistance. Positive routines have been successful in treating settling problems such as bed refusal. Many studies incorporate the use of a positive bedtime routine as a component of the treatment (Seymour, Bayfield, Brock & During, 1983; Weiskop, Mathews, & Richdale, 2001). It may be useful in combination with other behavioural techniques for the treatment of multiple sleep problems such as settling difficulties, night waking and co-sleeping.

Stimulus Control

Howlin (1984) demonstrated the use of stimulus fading in the treatment of settling difficulties, night wakings, and co-sleeping in a 6-year-old boy with autism. During baseline, the mother had resorted to co-sleeping with the child every night. The procedure involved the mother initially sleeping on a mattress in the child’s bedroom immediately next to the child’s bed so when the child woke she could cuddle and soothe him as usual. The position of the mattress was gradually changed increasing the distance between the mother’s mattress and the child’s bed. Approximately halfway through the intervention, the mother was requested to stop touching the child and use only verbal ways to comfort the child. The mattress was moved gradually towards the bedroom door, and then placed immediately outside the bedroom. The mother was able to return to her own bed in the next phase of the procedure. The intervention was successful in eliminating co-sleeping and reducing night waking and settling difficulties in the child. These improvements were maintained at a 3-month follow-up.

Allison, Burke, and Summers (1993) used a structured bedtime routine and stimulus control methods to treat co-sleeping in an 8-year-old girl with Down syndrome and profound intellectual disability. The stimulus control strategy involved replacing the mother with an almost life-size doll and gradually withdrawing the mother’s presence from the child’s bed and then the bedroom as outlined by Ferber (1985). At baseline, the child spent an average of 6% of the night asleep alone, and at the end of the treatment, this was increased to 78.2%. Improvements were maintained at 6-month follow-up.

Although results from these single-case studies are promising, methods employing stimulus control requires further research. Many interventions however, incorporate an element of stimulus control.
Standard Extinction

Standard extinction has been found to be an effective intervention and many studies have shown rapid and long lasting improvements in settling and night waking problems for typically developing children as well as children with disabilities. Standard extinction has also been referred to as unmodified extinction, systematic ignoring, standard ignoring, and planned ignoring. The extinction process requires the parents to ignore the child’s disruptive behaviours throughout the night. The parents are instructed to place the child in bed, say goodnight and leave the room, and then not attend to the child until the morning unless the child is ill or in danger of hurting himself or herself. Extinction thus provides neither reinforcement nor punishment for the child’s behaviours. Extinction is seldom used on its own and in fact, by its nature involves changing parent-child interactions and settling events. France and Hudson (1993) commented that extinction programmes invariably have an element of stimulus control, where regular bedtimes and bedtime routines are established, and the parents do not attend to the child unless it is deemed necessary.

The use of standard extinction procedures for sleep problems was first demonstrated by Williams (1959) to treat bedtime tantrums in a 21-month-old boy. Bedtime tantrums were effectively eliminated with the procedure and the child settled without tantrums on the third night on the programme. Mindeli (1999) classified extinction as a well-established intervention using the Chambless criteria (Chambless & Hollon, 1998). Kuhn and Elliott (2003) also evaluated extinction as being a well-established intervention based on the Chambless criteria (Chambless & Hollon, 1998). There is a considerably large body of research supporting the efficacy of extinction in typically developing children, with controlled studies (France et al., 1991; France, 1992; Rickert & Johnson, 1988), studies with large sample sizes (Richman, Douglas, Hunt, Lansdown, & Levere, 1985; Seymour, Bayfield, Brock, & During, 1983) and multiple baseline studies (France & Hudson, 1990; Seymour, 1987).

Standard extinction has also been successful in treating sleep disturbance in children with disabilities, and there have been several studies supporting its use. Didden et al. (2002) used extinction to treat settling and night waking problems in three children (aged 1 year 6 months, 6 years 5 months, and 7 years 3 months) and an adult (aged 25 years) with developmental disabilities. The multiple baseline design showed that extinction was successful in reducing disruptive night-time behaviours in all four participants, and effects were maintained at a 6-month follow-up. Didden, de Moor and Kruit (1999) demonstrated the use of extinction with a 2 1/2-year-old boy with a physical disability and near-normal intellectual functioning. The intervention successfully reduced night waking and a 3-month follow-up indicated no incident of disruptive night-time behaviours.
Dickens, Curfs, Sikkema, and de Moor (1998) presented six case studies of children with developmental disabilities and sleep problems aged 2 years, 4 years, 6 years, and 7 years. Analyses found that four children had sleep problems that were maintained by parental attention, one child had settling difficulty relating to conditioned anxiety, while an undiagnosed seizure disorder was found to be associated with night-time crying in another child. Intervention in the form of behavioural treatments (i.e. extinction and desensitisation), and pharmacological treatment (anticonvulsant medication) was used successfully in all six children respectively. Follow-up data obtained after 6 months indicated that changes were maintained. Extinction was shown to be an effective treatment for children whose sleep problems were maintained by parental attention. There were some methodological limitations associated with this study. First was the lack of inter-rater reliability data for sleep measures. Parents in the study refused to record behaviours independently. The study also used AB and B designs, with two case studies with missing baseline data. This study furthermore, did not incorporate an experimental control.

Weiskop et al. (2001) presented a case study of a 5-year-old boy with autism, with settling difficulties, night waking, and co-sleeping. The parents had stated two goals for intervention: the child would settle alone at bedtime, and sleep in his own bed for the entire night. Prior to starting the extinction procedures, the parents were taught how to implement a positive bedtime routine (the routine was presented on a pictorial chart for the child), and to give clear instructions. The parents were also taught how to use reinforcement procedures and to utilise these techniques when the child completed a component of the bedtime routine. The bedtime routine was taught to the child with the use of role-play and modelling with a doll. After the second week of intervention, the boy was able to fall asleep on his own and sleep alone in his bed for the entire night. By the end of the programme, both goals stated by the parents had been achieved with 100% success, and improvements were maintained at a 3-month and 12-month follow-up. The development of an appropriate intervention plan addressing the child’s learning needs lead to a successful outcome for the child.

Weiskop (2001) demonstrated the use of a parent-training programme to reduce sleep problems in six children with autistic spectrum disorder and seven children with fragile X syndrome aged between 2 and 9 years. The programmes incorporated training in bedtime routines, reinforcement, effective instructions, partner support strategies, and standard extinction. The results showed that the programme was effective in reducing the frequency of settling problems, night waking and co-sleeping to clinically significant levels in both groups. These positive outcomes were maintained at 3-month and 12-month follow-ups. The programmes however, were not effective in reducing early morning wakings, night rocking, or daytime naps.
The study reported high social validity indicating that the programme strategies were acceptable to the parents.

Thackeray and Richdale (2002) similarly taught reinforcement, instruction giving, partner support strategies, bedtime routines, and standard extinction to parents of three boys with intellectual disability aged 5 to 10 years with sleep problems. At the time of the referral, all three children required parental presence to fall asleep, two children had night waking difficulties, and two children had co-sleeping difficulties. After intervention, all three children were able to fall asleep independently. Co-sleeping was eliminated for both children, and night waking was reduced in one of the two children. Improvements were maintained at a 3-month follow-up. Two children experienced a PERB and one family reportedly took their child back to bed 259 times on the first night of the programme. The study supported the use of behavioural strategies with children with intellectual disability and the programme had high social validity. The difficulty of the PERB and the need for parental commitment to behavioural strategies is highlighted.

A study by Bramble (1997) incorporated extinction with stimulus control and cueing, to treat chronic sleep problems in 15 children (aged 3 years 6 months to 12 years of age) with severe learning disability. Rapid improvements in settling, night waking and co-sleeping behaviours were indicated within a few days of treatment. A 4-month and 18-month follow-up indicated that improvements in sleep patterns were maintained. Although the study had a reasonable sample size, the design lacked an experimental control.

Bartlet and Beaumont (1998) conducted a larger study, with 57 children with disabilities or chronic illness. Children included in the study had various disabilities (severe learning disability, Down syndrome, Rett’s syndrome, cerebral palsy, tuberous sclerosis, autism, blindness) or chronic illness (chronic upper-respiratory tract infections, ear problems, severe deafness, eczema, asthma, epilepsy and coeliac disease). The treatment incorporated cueing, graded change (breaking down the sequence of change into discrete steps for the parent to teach the child, e.g. falling asleep on their own), extinction, and positive reinforcement. Improvements were reported in 45 children (79%) and were maintained at a follow-up completed 3 to 6 months post-treatment. The study did not employ an experimental control.

Studies investigating the use of extinction in children with disabilities are still limited. There have been a number of single case reports (Didden et al., 1999; Weiskop et al., 2001) and several multiple baseline studies (Didden et al., 2002; Thackeray & Richdale, 2002; Weiskop, 2001) describing promising results. The results from single case and multiple baseline studies are suggestive that extinction is an effective behavioural intervention for children with disabilities, but due to small sample size and lack of experimental control, the results cannot verify its efficacy. Larger studies in this population group are sparse. There have been no controlled
randomised trials of extinction employed in children with disabilities to date. Research by Bramble (1997), Bartlet and Beaumont (1998) despite larger sample sizes are limited by the lack of experimental control groups. Future randomised controlled trials of extinction would be valuable in children with disabilities to determine its efficacy in this population.

France and Hudson (1993) noted that there are some problems associated with the use of extinction despite its proven efficacy. One such problem is parental acceptance of the procedure. Studies have noted that parents may be unwilling to use extinction because of concerns about ignoring their child and the belief that this may be harmful to the child and detrimental to the parent-child relationship. This concern may be heightened in parents of children with disabilities. Rickert and Johnson (1988) found that subject attrition was more prominent in families assigned to the extinction condition. Another difficulty with the extinction procedure is the PERB (France & Hudson, 1990; Rickert & Johnson, 1988), as the increased intensity of the child’s distress behaviour, such as crying and calling out to parents, may make it difficult for parents to ignore.

Two studies have investigated whether extinction has harmful effects on the child. France (1992) evaluated the behaviour characteristics and security scores of infants treated with extinction. The study compared 35 infants treated with extinction with 13 infants with sleep problems (untreated) and 15 normal sleep controls and found that there were no detrimental effects associated with extinction. In fact, the study found that security/emotionality/tension, and likeability scores improved as a result of treatment. Sanders, Bor, and Dadds (1984) similarly reported that preschoolers treated with extinction and stimulus control were not adversely affected by the procedure. Despite concern that extinction may have detrimental effects, research indicates that it does not have adverse consequences for the infant or child. Although, extinction has shown rapid results in treating sleep problems, the short-term distress that it may cause in parents and the child during the PERB, may still make this an unpopular treatment option for parents.

Chadez and Nurius (1987) attempted to address parental concerns in a case study of a 7-month-old girl. As part of the therapeutic process, the authors explored parental concerns about ignoring the child and leaving her to cry and beliefs about being a “bad parent” and presented alternative cognitions (e.g. “it’s better in the long turn not to pick her up right now”). The authors reported significant improvements in bedtime crying and night waking difficulties with the use of extinction. A follow-up 6 months and 12 months later revealed that improvements in sleep had been maintained. France, Henderson, and Hudson (1996) advocate a similar approach in an article entitled ‘Fact, Act, and Tact’: A 3-stage approach to treating the sleep problems of infants and young children. Fact involves informing the parent about infant and childhood sleep disturbance, and the difference between self-soothers and signallers, and the contributing factors
in the development of sleep problems, such as behaviour traps. Act refers to choosing an appropriate programme, informing the parents about the advantages and disadvantages of programme options, and supporting the parents to implement the chosen intervention, while Tact refers to addressing parental concerns, such as conflicting advice they may have been given in the past, and concerns about the possible impact the intervention may have on their relationship with their child. Using the 3-stage model may minimise parental concern and increase the likelihood of the completion of a successful treatment. This in fact is built into all good parent training programmes.

Parental doubt around the extinction procedure may lead to difficulties complying with the requirements of extinction; that is being able to consistently ignore all child behaviours at night-time. France and Hudson (1993) state: “The application of planned ignoring (extinction) has the potential to worsen night waking if the parents inconsistently implement the procedure and, thus, place the child’s night waking on an intermittent schedule of reinforcement” (p. 96). Standard extinction is therefore unsuitable for parents who feel unable to ignore their child and with children with serious medical conditions.

Modifications of Extinction

Due to parental concern about the practices of extinction procedures, modified versions of extinction have been explored. Because of difficulties parents typically encounter with the PERB, modifications of extinction have aimed to reduce this. Three types of modified extinction will be discussed: graduated extinction, the parental presence programme, and extinction in combination with trimiprazine tartrate. The most widely used modification is graduated extinction. This technique allows the parent to attend to the child and involves increasing the time interval before the parents attend to the child’s waking on each subsequent night. Parents are advised however, to keep interactions to a minimum. The most well known graduated procedure is described by Ferber (1985) in his book; Solve Your Child’s Sleep Problems. According to Mindell (1999) using the Chambless criteria (Chambless & Hollon, 1998), graduated extinction is considered probably efficacious given that there is a well-controlled, randomised study (Adams & Rickert, 1989) and a number of within-subject and multiple baseline studies (Durand & Mindell, 1990; Lawton, France, & Blampied, 1991; Mindell & Durand, 1993; Pritchard & Appleton 1988). Since the publication of Mindell’s (1999) review there has been another controlled study advocating the use of graduated extinction (Reid, Walter, & O’Leary, 1999). More recently, Kuhn and Elliott (2003) classified graduated extinction as a well-established intervention using the Chambless criteria (Chambless & Hollon, 1998).
Durand et al. (1996) investigated the use of a consistent bedtime routine combined with a graduated extinction procedure in four children (aged 2 years, 7 years, 11 years and 12 years) with varying developmental disabilities: Down syndrome, moderate intellectual disability due to a chromosomal abnormality, pervasive developmental disorder, and autism. Prior to treatment, two of the children had frequent night wakings and two children had bedtime disturbances. Individualised bedtime routines were developed for each child. The graduated extinction procedure adapted from Ferber (1985) involved gradually increasing the amount of time before the parents responded to the child. Parents were advised to attend to the child in a brief manner, telling the child to go back to sleep in a neutral tone of voice. The amount of time the parents were required to wait before attending was variable and was based on what the parents were comfortable with doing. Therefore, some parents responded after 2 minutes and increased this delay by 2 minutes on each subsequent night, while others were more comfortable with longer delay intervals, beginning with 5 minutes and increasing by 5 minutes on each subsequent night. A multiple baseline approach was utilised and results showed that this procedure was effective in the treatment of settling and night waking in all of the children. The authors commented that graduated extinction might have a paradoxical effect. By increasing the delay before the parent attended to the child it would seem that the parent would reinforce successively longer bouts of crying, which should in fact make the problem worse. Evidence from studies of graduated extinction suggests, however, that the relatively neutral behaviours of the parent while attending, may reduce the reinforcing effect of the interaction. The procedure changed the nature of the parent-child interactions. Prior to treatment, parents reported lying in bed with the child or using other soothing behaviours until the child fell asleep.

France et al. (1996) commented that graduated extinction has the advantage of being more acceptable with parents as it allows them the opportunity to check on their children. A disadvantage of the programme is that it may take longer than standard (unmodified) extinction programmes, requiring the parents to deal with crying and distressed behaviours for a longer period. Parents need to be committed and organised to carry out the programme, and careful not to increase the intensity of their contact throughout the procedure. There is strong research support for graduated extinction, with two controlled studies and many multiple baseline and single-case studies in typically developing children. More studies would be valuable in children with disabilities. The results of the study by Durand et al. (1996) with children with developmental disabilities are promising.

The parental presence programme is another modification of standard extinction. In this procedure, a parent sleeps in the child’s bedroom in a separate bed while standard extinction is carried out. Once the parent puts the child to bed and says goodnight, the usual procedure of
ignoring the child is implemented. If the child cries, the parent is advised to lie in the bed and feign sleep. The parent is able to leave the room once the child is asleep, returning to the room as soon as the child wakes. Again, in this circumstance the parent does not attend to the crying and instead pretends to sleep. The programme depends on the child being aware of the parent’s presence; therefore, sufficient light is necessary for the child to see the parent. The parent may also need to cough to indicate to the child they are in the room. Parental presence is implemented for about 1 week, after which the parent returns to sleep in his or her usual bed and a maintenance programme is used. The efficacy of this approach has been demonstrated by a between-groups design (Sadeh 1994), and a multiple baseline across subjects design (France & Blampied, 2002). Using the Chambless criteria (Chambless & Hollon, 1998), Kuhn and Elliot (2003) recognised extinction with parental presence as a promising intervention. According to France and Blampied (2002) there is some evidence to suggest there is a decrease in PERB with this approach.

Parental presence has the advantage of rapid treatment effects of standard extinction, but also has the added benefit of reducing infant crying and parental anxiety. The programme is suitable for parents who are willing to temporarily change their sleeping arrangements, or for children and parents who are not used to separation. This treatment is unsuitable for parents who feel unable to ignore their child’s crying without attending (France et al., 1996). Parental presence is a promising treatment option and further research would be valuable to assess its efficacy. There is no known research to date in children with disabilities.

Another alternative approach involves the short-term use of trimeprazine (a sedative known to reduce and not eliminate night wakings) in combination with standard extinction procedures. France, Blampied, and Wilkinson (1991) applied this approach to treat sleep disturbance in 45 infants aged 7 to 27 months. The rationale for using trimeprazine in decreasing doses over the first 10 days of extinction was that the short-term sedative effects might reduce the child’s initial reaction to the onset of extinction (the PERB), thereby reducing parent and child distress. The objective was to modify extinction so that it would be more acceptable for parents. Infants were randomly assigned to one of three treatment groups (extinction plus trimeprazine, extinction only, or extinction plus placebo). A double-blind procedure was used to allocate the trimeprazine or placebo. All three groups showed significant improvements in sleep disturbance, and these gains were maintained at follow-up. Measures of infant security and maternal anxiety were also improved with treatment. The reduction of night wakings was more immediate for children using the combined approach of medication and extinction. There was however, a slight rebound effect with the withdrawal of medication. The authors also noted that many parents in the medication group commented negatively on the “doped-up” condition of
their child during the daytime. France et al. (1996) commented that the combined approach has the advantage of minimising infant and parental anxiety and produces rapid change. A disadvantage may be the possible daytime effects of medication and the slight rebound effect associated with medication withdrawal. This approach may be suitable for parents who are extremely anxious about starting a programme for infants and children who have developed conditioned fear of the cot or bed, or vomiting. Some parents may find the use of medication unacceptable, despite its short-term use. Note that some children with intellectual disability in particularly autism can have paradoxical effects to such drugs (A. Richdale, personal communication, May 2004). Further investigation of this combined (pharmacological/behavioural) approach is necessary, as research is very limited with no current research in children with disabilities.

Concluding Comments

In this chapter, various treatment options were presented and it was argued that behavioural interventions are a more preferable option to pharmacological intervention, as they have shown to produce both short-term and long-term remediation of sleep problems. Side effects associated with medications may be less appealing as well as unsuitable for many children and their parents. Additionally, the long-term use and the safety of sedative medications are controversial, and so behavioural strategies are presented as a more appropriate and safe option. Various behavioural intervention approaches have been reviewed and shown to be successful in reducing sleep problems in infants and children. Strategies such as extinction and graduated extinction are well validated. Other behavioural interventions such as positive routines, stimulus control, and bedtime fading, have demonstrated positive treatment outcomes, however further research is required to assess their efficacy.

Studies evaluating behavioural interventions in children with disabilities have produced promising results but there have only been a few controlled studies. Studies employed in children with disabilities are limited not only in number, but also by methodological shortcomings, such as small sample size and lack of experimental control. Another limitation in disability research is the heterogeneous nature of the samples. There is a need to consider disability subgroups separately (e.g. autism, Down syndrome, cerebral palsy), as intervention outcomes may differ between different disabilities (Richdale et al., 2000; Wiggs & France, 2000). Suitability of interventions may be determined by parent and child characteristics associated with various types of disabilities (Weiskop, 2001). Hence, there is a need for future research to address disability subgroups separately.
Additionally, research on behavioural interventions in children with disabilities has predominantly focused on intellectual disability and developmental disabilities. There is very little research on physical disabilities and chronic illness with the exception of two studies. Bartlet and Beaumont (1998) investigated the use of an intervention employing a variety of behavioural strategies with children with disabilities as well as children with chronic illness, such as asthma, eczema, chronic respiratory and ear infections and problems, epilepsy, and coeliac disease. Didden et al. (1999) assessed the use of extinction in a case study of a 2½ -year-old boy with a physical disability and near-normal intellectual ability. Further investigations would be valuable to assess the efficacy of behavioural treatments within these neglected populations.

Despite these methodological constraints, there is sufficient evidence to support the use of behavioural interventions for children with disabilities in clinical practice. There is a wide range of behavioural approaches that have been shown to improve sleep problems in children. Some researchers have employed a combined approach, incorporating various behavioural techniques and reinforcement procedures such as praise or a start chart (e.g. Seymour, 1987). France et al. (1996) also outlines ways of modifying behavioural treatments for older children, by incorporating rewards and praise. There are many behavioural techniques available, each with their advantages and disadvantages. Practicality of behavioural options should be assessed for each family, as their ability to implement strategies will vary. For children with disabilities, modified and gradual interventions addressing the child’s special needs and parental concerns may be more suitable (Durand et al., 1996; Weiskop et al., 2001). A comprehensive assessment of the child and family circumstances and analysis of the presenting problems is important in determining the most appropriate intervention approach. France and Hudson (1993) advocate a “consumer model” whereby the therapist presents various approaches that are suitable and allows the parents to make the final decision. By informing the parent of the advantages and disadvantages of each treatment option, the parent is likely to choose the most appropriate intervention for their circumstances. This may increase parental compliance to the programme and therefore increase the likelihood of a successful treatment result.

RATIONALE FOR THE CURRENT STUDY

A review of literature has shown that children with disabilities are not only at greater risk of developing a sleep problem they are also more susceptible to developing persistent sleep problems. This is concerning due to the body of evidence indicating the negative impact of sleep problems on individuals as well as on family members. Sleep problems would have more repercussions for children with disabilities whose development and learning is already
compromised by their disability, as well as for family members who are placed under considerable stress and pressure of having a child with a disability. This highlights the need for interventions with sleep problems in this population.

Social learning theory indicates that parents play an important part in shaping and maintaining sleep problems such as bed refusal and night waking by inappropriate stimulus control and reinforcement (Blampied & France, 1993). Behavioural interventions are strongly founded on this premise (Adair et al., 1991) and involve the manipulation of these parental factors. In older and verbal children, reinforcement techniques can be helpful to encourage the pursuit of a common goal. A thorough assessment is necessary to determine the possible cause and maintaining factors of the child’s sleep problem. This is an important precursor to the development of an appropriate intervention plan. This current study examines the use of behavioural family interventions to reduce sleep problems in four case studies of children with disabilities.
CHAPTER 2. GENERAL METHOD

PARTICIPANTS AND SETTING

This research received approval from the University of Canterbury Ethics Committee (see Appendix A) and was conducted as part of the Canterbury Sleep Programme (CSP) at the University of Canterbury, Christchurch, New Zealand. The CSP accepts referrals from local medical practitioners, child health nurses, and parents for research purposes. Four children from three families were recruited to the study and treated at no charge. Interviews, meetings, and interventions took place in the family home of the participants. All the families lived in Christchurch, New Zealand. Two families were referred (one referral was made by a medical practitioner, the other was from a local agency) to the CSP and one family was recruited by word-of-mouth. A further family was referred but declined to participate. The criteria for inclusion in this study were that:

1. The child was perceived as having a sleep problem by the parent(s), experiencing one or more of the following: a) bed refusal, b) sleep onset delay, c) night waking, d) co-sleeping, or e) early morning waking. Definitions are given on pages 5 and 6.
2. The child had a disability, whereby the parent(s) had to alter their parenting to cater for the child’s special needs. This did not include disabilities that were primarily a mental health problem.
3. The child was between the ages of 2 and 12 years.
4. The child had no apparent medical or physical problem that was determined to be the primary cause of the sleep disturbance.
5. The child had no emotional, medical, or physical problem which would contra-indicate behavioural treatment.

Descriptions of the children and their families are summarised in Table 1 (p. 42). Pseudonyms have been used to preserve the anonymity of the participants. Two girls (aged 4 years, 5 months and 6 years, 11 months) and two boys (aged 3 years, 3 months and 6 years, 3 months) and their families participated in this research. One child was from a single-parent family, while the other three children were from two-parent families. The parents were between the ages of 25 and 38 years. All of the children had one or more siblings. One participant was first born, two were second born, and one was third born. Three children were NZ-European (pakeha) and one child was of NZ-European and Maori descent. English was the first language for all the families. The children covered a range, from lower to upper socio-economic status families, as rated by the Elley-Irving Index (Elley & Irving, 2003) (see Table 1, p.42). All four
children had a disability that had been diagnosed by a medical professional (paediatrician or orthopaedist). The parents of all four children reported long-term difficulty with their children’s sleep since infancy and had reported high levels of stress associated with the sleep behaviour. None of the children were taking medication for their sleep disturbance at the time of recruitment into the study, however all four children had tried sedative medication in the past without success (see Table 2, p.43). Three children were currently taking medication for medical conditions. Jenny was prescribed Lamotrigine, an anti-epileptic medication, while Molly and Lewis occasionally used a Ventolin inhaler for asthma relief. The parents reported their children as having long-term significant sleep difficulties but otherwise being in good general health.

The children included in this study had varying disabilities. Full information for each child is given in the individual case studies. A summary of the children’s disabilities and sleep difficulties are presented in Table 2 (p. 43). All of the participants exhibited multiple sleep problems, including bed refusal, sleep onset delay, night waking, and co-sleeping. One child experienced nightmares, while two children displayed bed refusal and sleep onset delay.

<table>
<thead>
<tr>
<th>Child</th>
<th>Gender</th>
<th>Age (at initial interview)</th>
<th>Ethnicity</th>
<th>No. of Siblings</th>
<th>Birth Order</th>
<th>Parents Residing</th>
<th>Parent’s Age</th>
<th>SES*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Jenny</td>
<td>Female</td>
<td>6yrs, 11mths</td>
<td>NZ European</td>
<td>2</td>
<td>1</td>
<td>2</td>
<td>M = 38</td>
<td>F = 38 1</td>
</tr>
<tr>
<td>Molly</td>
<td>Female</td>
<td>4yrs, 6mths</td>
<td>NZ European/Maori</td>
<td>1</td>
<td>2</td>
<td>1</td>
<td>M = 25</td>
<td>7</td>
</tr>
<tr>
<td>Hamish</td>
<td>Male</td>
<td>6yrs, 3mths</td>
<td>NZ European</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>M = 33</td>
<td>F = 37 5</td>
</tr>
<tr>
<td>Lewis</td>
<td>Male</td>
<td>3yrs, 3mths</td>
<td>NZ European</td>
<td>2</td>
<td>3</td>
<td>2</td>
<td>M = 33</td>
<td>F = 37 5</td>
</tr>
</tbody>
</table>

* Socio-economic status as rated on the Elley-Irving Scale (Elley & Irving 2003) based on the occupation of the principal income earner, where 1=highest and 6=lowest, 7 was added to the scale to represent a parent who was receiving a domestic purposes benefit.
Table 2: Type of disability and sleep disturbance

<table>
<thead>
<tr>
<th>Child</th>
<th>Type of Disability / Medical Condition</th>
<th>Current Medication</th>
<th>Type of Sleep Disturbance</th>
<th>Onset of Sleep Disturbance</th>
<th>Previous Sedative Use</th>
</tr>
</thead>
<tbody>
<tr>
<td>Jenny</td>
<td>Cerebral palsy</td>
<td>Lamotrigine</td>
<td>Night waking Co-sleeping</td>
<td>Infancy</td>
<td>Promethazine</td>
</tr>
<tr>
<td></td>
<td>Global developmental delay</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Epilepsy</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Molly</td>
<td>Global developmental delay</td>
<td>Ventolin</td>
<td>Bed refusal Sleep onset delay Night waking Co-sleeping</td>
<td>Infancy</td>
<td>Promethazine</td>
</tr>
<tr>
<td></td>
<td>Asthma</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hamish</td>
<td>Multiple epiphyseal dysplasia</td>
<td>None</td>
<td>Bed refusal Night waking Co-sleeping Nightmares</td>
<td>Infancy</td>
<td>Promethazine Trimeprazine</td>
</tr>
<tr>
<td></td>
<td>Asthma</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lewis</td>
<td>Multiple epiphyseal dysplasia</td>
<td>Ventolin</td>
<td>Bed refusal Sleep onset delay Night waking Co-sleeping</td>
<td>Infancy</td>
<td>Promethazine</td>
</tr>
<tr>
<td></td>
<td>Asthma</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

MEASURES

Descriptive Measures

An interview schedule (see Appendix B) adapted from the Canterbury Sleep Programme and from Sattler (1998) was conducted in the family home with the parents, and the child and his or her siblings if appropriate. Information gathered included, demographic information, family composition, the child’s sleep history and current sleep problem(s), developmental history, medical history and current health, as well as any previous or current medications used by the child. During the home visit, informal observations were made of the child, family situation, and the bedroom layout. The information gathered was used in the assessment of the child’s sleep problem.

Socio-Economic Status (SES)

The SES of the parents was determined using the Elley-Irving Socio-Economic Index: 2001 Census Revision (Elley & Irving, 2003). Earlier versions of the Index have been widely used by researchers. The current version of the scale was developed based on statistics from the 2001 Census of New Zealand (Elley & Irving, 2003). The Index ranks 630 occupations on a 1 to 6 scale (1= highest SES, 6= lowest SES) based on the average income and educational level for each occupation. The Index however, does not have a category for parents who are on a welfare
benefit, work part-time, or those who are full-time caregivers. In this study, one parent received a Domestic Purposes Benefit (DPB). To represent this parent, “7” was added to the scale.

**Sleep Measure**

The sleep diary (see Appendix C), a parental report form, was designed to keep a daily record of the child’s behaviour and parental behaviour at night-time. The sleep diary was used as the main assessment instrument, and was adapted from a diary used by the Canterbury Sleep Programme (France 1989). The Canterbury Sleep Programme diary was designed for infant sleep and so adaptations were primarily to make it suitable for older children in this study. Several sleep diary formats were considered (Durand 1998; Ferber 1995a; Weiskop, 2001) however, the Canterbury Sleep Programme Diary layout had the benefit of being able to record multiple waking during the night, as well as information about the child’s behaviour and the parents’ response. Parents were asked to record the time the child was settled into bed, the time the child fell asleep, the number of night wakings, and the time the child woke for the day. The parents were asked to note the child’s behaviour at each of these times and how they responded to the child. Daytime naps were also recorded on the sheet if applicable.

**Reliability**

The use of an infrared-time-lapse video was considered as a method to collect reliability data for all of the participants. This was a standard CSP procedure in which video recordings of the child’s sleep was compared with parental sleep diary records to calculate reliability. Although this was the original intention, in each case there were practical issues that obstructed the use of the video. The main problem was due to the age and mobility of the children in this study. The video is a common procedure used with infants, however all four participants were older, and in two cases damage to the equipment and the potential to cause injury to the child were factors that eliminated this as a feasible option to obtain reliability. A video camera placed on a tripod to record the child’s behaviour at night may have been potentially dangerous due to the child pushing or playing with the equipment. Mounting a video camera on the wall out of the child’s reach may have been a way to overcome these difficulties, however, this option would have caused damage to the walls and would be too costly to implement.

The novelty of the video equipment may also be distracting for the children and actually disrupt sleep, and so because of potential “first-night” effects, the infrared time-lapse video camera was not used. Several other options to obtain reliability were considered:
1. Another adult to record a sleep diary
The other parent (in two-parent families), a relative, or family friend to record an independent sleep diary for 3 nights during intervention and follow-up was considered to gain reliability measures. In two-parent families, both parents were asked to record sleep diaries for 3 nights during intervention and 3 nights during the follow-up period for reliability measures. In Case Study One, there was difficulty in getting the husband to complete reliability records, thus other methods were considered. This family also had limited family support thus another adult to complete reliability records would have been too difficult to implement practically. In the single-parent family (Case Study Two), this method was not employed, as it would be too demanding for a family to implement practically, as it would require an adult to stay at the house for 3 nights during intervention and also at follow-up. The family in this study had limited support from relatives and friends, so this was not a viable option.

2. A video camera outside the child's bedroom
A video camera placed outside the bedroom to record any waking sounds from the child and to record parental attending was another method considered to gain reliability measures. This option was finally deemed too intrusive for the families. These families were under extreme stress already and it was a further request of them that in discussion with my supervisor we were not prepared to make. Therefore, it was not an appropriate method to gain reliability.

Phone Calls
Sleep diary records (completed by the parents) were compared with notes made by the researcher during telephone contact as a further reliability check.

Calculating Reliability
Reliability was assessed by calculating the percentage of consistency between the mother's sleep diary and notes taken by the researcher during phone calls (for all the case studies), and in some cases (Case Studies One, Three, and Four) the percentage of consistency between the mother's sleep diary and the independent sleep diary records of the father was also calculated. A 15-minute margin for error was allowed between records.

Reliability (%) = \frac{\text{the number of consistent time records}}{\text{the total number of time records}} \times 100
Social Validity Measure

A Programme Evaluation Questionnaire (see Appendix D) developed by the CSP was used to determine the social validity of the sleep programme. Parents completed the questionnaire at the conclusion of the intervention. Parents were asked to rate (on a 4-point scale) the quality of help they received, general satisfaction with the programme, the amount of stress experienced from implementing the programme, as well as whether they would recommend this programme to a friend.

EXPERIMENTAL DESIGN

Originally, a multiple baseline design was considered to examine the effects of the intervention. It was planned that participants would be randomly assigned to 7-day, 10-day and 12-day baseline conditions however, it was not practical or ethical to implement this because of varying family circumstances. Nonetheless, some children had had baselines randomly allocated in the initial planning phase. In Jenny’s situation (Case Study One), the parents expressed extreme urgency to begin the intervention immediately because of the stress the sleep problem was placing on the marital relationship and family functioning. The mother had been seeking services for her child’s sleep for many years and desperately wanted to implement an intervention straight away. After discussion with my supervisor, the decision was made to utilise a 7-day sleep diary the mother had completed 3 months prior to the study (that had been requested by a GP as part of an assessment of Jenny’s sleep), as it seemed unethical to enforce the baseline condition in this situation. The mother indicated that this diary was a good representation of Jenny’s current sleep patterns. Information gathered from the assessment interview about Jenny’s current sleep behaviours were consistent with the mother’s sleep diary and this contributed to the decision to use this record to replace the baseline condition.

Furthermore, because of varying child and family characteristics and circumstances as well as the different sleep problems presenting in each child, interventions implemented were notably different. This restricted the applicability of a multiple baseline between-subjects design. A multiple baseline approach was considered with the two siblings, however it was anticipated that parental interactions would be altered with both children despite starting an intervention with one child. Furthermore, although the two boys had the same disability, two separate interventions were required to treat different sleep problems. It was therefore inappropriate to use a multiple baseline design for the two boys.

Due to these unforeseen practical issues, it was not possible to adhere to the multiple baseline design and the decision was made to consider each family as a separate case study. Furthermore, a case study presentation was more appropriate as each child had a different
disability, the interventions were different, and the variability of family circumstances would have made it difficult for formal comparison. Thus, the experimental design used was a single-case design.

PROCEDURE

Participants were initially contacted by telephone to establish suitability for the study as well as to determine the parents’ interest in participating in the research. Extensive background information and sleep history were obtained during an assessment interview in the homes of the participants. This information was used to determine whether the child met the inclusion criteria for the programme. The aims of the study were explained to parents and an information sheet outlining the study (see Appendix E) was given to the parents and written consent was obtained (see Appendix F).

Baseline

Parents were asked to complete sleep diaries for the child after the assessment interview for an assigned number of nights (range of 7 nights to 12 nights), except in Case Study One (see experimental design). At the end of the baseline phase, the sleep diaries were collected.

Analysis of Presenting Problems

An analysis was completed based on information gathered from the assessment interview, home visit, and baseline sleep data. This information was used to develop hypotheses about how the sleep problems were being maintained in each child. If the assessment suggested a behavioural cause to the child’s sleep difficulty as opposed to a medical or physical cause, the programme was continued and an appropriate family behavioural intervention plan was developed.

Intervention

Target behaviours were identified for each child. Interventions were designed to meet the needs of the family, and the child’s disability and learning needs were given special consideration. The intervention plan was discussed with the parents during a home visit, and an information sheet outlining the strategies was given. The children received treatment for a variety of sleep problems, such as bed refusal, sleep onset delay, night waking, co-sleeping and nightmares (see Table 2, p. 43). The interventions used in this research are described in the separate case studies. Interventions were introduced after baseline and families were given daily telephone advice and support during this phase. Parents were also advised to contact the
researcher at any time of the day or night if they were experiencing difficulties with their child’s sleep or implementing the programme. Intermittent home visits were organised according to each family’s needs. The parents were advised to stop the sleep programme during times of illness, or if they felt that the child’s health or safety was compromised.

Discontinuation of the intervention phase was jointly determined by the researcher and the parent(s), when sleep problems were reduced to a satisfactory level and when parents no longer felt a need for support and advice. The duration of intervention phase was 6 weeks and 5 days for Molly, 17 weeks for Jenny, 20 weeks and 1 day for Hamish, and 22 weeks for Lewis.

Maintenance

After the intervention phase, a home visit was organised and the parents were advised of the maintenance programme chosen for their child. The parents were advised that they may need to return briefly to the programme if there was a disruption to the child’s sleeping, such as in the case of illness. Daily telephone contact was discontinued with the understanding that parents were more than welcome to contact the researcher at any time for advice or support.

Follow-up

A follow up was completed 2 ½ to 3 months after the intervention phase and parents were asked to record a sleep diary for 2 weeks. In Case Studies Three and Four, follow-ups were conducted after 2 ½ months to avoid the Christmas period, as it is a particularly disrupted and busy time for families. Follow-up data was used to determine the long-term effects of the intervention on the child’s sleeping and whether changes in sleep behaviour and habits were maintained.

Data Coding

The sleep diary data were coded and graphed. The following variables were evaluated for all participants: Night wakings: was defined as any waking the parents were aware of, throughout the night before the scheduled wake time. For all the participants 6am was deemed an appropriate wake time and any waking prior to this time was determined to be a night waking. The frequency of night waking per night was recorded. Co-sleeping: was defined as the child sleeping with one or both parents, in the parents’ bed or the child’s bed during the night. Child illness: Nights when the child was ill were recorded as illness can significantly affect sleep. Parental non-compliance: was defined as parental actions that directly opposed recommendations given. This was documented as it could influence the progress of the intervention and potentially affect child sleep.
For Molly and Lewis a fourth variable was coded, namely sleep onset delay. This was the number of minutes between first being settled to bed by the parents and sleep onset. This may include settling difficulty whereby the child exhibited behaviour competing with sleep such as crying at bedtime. Another variable, Nightmares was only applicable to Hamish. A nightmare was defined as a bad dream that disrupted sleep, resulting in child distress to the degree that parental attention and comfort were sought.

Data Analysis

For every target behaviour visual analysis of the graphs was conducted to assess baseline versus the last 2 weeks of the intervention phase and baseline versus the follow-up phase to evaluate treatment effects. A 5-point rating scale modified by Weiskop (2001) from a rating scale developed by Hudson, Wilken, and Jauernig (1995) was utilised to make comparisons between these conditions. Ratings were: substantial improvement, moderate improvement, no change, moderate deterioration, or substantial deterioration (see Appendix G for criteria). Graphs were co-rated by a postgraduate Child and Family Psychology student for reliability of the visual analysis. The co-rater was given the indication of the desired direction of change, namely a decrease in undesirable behaviours. Child illness was taken into consideration in the analysis by the co-raters in the analysis.

Furthermore, graphs were analysed in accordance with guidelines stipulated by Cooper, Heron, and Heward (1987). Visual analysis of data within phases/conditions was assessed on three criteria: i) the variability of the behaviour, described as either stable or variable, ii) the level of behaviour (the value on the vertical axis on which a set of behavioural measures converge). The terms low, moderate, or high was used to describe the level of performance, and iii) the direction of any trends. The direction of trends was described as increasing, decreasing, or having a zero trend. Visual analysis between phases/conditions involved describing the change or lack of change in the level and trend between experimental conditions (i.e. baseline, intervention, and follow-up conditions).
CHAPTER 3. CASE STUDY ONE: JENNY

Jenny was 6 years and 11 months old. She has right-sided spastic hemiplegia cerebral palsy, epilepsy, and global developmental delay. She was delivered overdue at 43 weeks gestation. Jenny has severe intellectual disability, epilepsy, and limited language ability. Jenny had myoclonic seizures until the age of 5 to 6 years, and currently had complex partial seizures. Jenny lived with her mother, father, younger brother (aged 4 years), and sister (aged 23 months). The family’s socio-economic status was rated 1 on the Elley-Irving Scale (Elley & Irving, 2003).

Jenny was referred to the CSP with a primary sleep problem. Jenny had persistent night waking requiring constant parental attention. Her parents reported that she woke on average 5 times a night, which frequently resulted in their having to soothe her to sleep by sitting in a chair in her bedroom or by co-sleeping. Jenny’s parents indicated that her sleep difficulty was causing a lot of stress and disrupting their own sleep patterns. Her parents could recall only a few occasions when Jenny had slept throughout the night, for example on a family camping trip where the family shared a tent together. Jenny’s parents had made previous attempts to manage her sleep. At 4 years of age they tried a portable cot (on the advice of the family mental health services they were referred to). This stopped night waking for a period of time until she outgrew the portable cot and was transferred to an ordinary bed. Her parents have also tried on two separate occasions to establish an extinction procedure but said they gave up after 3 or 4 nights due to the stress involved with the procedure. At age 4 years, they closed the door and ignored Jenny’s crying and screaming. A parent attended to her at around 2am each night to put her back into bed, and then they continued to ignore her until the morning. This method was stopped after 3 to 4 nights. At age 6 years, the parents sat in a chair for 10 minutes until Jenny was asleep and they left the room, then they ignored her until the morning. After 3 nights they discontinued this strategy. Sedative medication (promethazine) has been tried in the past without success. The parents commented that the medication caused Jenny to be extremely drowsy during the day.

Jenny’s family was under a considerable amount of stress and there were disagreements between Jenny’s mother and father regarding her management. During the assessment interview the mother described the father as being “fed up” with the situation and advised me that he would not be participating in the sleep intervention. Jenny’s mother also stated that her husband’s desperation had led him to consider seeking permanent foster care for her.

CEREBRAL PALSY, EPILEPSY, GLOBAL DEVELOPMENTAL DELAY, AND SLEEP

The incidence of cerebral palsy is approximately 1.2 to 2.4 per 1000 births. (Hagerg & Olow, 1993 as cited in Pellegrino, 1997; Irie, 1999 cited in Kotagal, 2001). According to
Pellegrino (1997) there are many causes of cerebral palsy such as birth trauma, prematurity, and adverse intrauterine development. There are also different forms of cerebral palsy, which are classified according to the type of impairment and regions of brain that are affected. For Jenny, right-sided spastic hemiplegia implicates damage or dysfunction in the left side of the brain, as the motor neurons that control one side of the body are located in the opposite cerebral cortex (Pellegrino, 1997).

Cerebral palsy is commonly associated with other disabilities such as intellectual disability, visual impairments, hearing impairments, speech language disorders, feeding difficulties, growth abnormalities, and behavioural and emotional disorders (Pellegrino, 1997). Children with cerebral palsy are also at greater risk of developing medical conditions, in particular seizure disorders, with approximately 50% of children with cerebral palsy affected by seizures (Aksu, 1990 as cited in Pellegrino, 1997). Children with cerebral palsy are also at risk of developing a sleep problem (Didden et al., 2002; Quine, 1992). Shibagaki, Kiyono, and Takeuchi (1985) found abnormal EEG sleep patterns in infants with cerebral palsy, such as an absence of EEG patterns characteristic of wakefulness and NREM and REM sleep stages without characteristic spindles (bursts of rapid activity). Cerebral palsy has also been associated with sleep-wake or circadian rhythm abnormalities (Palm, Blennow, & Wetterberg, 1997).

Behavioural difficulties and psychological disturbance have also been associated with cerebral palsy. These factors also are commonly associated with sleep disturbance (Goodman, 1998; Goodman & Graham, 1996).

Most of the research into sleep and cerebral palsy has not differentiated the various subtypes and so it is unclear whether sleep is impacted differently in these subgroups. One study by Kotagal, Gibbons, and Stith (1994) examined nine patients with spastic quadriplegia and severe psychomotor impairments and found more apnoea (periods of cessation of breathing often caused by airway obstruction or cerebral dysfunction) and hypopnoea (abnormally slow or shallow breathing) per hour of sleep compared to age-matched controls.

Another study (Hayashi, Inoue, Iwakawa, & Sasaki, 1990) investigated the EEG of patients with athetoid cerebral palsy (a form of cerebral palsy where there are tonal abnormalities affecting the whole body, characterised by rapid, random, jerky writhing movements). The authors reported that there was a marked difference in the sleep architecture of patients with athetoid cerebral palsy with a significant decrease in the amount of REM sleep, as well as a lower rate of gross body movements during REM sleep when compared with typical sleep architecture. There has been little research analysing sleep in other specific forms of cerebral palsy, and so it is difficult to determine the relevance of these findings for Jenny. Discomfort and pain associated with cerebral palsy and problems with turning, may also relate to sleep problems.
At present, however there is no evidence to indicate that other subtypes of cerebral palsy differentially affect sleep.

Epilepsy may also increase the likelihood of a child developing a sleep problem. Many studies have shown that a high proportion of children with epilepsy have sleep difficulties (e.g. Didden et al., 2002; Quine, 1992). Stores (2001b) and Montplaisir (1990) reviewed literature on sleep and epilepsy and reported that disturbances in sleep physiology have been found in various forms of epilepsy. Furthermore, research indicates that not only can seizures disrupt sleep in children, sleep loss or disruption can increase the frequency of seizure activity. It has been noted that epilepsy can cause excessive daytime sleepiness, however this may be due to the use of some antiepileptic medications, and so a careful assessment is necessary (Stores et al., 1995). A study by Stores, Wiggs, and Campling (1998) compared 78 children with epilepsy aged 5 to 16 years, with 73 matched healthy controls, and found that children with epilepsy had significantly greater sleep problems (poor quality sleep, anxiety about sleep, disturbances during sleep, symptoms of sleep disordered breathing, and short duration of sleep). The authors also found that sleep problems were associated with daytime behaviour problems.

The association between intellectual disability and sleep is well documented (e.g. Bartlett et al., 1985; Didden et al., 2002; Wiggs & Stores, 1996a). The reader is referred to the Case Study Two for more information on sleep and GDD.

METHOD

During the assessment interview, it became apparent that the mother wanted the intervention to start urgently. Due to the high amount of stress the sleep difficulty was causing the parents and their desperate urgency to begin, the decision was made in consultation with the research supervisor, to start the intervention that night and to utilise previously collected data as baseline. The mother had completed a personal diary of her child’s sleeping for 7 nights, 3 months prior to involvement in this study (requested by a General Practitioner or GP, as part of an assessment of Jenny’s sleep) and this information was used as baseline data for Jenny. The mother advised that this sleep record was a good representation of her current sleep patterns. Furthermore, information gathered during the assessment interview confirmed that Jenny’s current sleep problems were consistent with the mother’s record. Jenny went to bed around 8pm every night, falling asleep within 10 to 30 minutes from initially being settled into bed. She woke on average 5 times a night and some nights waking every hour until her morning wake time between 6 and 7am. These night-time difficulties regularly resulted in co-sleeping.

Another meeting that day was scheduled to introduce the intervention strategies and sleep diaries.
Special Considerations for Intervention

Epilepsy

Although Jenny had epilepsy, which is known to interfere with sleep (Curtesy et al., 1999), a decision was made to include her in the study. As Jenny’s night waking could have been due to seizure activity, medical clearance from a GP was obtained prior to implementing a behavioural programme on the same day of starting the intervention. Jenny’s seizures were controlled with medication (Lemotrigine) and she had been seizure-free for 5 months prior to the study. Jenny had been able to sleep through the night during family camping trips (where the family shared a tent) and when co-sleeping, suggesting that her sleep difficulties were caused by behavioural factors and not her epilepsy.

Cognitive and Communicative Ability

Jenny had limited cognitive and communicative ability. Her receptive language allowed her to understand simple sentences, requests, and praise. Jenny was able to communicate some ideas such as “Jenny was stuck” but her language mainly consisted of babbling. Jenny’s cognitive and communicative abilities required special consideration when choosing an appropriate intervention. Intellectual disability meant that Jenny would require allowances for her ability to learn new things and more time. Strategies implemented would therefore need to be simplified, repetitious, and consistent. Additionally, good sleep behaviour needed to be reinforced as soon as possible.

Physical Disability

Jenny was able to walk slowly, but frequently bottom shuffled on the floor. She was learning to stand-up from sitting on the ground, but often required assistance. Jenny was able to get out of bed easily, but could not return to bed on her own. This meant that when Jenny got out of her bed at night she required parental assistance. Teaching Jenny to get into bed on her own was part of the programme.

Child Safety

Prior to implementing the intervention, Jenny’s room was prepared. This was to ensure the child’s safety throughout the programme. Jenny was prone to strew toys and clothing around the room during night awakenings, so unnecessary toys and furniture were removed. The wardrobe doors were also secured.
Analysis of Presenting Problems

Two target behaviours were identified for Jenny: night waking and co-sleeping. Jenny woke between 3 to 6 times every night. The data indicated that Jenny went to sleep with inappropriate stimulus control. Jenny required parental presence in order to fall asleep and required this stimulus to return to sleep when she woke during the night. This sleep onset conditioning has been present since Jenny was an infant when she would often fall asleep during breastfeeding. Jenny was described as a very unsettled and difficult baby who frequently required comforting. Currently at bedtime a parent would often stay in her room until Jenny fell asleep. The sleep onset conditioning of parental presence at bedtime was also targeted for intervention. Jenny’s mother or father was always present on awakenings to sooth Jenny back to sleep. Aversive behaviours during night waking exhibited by the child such as crying were reinforced by parental attention. The parents would often resort to co-sleeping to enable Jenny to sleep for the rest of the night. This was also reinforcing for the parents who were able to sleep undisturbed. It was determined after consultation with the parents that co-sleeping was not a normal part of the family’s cultural practice. It had been instituted as a response to the night awakening because the parents were concerned that Jenny was not able to sleep because of her disability. In summary, the analysis suggested that Jenny’s sleep problems were being maintained by parental attention and inappropriate stimulus control.

Intervention

An information sheet (see Appendix H) on the parental presence programme was given to the mother. Daily telephone contact (ranging from 5 minutes to 60 minutes duration) was made in the morning throughout the intervention phase to give advice and support to the mother. After 10 ½ weeks telephone contact was reduced to every second day. The intervention phase was 17 weeks in total. A follow-up was conducted 3 months after the intervention had been completed.

Positive Routines and Interactions

A positive bedtime routine was arranged with Jenny’s mother, and a regular bedtime and wake time were negotiated. Jenny’s bedtime was set at 8pm and her wake time at 6am. During the interview the mother had reported behavioural difficulties with Jenny and indicated that interactions with Jenny were often negative during the daytime. So an emphasis on positive time between the mother and child was an essential part of the programme. The mother was encouraged to include quiet-time in the bedtime routine, where she would spend some time with Jenny doing something relaxing and enjoyable, such as cuddling, reading a story, or a quiet game. Positive interactions during the daytime were also emphasised as the parental attention that
Jenny received during the night was to be eliminated. Jenny was settled into bed by the mother and was left to fall asleep on her own.

Reward System

Throughout the intervention phase, Jenny was reminded every night that she was to stay in her own bed until the morning and that she would receive a stamp if she did so. Jenny responded well to stamps on her hand and so they were incorporated into the sleep programme with the aim of reinforcing sleeping though. A star chart was also planned as reinforcement for her sleeping. A reward box was introduced (a box full of toys or stickers for Jenny to choose from).

Parental Presence

The behavioural intervention used was similar to the “parental presence” programme utilised by France and Blampied (2002). The rationale for using the Parental Presence method was to decrease child and parent anxiety while adhering to planned ignoring strategies (France, 1994), as well as to model appropriate sleep behaviour. The parental presence programme would not normally be used with mobile children but because Jenny had slept through under similar circumstances on a family camping trip it was decided to use it as the first intervention step. The mother was asked to sleep in Jenny’s room on a mattress beside her bed. The mother modelled appropriate bedtime behaviour (i.e. pretending to sleep) and was asked to ignore Jenny by lying in the bed feigning sleep.

Trimiprazine Tartrate

The parents were given the option of using the Parental Presence programme with or without the use of sedative medication. Jenny’s parents decided to use a decremental dose of trimiprazine tartrate (a mild sedative) in combination with behavioural techniques. The aim of this was to decrease the PERB and to minimise the stress on the child and family in implementing behaviour change (France et al. 1991). France et al. (1991) demonstrated that extinction in combination with medication was effective in reducing the PERB, in that treated children cried less. A letter was written to the family GP explaining the sleep programme and asking him to prescribe the medication (see Appendix I).

Implementation of the Programme

Issues of implementation and modifications to the programme are presented below.
Reward system

The reward box was implemented but eventually discontinued by the parents, as they felt that Jenny responded more positively to receiving a stamp on her hand.

Trimeprazine Tartrate

On the first night Jenny was given 5mls of Trimeprazine Tartrate solution (30mgs of trimeprazine tartrate per 5mls solution). This was increased to 7.5mls on the second night (on the advice of the GP) as the parents reported that there was no difference in her night-time behaviour. The higher dose had a sedative effect on Jenny. A decision was ultimately made to maintain this dose of sedative medication until there was a substantial improvement in Jenny’s sleeping. This was to ensure that Jenny’s learning was consolidated before sedative withdrawal. The dosage of trimeprazine tartrate was decreased on the 32nd night and completely withdrawn by the 42nd night. The withdrawal of medication was done gradually, reducing the dose by one-fifth every second night until there was no medication after 11 nights after commencing withdrawal.

Parental Presence

The parental presence programme was utilised for 6 nights. On the 4th night, Jenny’s night waking was reduced from 4 night wakings to 2 night wakings per night. Despite these improvements there were some practical difficulties with the use of the parental presence strategy. Jenny got out of bed and frequently attempted to sleep with her mother on the mattress. The mother was advised to assist Jenny back to her own bed with minimal interaction.

The parents expressed concern that the intervention was not working quickly enough, and so on the 7th night the parental presence approach was modified so that when Jenny woke, the mother was asked to remove herself from the bedroom for increasing amounts of time per waking. For example, on the first waking the mother went out of the room for 5 minutes, then on the second waking 10 minutes, and so forth. The maximum amount of time out of the room was set at 25 minutes. This graduated withdrawal method was utilised as a form of contingency for the night waking.

Standard Extinction

On the 18th night of the modified parental presence strategy, there was a marked disagreement between Jenny’s parents about the programme. A home-visit with the research supervisor, the parents, and the researcher was held. Both parents renewed their commitment to the programme and agreed to keep disagreements out of Jenny’s bedroom. Treatment options were discussed and the advantages and disadvantages of standard extinction and graduated
extinction were presented to the parents. A standard extinction protocol was decided upon because it would be more rapid and less confusing for Jenny.

The parents were requested to ignore all night wakings until the morning wake time. Jenny’s door was secured. Because of concern about Jenny’s safety it was agreed that Jenny’s mother would check on her through an outside window. Standard extinction was implemented for 7 nights but Jenny’s inability to return to bed on her own, necessitated a change in the standard extinction procedure. On the 20th night, the mother was advised to return Jenny to bed once quiet after a night waking. This minimal interaction was to ensure that Jenny did not have to sleep on the floor.

From the 66th night, Jenny started to have bowel motions during the night when she woke which was another concern for the mother who started to check Jenny’s nappy while she was asleep. She also began attending to Jenny 20 minutes after a night waking instead of waiting until she was quiet. This unscheduled checking decreased the progress of the intervention. Larger sized nappies that could not leak were bought. This reassured the mother and the programme was resumed.

For the rest of the programme the mother was instructed to ignore night wakings until Jenny was quiet, and if the wakings were close to the normal wake time (less than 1 hour) to ignore her until the scheduled wake time (usually 6.50am) and let her out of her room for the day. After 10 ½ weeks of intervention, telephone contact was reduced to every second morning. The intervention was terminated after 17 weeks, after consultation with the parents. The parents were satisfied with Jenny’s progress and felt they no longer required the support.

Teaching Jenny to Get into Bed

Teaching Jenny to get into bed was also set as a goal. On the 29th day, an occupational therapist was consulted and the bed was lowered (so that Jenny could comfortably sit on the edge) and the appropriate steps for Jenny to get into her bed were ascertained. The mother helped Jenny practice the steps required to get into bed. These were sitting on the edge of the bed; swinging her legs onto the bed; lying down; and pulling the sheets over herself. Jenny’s mother incorporated this practice into Jenny’s bedtime routine and gradually reduced her help. During the daytime, practice was in the form of play (bouncing, and swinging legs onto the bed). During the course of the intervention, the mother reported that Jenny began completing some steps on her own, such as pulling the sheets up, and lying down, and bringing her legs up onto the bed without assistance. On the 50th night, the mother reported that at bedtime Jenny sat on the edge of the bed, pulled up her legs onto the bed and then sat cross-legged in the middle of the bed. On
the 55th and 58th nights, Jenny pulled up the covers by herself. On the 76th, 79th, 81st, 84th, and 86th nights Jenny had returned to bed on her own to be covered once she was asleep.

**Maintenance**

At 17 weeks, although the parents had not reached their goal of Jenny’s sleeping through the night, they felt confident to continue with the programme independently. The parents were satisfied with their progress and no longer required the regular support. Regular telephone contact was stopped, however the parents were advised that they were able to contact the researcher at any time for further advice or support.

**Follow-up**

A follow-up was completed 3 months after the programme. The mother completed 2 weeks of sleep diaries. Three further nights of reliability data were collected 2 weeks after the follow-up phase. This was because Jenny’s father had failed to collect follow-up reliability when initially asked.

**Data Coding**

The following variables were coded for Jenny: i) night waking, ii) co-sleeping, iii) child illness, and iv) parental non-adherence. These have been coded and graphed. Sleep onset delay was also coded and graphed to demonstrate the change in Jenny’s sleep onset conditioning with the reduction of parental involvement at bedtime during intervention.

**Reliability**

The reliability between the mother’s sleep diary and the researcher’s notes taken during phone calls was 99%. The father completed 3 nights of reliability data during the intervention phase but failed to complete reliability recordings during follow-up as agreed. The parents however completed 3 nights of sleep diary for reliability at a later date (2 weeks after follow-up) and this was utilised. Reliability calculated from the mother’s sleep diary and the father’s independent sleep diary was low. The overall reliability for the mother and father’s sleep diaries was 69% (57% during intervention and 83% at follow-up).

The reliability between co-raters in the visual analysis of Jenny’s graph was 100%.
RESULTS

**Figure 1:** Sleep onset delay in minutes, across conditions: Jenny

**Figure 2:** Frequency of night waking per night, across conditions: Jenny
Sleep Onset Delay

Figure 1 presents sleep onset delay in minutes, across conditions. During baseline Jenny’s parents would sit in a chair in her bedroom until she was asleep. Sleep onset delay during baseline was within 5 to 10 minutes. With the parental presence programme, there was an initial increase in sleep onset delay for Jenny (20 to 25 minutes) however for the majority of nights Jenny fell asleep within 5 to 10 minutes from being settled to bed. When the mother was removed from the bedroom and the standard extinction strategy was implemented (18th night) there was a sudden increase in sleep onset delay to 45 minutes and 38 minutes on the subsequent night, followed by a rapid decrease to a low stable rate of sleep onset within 5 minutes for the rest of the intervention and follow-up phases.

Night Waking

Figure 2 presents the frequency of night wakings per night, across conditions. Baseline measures taken for a total of 7 nights indicated that Jenny was waking frequently every night. The number of night wakings during baseline was high but also variable, between 3 to 6 times per night (average of 3.9). During the initial stage of the parental presence programme night waking was at a stable level of 2 to 3 night wakings per night. With the change to modified parental presence, there was a sudden increase of night wakings to 6 per night. There was an average of 4 wakings per night during the parental presence phase. The implementation of the standard extinction procedure combined with trimeprazine, also resulted in an initial increase of night waking (up to 7 night wakings), followed by a rapid decrease in night waking. On the 24th night, Jenny slept through the night for the first time without waking. For the next 20 days or so, Jenny had a low but moderately variable rate of night wakings (range of 0 to 2 night wakings) during which she achieved many nights of undisturbed sleep. Jenny woke on average 1.1 times a night during the standard extinction with trimeprazine phase. During the unmodified extinction phase, overall there was a lower rate of night waking for Jenny compared to baseline with most nights having only one waking. There were two periods of marked night wakings for Jenny during this phase. The first occurring from the 57th to 64th nights, and the second occurring from the 70th to 76th nights. From the 83rd night until the end of the intervention phase (132nd night), there was a very low stable rate of night waking for Jenny with most nights having only 1 waking. Overall, during the unmodified extinction phase Jenny woke between 0 and 4 times a night (average of 1.0)

The 3-month follow-up also found a low stable rate of waking. When analysing Jenny’s follow-up data, 3 out of 14 nights were undisturbed and 11 out of 14 nights Jenny experienced
only 1 night waking (average of 0.8). An additional 3 nights of reliability data showed no night wakings. The overall trend of night wakings was decreasing.

Co-sleeping

Figure 2 presents the nights where co-sleeping occurred. Night waking behaviour would regularly result in co-sleeping, with Jenny being allowed to sleep in the parents' bed, or one of the parents sleeping with Jenny in her bed. Co-sleeping was recorded 5 out of 7 nights (71%) during baseline. Immediately with the intervention, co-sleeping was eliminated, with no incident of co-sleeping during intervention or follow-up (0%).

Child Illness and Parental Non-Compliance

There were two periods of child illness occurring from the 61st to 64th nights and 104th to 110th nights (Figures 1 & 2). Three incidents of parental non-compliance were recorded, on the 18th, 42nd, and 70th nights (Figure 2).

Visual Analysis

Night Wakings

The frequency of night waking during baseline and the last 2 weeks of intervention were compared. The change in the frequency of night waking was rated as showing a substantial improvement. Baseline and follow-up night waking data were also compared and rated as showing a substantial improvement by the co-raters.

Co-Sleeping

The frequency of co-sleeping during baseline and the last 2 weeks of intervention were compared and rated as showing a substantial improvement. Comparisons between baseline and follow-up co-sleeping were also rated as showing a substantial improvement.

Social Validity Measures

The programme had high social validity. Both parents reported high levels of satisfaction with the programme and commented that the programme procedures were easy to follow and logical. They indicated however that implementing the programme was very stressful. Both parents reported that the amount of telephone contact was beneficial and the mother advised that this constant support helped her get through the programme.
DISCUSSION

Overall, there was a substantial improvement in Jenny’s sleep difficulties supporting the use of behavioural intervention strategies with a child with cerebral palsy, epilepsy and GDD. Jenny was settling quickly (within 10 minutes) during baseline however, the parents were always present until Jenny fell asleep. During the parental presence programme Jenny had a slightly variable rate of sleep onset, but once the mother was removed from the room to start the standard extinction programme, Jenny’s sleep onset delay increased suddenly to 45 minutes, indicating a PERB. This was followed by a rapid decrease in sleep onset delay to a low stable rate of 5 minutes, suggesting rapid sleep onset for Jenny once she was placed in bed. The programme was successful in reducing parental interactions and changing the sleep onset conditions for Jenny at bedtime.

Jenny’s baseline data indicated that she was waking between 3 to 6 times per night, commonly resulting in the parents attending to the night wakings and co-sleeping. The number of night wakings decreased over the course of the intervention indicating a positive treatment effect. It is important to note that towards the end of the intervention there were several nights where night wakings occurred close to the scheduled wake time between 5am and 6am. This suggests that Jenny’s wakings were shifting to a later time and that she was almost sleeping through the night.

Improvements in night wakings were maintained at follow-up. During the follow-up phase Jenny woke only once a night. Although it is not illustrated in the graph, there were further improvements in Jenny’s night waking behaviour. Follow-up data indicated that Jenny was waking at later times, closer to the scheduled wake time, with most wakings (10 out of 14 nights) occurring between 5.30 and 5.55am. No night wakings were recorded in the three extra nights of reliability data. This suggests that there were long-term treatment effects. Furthermore, the programme was successful in eliminating co-sleeping. This was also maintained at follow-up.

There was evidence of a PERB, during the modified parental presence and unmodified extinction phases, with an increased frequency of night wakings reported (6 to 7 night wakings per night). This was followed by a marked decrease in night wakings and from the 32nd night the dosage of trimeprazine tartrate was reduced gradually. There was a slight increase in night waking during this time, a possible rebound effect from sedative withdrawal.

Jenny’s sleep progress was disrupted by illness on two occasions. The first time of illness, Jenny’s night waking increased to 3 to 4 wakings per night. Jenny’s night waking during the second time of illness was not as marked, however, after illness Jenny began to wake at earlier times (between 4 and 5am). This indicates that child illness was disruptive to the programme.
Furthermore, there were three separate incidents of parental non-adherence during the intervention phase that impacted intervention progress.

The case study shows that an intervention employing a variety of behavioural techniques (a positive bedtime routine, parental presence, modified extinction, unmodified extinction, and a reward system) is effective in reducing persistent sleep problems in a child with a disability. The assessment process enabled the development of an intervention plan specifically tailored for Jenny and her family. Jenny’s case illustrates the importance of accommodating a child’s special needs into a treatment plan.

Attempts were made to minimise stress for Jenny and her family during intervention, such as using a modified extinction process with the use of trimeprazine, as well parental presence and graduated strategies. Despite these attempts, the parents indicated that the programme was very stressful. Implementing behavioural change in children with disabilities may be difficult, especially if the child has limited communicative ability. Jenny’s parents may have felt more anxious about making changes, as they were less able to explain to Jenny what they were doing. The change would also have been stressful for Jenny as she would be less able to understand it. Emphasis is placed on providing frequent support for the family during behavioural intervention. Both parents reported that regular telephone contact and support was important for the completion of the programme. Despite the high stress involved in implementing the strategies both parents reported being very satisfied with the programme.

Jenny’s sleep programme was effective in reducing problematic sleep behaviours, however it is important to note that the length of the programme was 17 weeks, suggesting that treatments in some cases may take longer for children with disabilities. Jenny’s ability to learn new behaviours and skills may have been affected by her intellectual and communicative level. The length of the intervention may have been prolonged because of several factors associated with Jenny’s disability. Families with children with disabilities are already under pressure, and implementing a behavioural programme is timely and labour intensive. The parents’ ability to implement an intervention may have been at times under strain. This may have underpinned the incidents of parental non-compliance in the programme. Child illness was also a factor that disrupted progress. Jenny was less able to communicate her needs and so her parents had to be more responsive to her. Toileting was also an issue. Jenny was not fully toilet trained and so bowel motions during the night interfered, at one stage, with the intervention progress as it limited the mother’s ability to ignore wakings. This emphasised the need not only to consider individual and family circumstances in the development of an intervention plan but also the need to adapt the programme in response to the family’s changing needs.
The treatment of sleep problems in children may have other associated benefits for the individual and family. It is well documented that children's sleep problems can affect family functioning and relationships (Chavin & Tinson, 1980; Stores, 1996; Quine, 1992), marital relationships and satisfaction (Chavin & Tinson, 1980), parental stress (Richman, 1981, Quine, 1991) as well as having detrimental effects on the individual, such as on behaviour (Lavigne et al., 1999; Zuckerman et al., 1987), cognitive and academic functioning (Meijer et al., 2000; Wolfson & Carskadon, 1998). The mother reported positive change in Jenny as a result of improvements in her sleep as early as the 40th day of the programme. She advised that Jenny’s schoolteachers had commented that Jenny had improved concentration at school and was learning new tasks quickly and Jenny’s mother had also noted improvements in Jenny’s concentration and behaviour at home. After the completion of the intervention, the mother reported that Jenny was talking more. Both parents reported improvements in their own sleep with the programme. Jenny’s mother indicated that her relationship with her husband was under less stress because of improvements in Jenny’s sleeping. Furthermore, Jenny’s father was no longer considering alternative care. Jenny’s parents attributed these improvements to the success of the sleep intervention. Although this information is subjective, it does however demonstrate the impact of sleep problems on families and the importance of intervention for children with disabilities.
CHAPTER 4. CASE STUDY TWO: MOLLY

Molly was a 4 years and 5 months old girl who lived with her mother and older brother (aged 6 years). At birth Molly had an Apgar score of 1 and her score after 5 minutes increased to 9 after resuscitation. Her developmental milestones such as rolling over, sitting and walking were delayed, and at age 2 years a paediatrician diagnosed Molly as having severe Global Developmental Delay (GDD) with no chromosomal abnormality. Molly is severely intellectually disabled and nonverbal. She had very limited receptive language, understanding only simple instructions or prompts in context, such as “Molly, get in the car.” At 7 months of age, Molly was hospitalised due to asthma and had been hospitalised several times over the years. Her mother advised that Molly was currently in good general health, and she had not been in hospital for the past year. She was currently using Ventolin for asthma relief and was not on any other medication. Socio-economic status was rated as 7 on an adapted version of the Elley-Irving scale (Elley & Irving, 2003). Molly and her family had very limited support from family and friends. Molly attended kindergarten in the morning 4 days a week.

Molly was referred to the CSP by a community agency for sleep onset difficulty and frequent night waking, approximately 3 to 4 times a night. The mother reported that Molly had had these sleep difficulties since infancy, and she continued to settle Molly with a bottle at bedtime and throughout the night when she woke. According to her mother, Molly would settle at night anytime between 7.30pm and 1am, often waking during the night for “a couple of hours” playing with her toys as well as opening the bedroom windows. Molly would regularly go to her mother during the night and sleep in her bed. Molly had been given sedative medication (promethazine hydrochloride) in the past with little success, and her mother reported that there was no difference in Molly’s sleeping with the medication. Molly’s sleep difficulty was causing a lot of stress for her mother who also reported tiredness due to the constant attending at night.

GLOBAL DEVELOPMENTAL DELAY AND SLEEP

Developmental delay is the term used when there is a failure to achieve age-appropriate milestones due to atypical neurodevelopment (Brown & Eksnin, 1994 as cited in Batshaw & Shapiro, 1997). Global developmental delay (GDD) is a generic term often used in New Zealand to describe delays or impairments across many areas such as cognition, speech and language, gross/fine motor skills, social and personal skills, and/or the person’s ability to complete activities of daily living due to developmental disabilities. Therefore, sleep literature on developmental disabilities (Durand et al., 1998, Didden et al., 2002), learning disabilities (Quine,
1991; Saxby & Morgan, 1983), and intellectual disability (Didden et al., 2002; Johnson, 1996; Richdale et al., 2000; Piazza et al., 1996) would be relevant. Crocker (1989) reported that the most common cause of intellectual disability requiring extensive support is chromosomal abnormalities (35%) such as Down syndrome and fragile-X syndrome. Other identified causes include multiple congenital anomalies, single-gene defect, early pregnancy problems, perinatal insults, and postnatal brain damage (as cited in Batshaw & Shapiro, 1997). In Molly’s case the cause was not identified. The prevalence of moderate to profound range of intellectual disability is between 0.8 and 1.2%. Intellectual disability occurs more frequently in boys than in girls, at a ratio of 2:1 (McLaren & Bryson, 1987 as cited in Batshaw & Shapiro, 1997).

The association between sleep and intellectual disability and developmental disabilities is well documented (Didden et al., 2002; Richdale et al., 2000; Quine, 1991, 1992; Wiggs & Stores, 1996a). As mentioned in the Chapter One there are several contributing factors implicated in the development of sleep problems for this population. Quine (1991) demonstrated that children’s communicative ability was strongly associated with the development of sleep problems, suggesting that children who were less able to express their own needs as well as less able to understand and learn appropriate sleep behaviours were more likely to develop sleep difficulties. Okawa and Sasaki (1987) suggested that children with intellectual disability have impaired perception of Zeitgebers, impacting on their ability to develop appropriate sleep patterns. Okawa and Sasaki also proposed that sleep is affected in children with intellectual disability because of impairments or malfunctions in brain regions responsible for sleep. Research has also demonstrated that parental factors such as their perception of their child’s disability, concern about the child’s health, and expectations around sleep are also associated with sleep problems (Didden et al., 2002; Stores & Wiggs, 2001; Quine, 1991, 1992). Additionally, parents with children with disabilities may be under a lot of pressure making them more susceptible to coercive behaviour traps. Blum (1999) found that children’s sleep problems were associated with maternal over-concern about the child’s health and needs.

METHOD

A 7-day baseline condition was randomly assigned to the family with a toss of a coin method (out of 7-day or 10-day baseline options). A home visit was arranged after baseline and the mother was given an information sheet on Molly’s specific sleep programme (Appendix J). Unfortunately there was a delay in starting the programme owing to the time elapsed prior to securing Molly’s room safely. The mother was contacted regularly during this period until she
was ready to start the programme. On the 25\textsuperscript{th} night the intervention was started. Daily telephone calls were made to the mother during the intervention phase.

\textbf{Special Considerations for Intervention}

\textbf{Cognitive and Communicative Ability}

There were several issues to take into consideration due to Molly’s special needs and level of cognitive functioning and communication. Firstly, a reward system was considered but rejected. It was concluded that Molly would not understand the relationship between her sleep behaviour at night and receiving a reward in the morning (a temporarily delayed reinforcement). Praise was incorporated into the bedtime routine and given when Molly complied with her mother’s requests. Secondly, Molly’s limited communicative ability meant that she could only understand simple phrases. The mother was encouraged to use a simple prompt such as “Bedtime Molly” every night and to establish a consistent night-time routine. This meant setting appropriate limits for play (emphasising that night-time was for sleep, and daytime was for play) and requesting Molly to return to sleep during the night instead of allowing her to stay up.

\textbf{Asthma}

Molly’s mother was advised to check on Molly and to give her medication when it was necessary.

\textbf{Child Safety}

The mother was encouraged to prepare the bedroom for the intervention by removing toys to make the room less stimulating and safe for Molly prior to beginning the programme. All the toys (except a few stuffed toys) were removed from the bedroom. This was done to reserve the bedroom for sleep and to help Molly understand appropriate night-time behaviour and rules. Molly enjoyed opening her bedroom windows. It was initially decided to secure the windows and doors but ultimately only the door was secured. However, the mother agreed to check regularly on Molly through a window in her bedroom door. Molly’s bedroom door was secured, but her mother remained within hearing range.

\textbf{Analysis of Presenting Problems}

Sleep onset delay and frequent night wakings were identified as target behaviours for intervention. The mother indicated that Molly would regularly co-sleep but this did not occur during baseline. The analysis indicated that the mother was permissive with Molly’s behaviour at night and there was no structured bedtime routine or set bedtime. Molly would play with her toys...
or open her windows for extended periods at bedtime. During night wakings, Molly would sometimes play with toys for long periods of time. The mother had always settled Molly with a bottle, so this was part of the conditions Molly required for sleep onset. The regular provision of a drink during the night probably increased and maintained waking. Parental attendance at night possibly stemmed from concern about Molly’s communication problems as well as issues relating to her health such as asthma, illness, and hospitalisations.

**Intervention**

The intervention began on the 25th night once the room had finally been prepared. Molly’s mother began implementing an intervention incorporating a positive bedtime routine, limits set on bedtime and behaviours around sleep, and graduated extinction.

**Positive Bedtime Routine and Parental Limit Setting**

The importance of a positive and consistent bedtime routine was explained. A suitable bedtime and wake time was negotiated (7.30pm and 7am respectively). The mother was encouraged to keep to the scheduled bedtime and wake time as consistently as possible. A positive bedtime routine involved having a quiet-time for cuddles and/or a story in the living room before settling Molly into bed. This was to encourage calm and restful behaviours before bedtime, as well as to encourage positive interactions between mother and child. Once Molly was settled into bed, the mother was advised to leave the room and allow Molly to fall asleep on her own. If Molly tried to get out of bed, the mother was advised to firmly prompt her to return to bed, followed by verbal praise if she complied with the instruction.

Rather than dispensing with the bottle altogether, it was incorporated into the bedtime routine. The mother was asked to give a bottle to Molly in the living room but to eliminate the use of the bottle during night wakings. The physical distinction of where Molly was given the bottle was made to simplify the learning task and to reinforce the bedroom as an environment for sleeping only. The bottle was also seen as a form of comfort and attachment for Molly and it was utilised in the bedtime routine to ensure the continuation of positive interaction between the mother and child. This was considered important to minimise separation difficulties.

**Graduated Extinction**

The mother was presented with a choice of standard or modified extinction (combined with trimeprazine, or graduated extinction) programmes and the advantages and disadvantages of the options were explained in detail. The mother preferred graduated extinction as she viewed it as the more gentle and child friendly approach. The graduated extinction procedure outlined by
Ferber (1985) was described. The strategy involved systematically increasing the amount of time before the parent attended to the child when she woke during the night (starting with 5 minutes and then increasing the period of time with 5-minute increments). When attending to the Molly at night, the mother was encouraged to attend to her briefly restoring sleeping position with minimal interaction so that it would be less stimulating and reinforcing. It was also emphasised that Molly should settle to sleep on her own.

Implementation of the Programme

Although the mother expressed her intention of implementing the graduated extinction strategy, she continued to attend to Molly immediately upon awakenings, settling her with a bottle throughout the programme. The mother reported that she was too tired to implement the approach and that she continued to attend to Molly straight away and gave her a bottle. So in fact, the programme the mother actually implemented consisted of reducing the amount of attention she gave to Molly when she attended to her waking and being firmer with Molly with bedtime rules and behaviour. The mother was pleased with the improvements in Molly’s sleeping resulting from a positive bedtime routine and rules. After 4 weeks and 4 days, it was jointly decided to terminate the intervention. The mother noted improvements in Molly’s sleeping and was happy to continue to give her a bottle when she woke. She no longer felt it necessary to remove the bottle at this stage as the difficulties associated with Molly’s night waking had reduced to manageable levels.

Maintenance

The mother was advised to continue with the positive bedtime routine and rules, so that improvements in Molly’s sleep behaviour could be consolidated. During this phase, regular telephone contact was stopped, however the mother was advised that she was more than welcome to contact the researcher if she required further advice, or if she changed her mind about implementing the graduated extinction strategy and eliminating the night-time use of the bottle.

Follow-Up

A follow-up was organised 3 months after the intervention. The mother was asked to complete a 2-week sleep diary, however she advised that she had difficulty recording the follow-up data because she and Molly were ill. Although 14 nights of data were obtained, this was over the course of 3 weeks and not 2 weeks as planned. Follow-up data was therefore not over 14 consecutive nights.
Data Coding

The following variables were coded and graphed for Molly: i) sleep onset delay, ii) night waking, iii) co-sleeping, iv) child illness, and v) parental non-adherence.

Reliability

The mother’s sleep diary was compared with notes made by the researcher during telephone conversations during the intervention phase. Reliability was 84%. The reliability of visual analysis of Molly’s graphs was 100% between co-raters.
Figure 3: Sleep onset delay in minutes, across conditions: Molly

Figure 4: Frequency of night wakings per night, across conditions: Molly
Sleep Onset Delay

Figure 3 presents sleep onset delay in minutes, across conditions. Prior to intervention, Molly took on average 32 minutes (a range between 5 and 90 minutes) from initially being settled into bed and sleep onset. During baseline there was an extremely variable rate of sleep onset delay. There was an immediate decrease in sleep onset delay during the intervention phase and Molly had a low stable rate of sleep onset, averaging 5 to 10 minutes. There were 2 nights (33rd and 34th night) where Molly took longer times to fall asleep. The follow-up also indicated a low stable rate of sleep onset delay.

Night Waking

Figure 4 presents the frequency of night wakings per night, across conditions. Baseline data indicated that Molly woke 5 out of 6 nights (83%). The data during this phase was high and variable, ranging from 0 to 4 night wakings (average of 1.5). The rate of night wakings immediately reduced to a low and more stable level during the intervention phase, with the majority of nights having 0 to 1 waking. There were several nights of increased night waking during the intervention phase. On the 28th and 39th nights Molly woke twice, while on the 44th night she woke 4 times. Molly woke 10 nights out of the 28 nights recorded during the intervention phase (36%). During follow-up, there was a low stable level of night waking (0 to 1 waking per night). Molly woke 6 out of 14 nights during this phase (43%). There was one night where Molly experienced 2 night wakings on the 167th night. The overall trend of night wakings was decreasing.

Co-sleeping

Figure 4 presents the nights where co-sleeping occurred. There was no co-sleeping reported during baseline or during intervention. Co-sleeping was recorded at follow-up, 5 out of 14 nights (36%).

Child Illness and Parental Non-Adherence

Child illness occurred on the 34th and 44th nights as well as from the 163rd to the 167th nights (Figures 3 & 4). On the 33rd night there was an incident of parental non-adherence during bedtime (Figure 3).
Visual Analysis

Sleep Onset Delay

Sleep onset delay during baseline and the last 2 weeks of intervention were compared and rated as a *substantial improvement*. Baseline and follow-up was also rated as showing a *substantial improvement*.

Night Waking

The number of night wakings during baseline and intervention were compared and rated as showing a *substantial improvement*. Baseline and follow-up was also rated as showing a *substantial improvement*.

Co-Sleeping

Molly’s co-sleeping during baseline and intervention were compared. This was rated as showing *no change*. Baseline and follow-up co-sleeping was rated as showing a *moderate deterioration*.

Social Validity Measures

The mother commented that she was very satisfied with the programme, and indicated that the intervention had met her needs. She reported that the instructions and strategies were easy to follow, and telephone support was frequent but helpful. The mother rated the programme as *somewhat stressful* to implement.

DISCUSSION

There were substantial improvements in Molly’s sleep behaviour as a result of the behavioural intervention. There were marked reductions in sleep onset delay and the number of night wakings for Molly. During baseline Molly’s sleep onset delay averaged 35 minutes and the longest recorded sleep onset delay during this phase was 90 minutes. A positive bedtime routine and appropriate parental limit-setting reduced sleep onset delay to 5 to 10 minutes indicating rapid sleep onset from the time of being settled to bed. On the 25th night Molly got out of bed and started banging and opening her bedroom windows. Her mother reported being firm with Molly, resettling her to bed with a teddy bear. Molly achieved sleep onset within 5 minutes on this night. During the intervention phase there were only two incidents of exceptionally delayed sleep onset for Molly. On the 33rd night, Molly’s sleep onset delay was 40 minutes, explained by an incident of parental non-adherence. The mother indicated that she had allowed Molly to get out of bed and play with toys on this night and as soon as she prompted Molly she fell asleep.
within 10 minutes. Thus the mother had not set appropriate limits for Molly's bedtime behaviour. On the 34th night, the mother recorded that Molly took 80 minutes from initially being settled to bed to sleep onset, this extreme delay of sleep onset was explained by illness. Despite these disruptions, the overall trend showed an improvement in sleep onset delay and this was maintained at follow-up with Molly settling consistently within 5 to 10 during this phase.

Prior to intervention Molly was waking up to 4 times a night. This was reduced to between 0 and 1 night waking per night during intervention and follow-up phases. There were some disruptions noted in the programme. On the 29th night, an aunt and cousin stayed at Molly's house and the mother commented that Molly appeared unsettled. Molly had 2 night wakings that night. A marked level of night wakings on the 44th night (4 night wakings) coincided with illness. Overall there were substantial improvements in night wakings and these changes were maintained at follow-up. The highest reported number of night wakings during follow-up was 2 night wakings on the 167th night. This was also associated with child illness.

Improvements in Molly's sleep behaviour were seen early in the intervention phase. There was also no observed PERB when the intervention was implemented and the mother did not report heightened crying or signalling behaviours. Because of these factors, it is likely that the mother in fact started the intervention earlier than agreed upon. The mother indicated that started changing her responses to Molly at night, by minimising her interactions, being firmer with her instructions and setting limits on her behaviour. This would explain the rapid resolution of sleep onset delay and improvements in night waking observed from the beginning of the intervention phase.

The mother reported that co-sleeping was a regular issue for Molly, but there were no incident of co-sleeping recorded during baseline or intervention phases. Co-sleeping was recorded during follow-up 5 out of 14 nights. This was rated as a moderate deterioration in the visual analysis. The low rate of night wakings during this follow-up period may be partly explained by the frequency of co-sleeping. Molly would be less likely to wake frequently during the night if she was sleeping in her mother's bed. However, it is also important to note that Molly was ill during the follow-up phase for several days and had asthma. The mother's concern for Molly may have contributed to co-sleeping during this time, especially as Molly could not communicate her needs. Although during the intervention there were no accounts of co-sleeping, this was not maintained at follow-up with data showing an increase in the occurrence of this behaviour.

Overall the mother reported being satisfied with the sleep programme and indicated that it met her needs. She advised that the intervention was somewhat stressful to implement. She also reported that the telephone contact and support throughout the intervention was helpful.
Molly showed marked improvements in her sleeping after the implementation of an intervention that combined a positive bedtime routine and parental limit setting strategies. The mother decided not to eliminate the provision of a bottle during night wakings and declined to start the graduated extinction strategy although she had initially chosen to do so. There may be several reasons why the mother decided against making these changes. Firstly, the mother was pleased with the improvements that she had observed with Molly’s sleeping. She indicated that less effort was required to settle Molly to sleep and that night wakings were shorter and less frequent. This may have contributed to her decision not to use other behavioural strategies.

Secondly, the mother’s ability to implement a graduated extinction programme may have been limited due to tiredness and stress associated with having a child with a disability as well as family circumstances such as being a single-mother with limited financial means and social support. Molly had a sibling and this would have divided the mother’s time and may have affected her ability to implement an intervention. The mother mentioned during one telephone conversation in reference to starting the graduated extinction technique, “last night, I wasn’t in the state of mind. I didn’t feel I had the patience”. It is difficult to determine whether this was due to her motivation and commitment to the programme, or whether her ability to cope was affected by her stressful circumstances, or a combination of both. Parental attendance during the night and the provision of a bottle were long-standing aspects of a behaviour trap for this family. The continuation of this behavioural repertoire would be reinforcing for the mother who could avoid the aversive PERB. Fatigue was frequently reported by the mother as a major obstacle to implementing change in her night-time attending. This emphasises the importance of supporting families during intervention as well as adapting interventions to suit individual needs.

Molly’s sleep disturbance was contributed to by an inconsistent bedtime routine and rules, parental attendance and sleep onset conditioning of a bottle. A likely factor contributing to the development of Molly’s sleep problems was her mother’s concern about issues relating to her disability and health. Asthma and hospitalisations would have contributed to the mother’s concern about Molly’s health and increased her responsiveness to her needs at night-time especially as Molly had limited ability to communicative her needs. A behavioural intervention involving a positive bedtime routine and parental limit setting was successful in reducing sleep problems such as sleep onset delay and night wakings for this child with GDD, to a level that was satisfactory to the parent.
CHAPTER 5. CASE STUDIES THREE AND FOUR: HAMISH AND LEWIS

In this chapter two, case studies are presented together. There is considerable overlap of information between the two cases because the two boys were siblings and had the same disability. Nonetheless, the two boys had different sleep problems and required separate interventions. In the first section of this chapter, general information about the two boys and their disability are discussed and intervention strategies and methodology relevant to both children will be addressed. In the second section, the analysis of presenting problems and specific intervention strategies as well as the results of the intervention will be presented for each child separately. A general discussion completes this section.

Hamish was 6 years and 3 months of age and Lewis was 3 years and 3 months of age. The two brothers lived with their mother, father, and older half-brother, Jamie, aged 9 years. The socio-economic status of the family was rated 5 on the Elley-Irving Scale (Elley & Irving, 2003). Hamish attended a mainstream primary school and Lewis attended kindergarten in the mornings. Hamish had been diagnosed with multiple epiphyseal dysplasia (MED) at the age of 3 years by an orthopaedist. At the time of the intervention, Lewis’s diagnosis of MED was suspected and was confirmed by follow-up. Lewis was included because he presented with the same signs as his brother such as walking stiffly, complaining of sore legs and tiredness, and frequently requesting to be picked up and carried. He also had the more disrupted sleep problem and slept in the same room as his brother. The parents reported that both children had had sleep problems since infancy. The role of MED in the children’s sleep problems is complex. Initiating factors in infancy such as the parental responsiveness to child behaviours at night were maintained and exacerbated by the diagnosis of MED in early childhood. Sleep problems for the two children need to be understood in relation to the role of pain, parental anxiety about pain and the children’s wellbeing, as well as children’s fears (particularly Hamish) about his illness.

Hamish and Lewis were referred to the CSP by their parents who had discovered the CSP by word of mouth. Hamish was referred for night waking, co-sleeping, and nightmares, while Lewis was referred for bed refusal, sleep onset delay, night waking and co-sleeping. The parents had tried various methods in an attempt to eliminate the boys’ sleep problems. These were installing night-lights, putting a television and radio in the boys’ room, unmodified and graduated ignoring. None of these approaches had worked. A star chart had also been used but the parents forgot to mark it. Both Hamish and Lewis have been given promethazine while Hamish had also been prescribed trimeprazine. The parents reported that the medication had had no effect and both the boys continued to wake frequently.
MULTIPLE EPiphySEAL DYSPLASIA (MED), PAIN, AND SLEEP

MED is an autosomal dominant genetic disorder characterised by abnormalities in the growth and remodelling of cartilage and bones (Sillence, Horton, Rimoin, 1979, as cited in Trivedi, 2003). The reported incidence of bone dysplasias is approximately 1 in 3000 to 1 in 5000 births (Anderson, 1989, cited in Trivedi, 2003). The main clinical findings of MED are frequently painful joints (especially weight bearing joints) with restricted mobility and waddling gait, normal or moderately short stature with normal body proportions, and back pain (Spranger, Brill, & Poznanski, 2002; Trivedi, 2003). These clinical manifestations usually present after the second year of life, but sometimes are not recognised until early adulthood (Spranger et al., 2002). MED frequently leads to progressive degenerative osteoarthroses, where in severe cases if left untreated, patients may lose their ability to stand and walk by the age of 50 years due to crippling arthritis (Spranger et al., 2002; Trivedi, 2003). Joint replacement and surgery may also be necessary in adulthood, and in some cases in adolescence. Non-weight bearing exercise such as swimming and biking is encouraged to help maintain joint function and stability. Monitoring of weight is also advised to minimise the impact on joints (Spranger et al., 2002).

The sleep research on physical disabilities and chronic pain is limited. As mentioned in Chapter One, there has been some evidence to suggest that children who have chronic pain such as rheumatoid arthritis (Zamir et al., 1998), and burn injury (Lawrence et al., 1998) are at greater risk of developing a sleep problem than the general population. Lewin and Dahl (1999) in their review of sleep and paediatric pain management state that not only can pain interfere with the quality and quantity of children’s sleep, insufficient sleep can lead to behavioural and emotional changes, such as negative mood, impaired self-control and attention, and diminished motivation towards goals and rewards. The authors note that these changes may impact on the coping skills necessary for effective pain management. Hence, sleep is indicated as playing an important role in the management of paediatric pain. Pain has been associated with prolonged sleep onset as well as disruptions in the quantity and quality of sleep. Cognitive and psychological disturbance such as anxiety and depressed mood have been indicated in children with chronic pain (Pao, 1998 cited in Lewin & Dahl, 1999). Fears and anxiety in children have been shown to negatively affect both the amount of pain reported and sleep patterns (Lewin & Dahl, 1999).

METHOD COMMON TO HAMISH AND LEWIS

An assessment interview was conducted in the family home with the parents, all three children, and the researcher. A 12-day baseline was randomly assigned to the family with a “toss of a coin” method out of a 10-day or 12-day baseline options. A home visit was arranged after
the baseline (again with the parents, children and researcher), and an information sheet about the sleep intervention was given (Appendix K).

Once baseline was completed, the intervention phase was started on the same night (13th night) with both children. Daily telephone contact (ranging from 5 minutes to 60 minutes duration) was made during the intervention to give advice and support to the parents, usually the mother. After 19 weeks, contact was reduced to every second day.

**Intervention**

A positive bedtime routine and rules, and rewards for appropriate sleep behaviour were implemented with all of the children in the family, including Jamie who did not have a sleep problem. This was to ensure a joint family effort with the sleep programme and to make it enjoyable for the children and to minimise Jamie’s feelings of exclusion.

**Positive Bedtime Routine and Bedtime Rules**

During a home-visit prior to beginning the intervention, the importance of a positive and consistent bedtime routine and bedtime rules were discussed. The family was assisted to establish appropriate bedtimes and a list of bedtime rules together. The children recorded these on paper and this list was put on the refrigerator. Lewis’s bedtime was set at 7pm, Hamish’s was at 7.30pm, and Jamie’s bedtime was set at 8pm. Bedtime rules included “brushing my teeth before bedtime”, “going to bed at my bedtime”, “no telling of scary stories before bedtime”, and “staying in my own bed”. The importance of all family members to keep to the family rules was emphasised. A positive bedtime routine including “quiet-time” was also incorporated. Quiet time was fifteen minutes for a quiet activity and/or cuddles with a parent before bed. This was to promote positive interactions between the parents and children as bedtime had become full of negative interactions. Positive interactions during the day were also emphasised to compensate for the attention to be eliminated at night.

**Reward System**

A reward box, with inexpensive toys and stickers to choose from, was introduced to the family with the aim of reinforcing positive sleep behaviour. Jamie received a reward from the reward box for not telling scary stories to Hamish. Larger rewards such as more expensive toys or a family day were reserved as a reward to reinforce several nights of undisturbed sleep. This was left to the discretion of the parents. A star chart was also suggested to the family but was not implemented. The parents had tried a chart in the past and preferred not to use one.
Follow-up

A follow-up on the two boys was completed 2 ½ months after Lewis had completed his programme. The follow-up phase was implemented concurrently for the two boys to make it easier for the parents. The follow-up for this family was conducted earlier than originally planned to avoid the busy Christmas period. The mother completed 2 weeks of sleep diaries.

Data Coding

The following variables were coded and graphed for Hamish and Lewis: i) night waking, ii) co-sleeping, iii) child illness, and iv) parental non-adherence. Sleep onset delay was coded and graphed for Lewis, and nightmares was coded and graphed for Hamish.

Reliability

The reliability between the mother’s sleep diary and the researcher’s notes was 97% for Hamish, and 98% for Lewis.

The father was asked to complete a sleep diary independently for 3 nights during intervention and 3 nights during follow-up for each of the two boys. This record was compared with the mother’s sleep diary to calculate reliability. The reliability between the parental records during intervention and follow-up phases for both Hamish and Lewis was 100%.

The reliability of visual analysis was 100% between the co-raters.

CASE STUDY THREE: HAMISH

Hamish presented with regular night wakings and co-sleeping difficulties. He also experienced frequent nightmares. Because nightmares are a type of parasomnia, this was beyond the scope of the original research. Therefore, a brief literature review on nightmares is presented below.

Nightmares

Nightmares are bad dreams. They occur in the REM sleep state. Most people experience nightmares from time to time, however, nightmares can become a problem if they occur frequently, are extremely distressing for the child, and require constant parental comforting. Nightmares usually occur in the second half of the night when REM sleep frequency is higher. Ferber (1985) commented that nightmares often represent emotional conflicts that take place during the day. In Hamish’s case a concern was that his nightmares might have reflected his worries about his illness.
Nightmares are a common sleep problem reported in children. Salzarulo and Chevalier (1983) studied a range of sleep problems in their sample of 218 children aged 2 to 15 years from paediatric and psychiatric consultation clinics. Overall, nightmares were reported in 28% of children. Nightmares occurred more frequently in children aged between 6 to 10 years with 41% reported to have nightmares, compared with 24% of children aged 2 to 5 years, and 22% of children aged 11 years. Vela-Bueno et al. (1985) looked at the prevalence of nightmares in their study of 900 typically developing children aged 6 to 12 years. The authors found that 22% of children had nightmares. A review by Leung and Robson (1993) reported that the prevalence of nightmares peaks between the ages of 3 to 6 years. Developmental, genetic, psychological, and organic factors have been implicated in nightmares. A study by Mindell and Barrett (2002) found that children who experienced nightmares had higher levels of anxiety than children who did not experience nightmares as rated on the State-Trait Anxiety Inventory for Children. This emphasises the importance of the assessment of anxiety in children who present with frequent nightmares.

Children’s chronological age and developmental level may also be related to the type of fears expressed and the content of scary dreams. Bauer (1976) found that younger children were more likely to report fears and scary dreams of ghosts and monsters than older children (74% of children aged 4 to 6 years compared with 5% of children aged 10 to 11 years), while older children were more likely to report fears and scary dreams of bodily injury and physical danger (55% of children aged 10 to 11 years compared with only 11% of children aged 4 to 6 years). Muris, Merckelbach, Gadet, and Moulart (2000) reported that in a group of typical children aged 4 to 12 years of age, 75.8% had fears, 67.4% had worries, and 80.5% had nightmares. Children aged 4 to 6 reported nightmares of imaginary creatures, personal harm or harm to others, and animals. Children aged 7 to 9 years had the highest reported nightmares at 95.7% with the most common dream content being imaginary creatures, being kidnapped, and personal harm or harm to others. The children in this study attributed their scary dreams to influences such as television.

Muris, Merckelbach, Ollendick, King, and Bogie (2001) found that 73.3% of normal school children (N=176) reported night-time fears. The prevalence of nightmares was more elevated in children aged 7 to 9 years. The study found that in 10% of children, night-time fears were related to one or more DSM-III-R anxiety disorders. This highlights the need for a thorough assessment for anxiety when children present with night-time fears and nightmares.
Interventions for Children’s Nightmares

Maurer and Shaefer (1998) advised that for occasional nightmares a parent should explain to the child that dreams are not real and reassure the child of his or her safety. Exposure to potential daytime stressors such as television with scary or distressing content should be limited. It is common for children to have nightmares, however if they become persistent and frequent then treatment is recommended (Ferber, 1985; Maurer & Shaefer, 1998). The literature on the treatment of nightmares in children is sparse, and there is only a small number of single case studies. A review by Owens, France, and Wiggs (1999) reported that there were no controlled studies addressing behavioural or cognitive-behavioural treatments for nightmares. Thus the efficacy of these treatments needs further exploration.

Roberts and Gordon (1979) incorporated a response prevention procedure, extinction, and systematic desensitisation to reduce the occurrence of nightmares and night terrors in a 5-year-old girl who received burns to approximately 20% of her body in an accident. The girl had developed a strong phobia to fire-related stimuli. The mother was asked to stay in the child’s room for the entire night and to awaken the child as soon as she began to show the first signs of a nightmare (i.e. clutching her nightgown) and to allow her to resettle to sleep without attending or talking to her. The extinction of this parental response to the nightmares then followed.

Systematic desensitisation techniques were implemented when nightmares reoccurred after a subsequent hospitalisation. Nightmares that occurred 5 to 22 times per night during the baseline phase were successfully eliminated with this intervention. This progress was maintained at a 6-month follow-up.

Other strategies that have been used to treat children’s nightmares include, systematic desensitisation, guided imagery and relaxation. Cavior and Deutsch (1975) used systematic desensitisation techniques to reduce anxiety produced by a recurrent aversive dream in a 16-year-old boy. Therapist sessions involved guiding the boy through the course of the dream with instructions to remain calm. Although the dreams continued to occur, the treatment was successful in eliminating the anxiety associated with the dream after three sessions. This was maintained at 6-month follow-up. Palace and Johnson (1989) treated a 10-year-old boy with co-sleeping and nightmares related to two traumatic events (two separate automobile accidents) with a dream re-organisation approach. The intervention involved systematic desensitisation with coping self-statements and relaxation techniques. The therapist guided the child to master a hierarchy of dream scenarios with the use of an imaginary superhero. Nightmares and co-sleeping were eliminated with the intervention and this was maintained at a 6-month follow-up.

White (1985) developed “a fear busting and monster taming” approach to treating children’s fears and nightmares. The strategy involves addressing the interactions between
family members by changing the *fears lifestyle* that reinforces the child’s fears. The child is also asked to draw pictures of his or her fears and then to put them in a designated box at night-time securing it with string. According to White (1985) the box should be suspended, so that it is difficult for monsters to escape or cause havoc, as their feet cannot touch the ground. In the morning the monsters are released. This is explained by the *fourth rule of monsters*: “Since monsters grow more fearsome with night-time practice and more funny with day-practice, if children want to have a funny time then they should stop their monsters from having night practice” (White, 1985, p. 31). This strategy was illustrated with a case example.

Research on the treatment of children’s nightmares is extremely limited and there have only been a handful of single-case studies. None of the studies have employed an experimental control. Most of the research has addressed nightmares relating to specific anxieties or trauma and have not addressed nightmares of a more general or developmental nature. The article by White (1985) described a novel approach to treating typical fears and nightmares in children. However, this strategy is yet to be tested in an experimental design. There has been no controlled study in this area and so efficacy of these treatments has not been established.

**METHOD**

**Special Considerations for Intervention**

**Child Anxiety**

The initial assessment interview indicated that Hamish did not meet DSM-IV-TR (American Psychiatric Association, 2000) criteria for an Anxiety Disorder. A clinical interview with Hamish was also conducted to discuss his nightmares and daytime worries. The content of Hamish’s nightmares were developmentally typical involving dinosaurs, monsters and tigers.

Partway through the intervention, however, Hamish had two nightmares that were more concerning in nature and possibly anxiety related. One nightmare was about a man who was hiding under his bed with a bat and waiting to swipe his legs. In the second nightmare, Hamish was biking with his older brother and they were crossing the railway lines and the brother did not bike fast enough over the tracks and was hit by the train. This raised concern that his nightmares could stem from anxiety relating to his MED.

These two nightmares occurred during a time when there was increased talk amongst the parents about MED because Lewis was being reassessed. In response, the researcher facilitated a meeting with Hamish and his family to discuss MED and to clarify his understanding of the disorder. The meeting revealed that Hamish knew that he had MED but had no knowledge about what it was. This may have been a source of anxiety. The session incorporated educating Hamish about MED as well as discussing associated conditions such as pain and fatigue. During this
meeting, Hamish disclosed that he was being bullied at school about his gait and his limited ability to participate in sport. The parents agreed to contact the school in regards to this bullying problem, as this would have further contributed to his anxiety about his illness.

Child Pain

Pain is commonly associated with MED and the experience of pain often increases as the condition becomes more advanced. Hamish certainly experienced pain to a greater degree than typically developing children. Although Hamish did not cite pain as a problem that affected his sleep, pain still may have contributed to sleep fragmentation (Lewin & Dahl, 1999). His parents also, were concerned about pain and would place extra blankets and dress him in warmer clothes on colder nights. Concern about pain may have led Hamish’s parents to be more responsive at night and more lenient with night-time behaviours such as co-sleeping. The parents were instructed to consider pain during the daytime and to ask Hamish before bedtime whether he was experiencing pain so they could attend him accordingly (such as providing warmer clothes and blankets, massaging his legs, or giving him a bath). The programme therefore incorporated addressing parental concerns and their interactions with Hamish at night but did not intervene directly on pain management.

Analysis of Presenting Problems

Night waking, co-sleeping, and nightmares were identified as target behaviours. Hamish was easy to settle but from infancy the mother’s feeding at night-time contributed to inadequate stimulus control. As Hamish grew older, parental presence during night wakings continued and co-sleeping was also established. Parental concern about pain because of MED further contributed to attending behaviours at night-time, and may have increased the likelihood of falling into a behaviour trap (France & Blampied, 1999).

Hamish started to have frequent nightmares during his first year of school and reported being scared at night. His parents wondered if this was an excuse to sleep in their bed for Hamish. Co-sleeping was associated with comfort and an escape from fear, whereas awakening during the night was associated with the experience of nightmares, fears and distress. This conditioning is likely to have maintained Hamish’s sleep problems and his need to seek parental attention and comfort even when he did not have a nightmare. Hamish’s parents were highly responsive to him at night-time and had inappropriate limit setting. Hamish had learnt that seeking attention from his parents during the night, helped him avoid feeling scared and going to bed alone. His parents had learnt how to avoid Hamish’s distress behaviours by comforting him and engaging in co-
sleeping. This established behavioural repertoire is likely to have prevented Hamish from learning to how to deal with his fears on his own.

Hamish indicated that he generally had nightmares about dinosaurs, monsters, and tigers chasing him. Jamie regularly told Hamish scary stories and this may have stimulated the occurrence of nightmares. Although issues relating to MED and pain were considered, an assessment did not indicate clinical anxiety. In general the content of his nightmares was developmentally typical, but on occasion may have reflected anxiety about illness and bullying.

**Intervention**

Hamish’s sleep programme incorporated a positive bedtime routine and a reward system. These strategies were utilised throughout the intervention. The intervention for Hamish’s nightmares was modelled on the *Fear Busting and Monster Taming* approach, developed by White (1985) with the goal of obtaining a *Fear Busting and Monster Taming Certificate*. The strategy requires the child to draw pictures of his fears and monsters and to place them in a box secured with string at bedtime every night. The monsters are let out during daytime.

**Implementation of the Intervention**

The first step was to implement a positive bedtime routine, bedtime rules and a reward system. Because Lewis’s sleep problem was causing the parents the most stress and may have limited their ability to employ multiple strategies, the decision was made to delay implementing specific intervention for Hamish’s nightmares. Meanwhile the parents were asked to start just setting appropriate boundaries for Hamish. Upon a night awakening the parents prompted him to return to bed. If Hamish had a nightmare, the parents were advised to reassure him that he was safe and to comfort him briefly before settling him back into bed, this was continued until the parents were ready to implement other strategies for Hamish.

**Fear Busting and Monster Taming Certificate**

On the 20th day a *Fear Busting and Monster Taming Certificate* (White, 1985) was explained to Hamish and a goal of 12 consecutive nights of nightmare-free sleep was negotiated. Hamish’s night wakings and nightmares began to subside with the introduction of the certificate as a goal. Hamish received his certificate on the 46th night. Hamish maintained sleeping through the night without nightmares for a further 23 consecutive nights.
Fear Busting and Monster Taming Box

The mother reported that Hamish had nightmares on the 70th, 79th, and 86th nights and this re-occurrence of nightmares led to the decision to implement the monster-taming box (White, 1985). On the 89th day of the programme the use of a fear busting and monster taming box was suggested to Hamish and his parents. The mother did not start using the box until the 101st night of the programme.

Advanced Certificate in Fear Busting and Monster Taming

A home-visit was arranged on the 120th day and Hamish was introduced to the concept of working towards a second certificate, this time an advanced certificate in fear busting and monster taming. This was emphasised as a challenging goal and Hamish was told we know of no other child in the city who had attempted it. During this meeting, the criteria for the certificate were negotiated with Hamish and the mother. The goal of 4 weeks was established. Because this was a long period of time, it was emphasised that it would be acceptable for Hamish to have a few nightmares during this time. It was negotiated with Hamish that he was able to have three nightmares during the 4 weeks and still obtain the certificate.

The intervention was finished once Hamish received his second certificate after 20 weeks and 1 day. Hamish was having fewer nightmares but was also managing his nightmares and fears better, such as turning on the hall light on his own, and not seeking parental attention as often. Hamish’s mother reported that he no longer seemed distressed upon awakenings. His mother also indicated being happy with the improvements and felt that intervention was no longer necessary. During a home visit when the certificate was presented to Hamish he also told the researcher that he no longer needed my help and no longer needed to be part of the sleep programme. He reported sleeping through the night and having good dreams instead of nightmares.

Maintenance

The parents were advised to maintain the programme by using minimal methods to reassure Hamish (such as verbal reassurance and prompts to go back to bed) if he woke with a nightmare and came to seek parental comfort. They were also advised to encourage Hamish to settle in his own bed. Furthermore, Hamish’s parents were asked to praise Hamish when he managed fears and nightmares on his own. If frequent nightmares re-emerged the parents were asked to consider possible precipitants and to seek advice if there was concern about anxiety or other psychological problems. Regular telephone contact was discontinued, and the parents were advised to contact the researcher if they wanted further advice or support.
RESULTS

Figure 4: Frequency of night wakings per night, across conditions: Hamish

Night Waking

Figure 4, presents the frequency of night wakings per night, across conditions. Hamish woke regularly during baseline, with night waking occurring 7 out of 12 nights (58%). There were 2 night wakings on the first night of the baseline phase, otherwise the level of night wakings was stable with most nights having 0 to 1 waking per night. During the implementation of the positive routine and reward box, night wakings were less frequent, with Hamish waking only 2 out of 7 nights (28%). After the introduction of the first certificate on the 20th night of the programme, there was night wakings on the 22nd, 24th, 25th, 29th, and 33rd nights followed by a stable rate of 0 waking per night for 16 consecutive nights. On the 58th night there was a marked level of night waking, with 4 night wakings that night. There was a further 6 nights with wakings during this phase. Hamish woke 14 out of 81 nights during this phase (17%). During the monster taming box phase, Hamish did not wake for the majority of nights. Only 2 out of 19 nights were recorded with wakings (11%). With the introduction of the second certificate sleep was undisturbed on most nights. Hamish woke 4 out of 36 nights (11%). During the follow-up phase there were 2 out of 14 nights where Hamish woke during the night (14%). The overall trend of night wakings was decreasing.
Nightmares

Figure 4 presents the nights where nightmares occurred. During baseline, Hamish was reported to have a nightmare on 5 out of 12 nights (41%). With the implementation of the positive routine and the reward box, Hamish had a nightmare 1 out of 7 nights (14%). With the introduction of the first certificate, there were initially two nightmares on the 24th, and 33rd nights. This was followed by a 36-day period without nightmares (from the 34th to 69th nights). Hamish had a nightmare on the 70th night as well as on the 78th night. There was a cluster of three nightmares on the 86th, 89th, and 91st nights. During this phase, Hamish had a nightmare 7 out of 81 nights (9%). After the introduction of the monster-taming box, there were two reported nightmares out of 19 nights (10%). One of the two reported night wakings on the 104th night was recorded as a nightmare. Hamish had a nightmare on the 105th night. During the phase of the second certificate, there were four nightmares recorded out of 36 nights (11%). The overall trend of nightmares during the intervention was decreasing. There were no nightmares recorded during the follow-up phase (0%).

Co-sleeping

Figure 4 presents the nights where co-sleeping occurred. There were 5 out of 12 nights of co-sleeping coded for the baseline phase (42%). Once the intervention was implemented there was 6 nights of co-sleeping recorded out of a total of 143 nights (4%). These occurred on the 16th, 25th, 29th, 33rd, 70th, and 105th nights. There were higher rates of co-sleeping reported during baseline and at the beginning of the intervention. There was no record of co-sleeping during the follow-up phase (0%).

Child Illness and Parental Non-Adherence

Child illness was recorded on 4 nights during the intervention phase (58th, 94th, 95th, 96th nights). Co-sleeping during intervention was recorded as parental non-adherence and occurred on the 16th, 25th, 29th, 33rd, 70th, and 105th nights. On the 79th night Hamish watched an adult television programme with witches and vampires. This was also considered to be parental non-adherence, as they had agreed to restrict Hamish’s access to materials of this nature. There were 2 nights (237th and 240th nights) during follow-up that were recorded with child illness (Figure 4).
Visual Analysis

Night Wakings

The frequencies of night waking during baseline and the last 2 weeks of intervention were compared. This was rated as showing a *substantial improvement*. Baseline and follow-up phases were compared and determined as showing a *substantial improvement*.

Co-sleeping

Baseline and intervention comparisons of co-sleeping led to ratings as a *substantial improvement*. The comparisons between baseline and follow-up were also rated as showing a *substantial improvement*.

Nightmares

The frequencies of nightmares during baseline and the last 2 weeks of intervention were compared and rated as showing *no change*. Comparisons between baseline and follow-up however were rated as showing a *substantial improvement*.

DISCUSSION

There were substantial improvements in night waking and co-sleeping behaviours after behavioural intervention, and these improvements were maintained at a 2½-month follow-up. Hamish woke 7 out of 12 nights during baseline and there was a marked reduction in night wakings with intervention. There were only 3 night wakings indicated in the final 2 weeks. At follow-up, Hamish had only 2 nights of disrupted sleep suggesting that improvements were maintained. It is likely that these wakings were caused by illness. Co-sleeping was frequent during baseline, reported 5 out of 12 nights, and eliminated with the intervention. No incident of co-sleeping was reported in the last 2 weeks of the intervention or during follow-up.

The frequency of nightmares also decreased but a reoccurrence during the last 2 weeks of intervention resulted in a *no change* rating. During the last 2 weeks of intervention Hamish had reported three nightmares. Although this was equivalent to the number of nightmares during the baseline phase, it is important to comment that Hamish had improved his management of nightmares. On the nights where he had a nightmare, Hamish woke, told his mother and returned to bed on his own with a small verbal prompt; settling within 5 minutes of wakening. This contrasts to co-sleeping during baseline. There were several nights towards the end of the intervention where Hamish woke, turned on a light and resettled without waking his parents. The follow-up was rated showing a *substantial improvement* in the frequency of nightmares, with no recorded nightmares during this period.
There were several incidents that disrupted the progress of the programme. On the 58th night Hamish was ill and there were 4 night wakings recorded that night. The second time of illness (94th to 96th nights) did not appear to markedly affect his sleeping. Parental non-adherence was initially high at the beginning of the programme but this decreased as the programme continued. The parents allowed Hamish to sleep in their bed after a nightmare on the 16th, 25th, 70th and 105th night, and this may have delayed the intervention progress. On the 79th night the parents allowed Hamish to watch an adult television series about witches and vampires and Hamish experienced a nightmare on this night. On the 105th night Hamish’s waking was related to excitement over a family event.

The behavioural intervention was successful in eliminating co-sleeping and reducing night waking and eventually nightmares to low levels of occurrence in a child with MED. In addition, Hamish was less irritable during the daytime and improved his performance at school. The parents attributed these improvements to the sleep programme.

CASE STUDY FOUR: LEWIS

METHOD

Special Considerations for Intervention

Asthma

Lewis had asthma. The parents were instructed to attend to the him and to give him medication during asthma bouts.

Child Pain

Parental awareness of pain associated with MED would also have contributed to parental concern about pain for Lewis. Parental anxiety about pain may have led the parents to become more responsive and more permissive with night-time behaviours such as co-sleeping. The programme involved addressing parental concern in relation to interactions with Lewis at night. The parents were encouraged to ask Lewis if he was experiencing pain and to attend to his needs when necessary by providing warmer clothes and blankets, massaging his legs, or giving him a bath.

Developmental Considerations

Because of Lewis’s age and cognitive development, tangible rewards were able to be used to reinforce appropriate sleeping behaviours. Stickers, inexpensive toys and treats were incorporated in the programme with the aim of reinforcing good sleep for Lewis.
Toileting was another issue that needed consideration. Lewis was learning to be dry at night. Lewis was allowed to go to the toilet during the night and the parents were advised to take him back to his own bed with minimal interaction.

Analysis of Presenting Problems

Lewis presented with bed refusal, sleep onset delay, night wakings and co-sleeping. As an infant Lewis was difficult to settle and woke frequently. These sleep problems were exacerbated by feeding difficulties and reflux. Lewis had always been a fussy eater and his parents had difficulty trying to get him to put on weight as an infant and toddler. Parental concern about his health may have underpinned over responsiveness to his night-time behaviours. Asthma has also contributed to parental concern and attending behaviours. His asthma required the parents to check and comfort him during the night and occasionally administer a Ventolin inhaler for breathing difficulties. Furthermore, the possibility of MED would have added to parental concern. Therefore, issues relating to child health and concern about MED may have increased the likelihood of the parents falling into a behaviour trap (France & Blampied, 1999).

The analysis indicated that the parents were highly responsive to Lewis’s night-time behaviours and had inappropriate limit setting. Lewis had learnt that coercive behaviours such as tantrums and crying helped him avoid going to bed at the expected time and allowed him to stay with the parents in the living room. Despite being the youngest child in the family, Lewis was going to bed the latest. When the parents tried to settle Lewis to bed at an earlier time, sleep onset would be delayed due to crying and tantrums. The parents had also learnt to avoid these negative child behaviours by allowing Lewis to stay up until he was tired or asleep on the couch. They would then transfer him into bed. Owing to this inappropriate parental limit setting and stimulus control, Lewis had associated parental presence with sleep onset, and therefore required parental presence during the night. Lewis would often wake and sleep in his parents’ bed in the middle of the night. Occasionally one of the parents would co-sleep with Lewis in his bed to help him sleep through. Co-sleeping was a frequent problem for his parents.

Intervention

Lewis’s sleep programme incorporated a positive bedtime routine and a reward system. The parents were asked to remind Lewis everyday about the rewards for sleeping through the night in his own bed. Lewis’s parents were advised to keep to the regular bedtime as much as possible and resist child demands and protests. The bedtime routine incorporated quiet-time, having a bath, getting changed, and brushing his teeth. Lewis was settled into bed around 7pm. The parents were asked to ignore Lewis’s negative behaviours as long as he remained in his bed.
If Lewis got out of his bed, then the parents were asked to lead Lewis back to his bed without talking and to shut the bedroom door. The parents were advised to hold the door closed until Lewis settled himself to sleep. This procedure was also to be implemented for night wakings if he came out of his bedroom. The procedure had been explained to Lewis during the day.

Implementation of the Intervention

Lewis's parents implemented the positive bedtime routine and made changes to their interactions with Lewis at night-time. The reward system was introduced and gradually phased out as the intervention progressed by systematically increasing the number of nights (of staying in his own bed) required to earn a reward. The parents began to set limits on Lewis's behaviours during the night-time, such as enforcing the regular bedtime. Although closing the door was part of the programme, the parents did not implement this approach. They warned Lewis that his door would be shut but did not do so. Ultimately, the intervention that the parents implemented was to attend to Lewis upon night awakenings and to settle him with minimal interaction.

On the 100th day of the programme, a home-visit was arranged and the parents' concerns about ignoring Lewis were addressed. The parents believed that Lewis was waking for genuine reasons such as outside noises. The parents reaffirmed their commitment to the programme and agreed to allow Lewis the opportunity to settle himself back to sleep in these instances. Instead of shutting the door for the whole night, it was agreed the door would be shut for a few minutes only and for progressively longer times upon each night awakening (Ferber, 1985). Although the parents agreed to use this graduated strategy they did not do it, using warnings only.
Figure 6: Sleep onset delay in minutes, across conditions: Lewis

Figure 6: Frequency of night wakings per night, across conditions: Lewis
Bed Refusal / Sleep Onset Delay

Figure 6 presents sleep onset delay in minutes, across conditions. For all but 2 nights during baseline, Lewis was taken to bed. These were the 2 nights during the baseline phase where the parents had sent him to bed awake at earlier times of 7.30pm and 8.30pm.

In the initial stages of the programme there was a sudden increase in sleep onset delay with high and variable rates (0 minutes to 75 minutes) followed, after night 34, by a low stable rate (5 to 10 minutes). Sleep onset delay remained low for the rest of the intervention phase with only 3 nights of exceptions (54th, 91st, and 159th nights). The follow-up data indicated a moderately variable rate of sleep onset delay, with most nights ranging from 0 to 20 minutes, with one exceptional night of sleep onset delay of 90 minutes (249th night). The overall trend of sleep onset delay was decreasing.

Night Waking

Figure 7 presents the frequency of night wakings per night, across conditions. Night waking was high and variable during baseline, ranging from 0 to 4 wakings per night (average of 1.2). Lewis woke 10 out of 12 nights (83%) during the baseline phase. During intervention, the number of night wakings reduced to a more stable rate of 0 to 2 wakings per night. From the 71st night the number of night wakings increased to a high variable rate of night waking with up to 4 night wakings a night, this was followed by a decrease in the frequency of night wakings from the 94th night to 0 to 1 night waking per night. During the intervention phase, Lewis woke 55 out of 147 nights (37%). Overall the trend of night wakings was decreasing. At follow-up, there was a low stable rate of night waking with only 3 disrupted nights out of 14 nights (21%). Two nights had only 1 night waking while one night had 4 night wakings (252nd night).

Co-sleeping

Figure 7 presents the nights where co-sleeping occurred. The frequency of co-sleeping during baseline was high (7 out of 12 nights). In the early stage of the intervention, there were three nights of co-sleeping recorded (14th, 17th, and 20th nights). Then further co-sleeping on the 97th, 116th, 132nd, and 145th nights. During follow-up co-sleeping was recorded once. The overall trend of co-sleeping was decreasing.

Child Illness and Parental Non-Adherence

Child illness was recorded on two occasions during intervention on the 87th to 93rd night as well as on the 146th and 147th nights. At follow-up, illness was recorded from the 250th to
253rd night (Figures 6 & 7). Parental non-adherence was recorded on 9 nights of the intervention period (Figure 7).

**Visual Analysis**

**Sleep Onset Delay**

Sleep onset delay during baseline and the last 2 weeks of intervention were compared and rated as showing a *substantial improvement*. Baseline and Follow-up sleep onset delay were compared and rated as showing a *substantial improvement*.

**Night Waking**

The frequency of night waking during baseline was compared to the last 2 weeks of intervention and rated as showing a *substantial improvement*. Baseline and follow-up comparisons were rated as showing a *substantial improvement*.

**Co-Sleeping**

Rates of co-sleeping were compared during baseline and the last 2 weeks of intervention and rated as showing a *substantial improvement*. Baseline and follow-up night waking data were also compared and determined as showing a *substantial improvement*.

**DISCUSSION.**

Overall, there were substantial improvements in Lewis’s sleep difficulties. This supports the use of behavioural intervention for multiple sleep problems such as bed refusal/sleep onset delay, night waking, and co-sleeping in a child with MED. With intervention there was an initial increase in sleep onset delay for Lewis. This was due to the shift in his bedtime from between 8.30 and 10.30pm to a bedtime of 7pm as well as to settling without parental presence. The increase in sleep onset delay in the initial stages of the intervention is also likely to be a PERB. Lewis’s parents reported that there were increased levels of crying and tantrum behaviours. Sleep onset delay decreased from the 35th night to low stable levels of 5 to 10 minutes indicating rapid sleep onset from the time of being settled into bed. The follow-up indicated that sleep onset delay was still relatively short in duration, with one exception during illness. The changes in sleep onset delay were rated as showing a *substantial improvement*.

Lewis was waking frequently during the night and these wakings would normally result in co-sleeping in the parents’ bed or a parent sleeping with Lewis in his bed. The frequency of night waking was substantially reduced with intervention. Lewis was waking 0 to 4 night wakings per night during baseline, and this was reduced to 0 to 1 night waking per night by the
end of the intervention and also during follow-up. There were many nights of undisrupted sleep towards the end of intervention as well as during follow-up indicating positive treatment effects. Co-sleeping was also high during baseline with 7 out of 12 nights reported. After intervention the number of nights of co-sleeping were reduced and there was no co-sleeping reported in the last 2 weeks of the intervention phase, and only one reported incident during follow-up. The change in the frequency of co-sleeping with intervention was also rated as a substantial improvement.

The duration of the sleep intervention was 22 weeks for Lewis. The sleep programme was disrupted on one occasion by child illness but markedly disrupted by parental non-adherence. Parental non-adherence was high, with a total of nine incidents throughout the intervention phase. Seven of these nights involved co-sleeping. Parental concern about Lewis, especially because of the uncertainty surrounding MED, may have affected their ability to adhere to the programme recommendations. On the 97th night for example, the mother said that she allowed Lewis to sleep in their bed because he was cold. According to the parents, pain associated with MED is influenced by colder temperatures. Although the parents wanted Lewis to be able to sleep through the night, they often attributed his waking to external factors. The parents felt that these were genuine reasons to wake and so they attended to Lewis when he woke. These parental expectations around sleep behaviour impacted on the progress of the sleep programme.

Overall, there were substantial improvements in Lewis’s sleep behaviours and sleep onset delay, night waking, and co-sleeping were reduced to low levels that were satisfactory to the parents. The parents reported that the sleep programme impacted positively on the daytime behaviour of Lewis.

RESULTS COMMON TO HAMISH AND LEWIS

Social Validity Measures

The programmes for Hamish and Lewis had high social validity. The parents reported being very satisfied with the programme, which met all of their needs. Both parents indicated that the programme was somewhat stressful to implement. Hamish and Lewis’s parents also reported in the evaluation questionnaire that the quality of the help was excellent and that they would definitely recommend the programme to a friend if had a similar problem with a child.

DISCUSSION COMMON TO HAMISH AND LEWIS

Interventions employing a variety of behavioural techniques were successful in eliminating or reducing multiple sleep problems for two children with MED. The duration of the interventions for Hamish and Lewis were long, 20 weeks and 1 day and 22 weeks respectively.
The length of the interventions may have been due to the parents having to intervene with two children’s sleep problems concurrently. Although there were some strategies in common between the interventions, treating two children at once would have been extremely time-consuming, labour intensive, and tiring. An attempt was made to address this issue by delaying the use of strategies for Hamish’s nightmares. It is unclear whether this was beneficial to the parents. The difficulties associated with treating two children at once may have exacerbated parental non-adherence and there were incidents of co-sleeping during the intervention for both Hamish and Lewis.

The parents noted that there were positive changes to Hamish’s schooling and learning and some improvements in Lewis’ daytime behaviour. They reported improvements in their own sleeping as a consequence of improvements in sleep in their children. The parents also said that their stress were reduced as a result of the programme and the mother indicated that she was feeling less irritable and coping better with daily stressors.

Overall, the behavioural interventions were successful in eliminating or reducing sleep problems to low levels of occurrence for both children. The parents indicated high satisfaction with the programme and were pleased with the outcome for both children. The parents further reported that the interventions were only somewhat stressful to implement. Multiple sleep problems were addressed such as bed refusal and sleep onset delay, night waking, co-sleeping, and nightmares and improvements were noted in all of these sleep-related difficulties. The programme supports the use of behavioural interventions with children who have a physical disability.
CHAPTER 6. GENERAL DISCUSSION

SUMMARY OF FINDINGS AND COMPARISON TO THE LITERATURE

The present research aimed to demonstrate the effectiveness of behavioural interventions for the treatment of sleep problems in children with varying disabilities such as cerebral palsy, epilepsy, GDD, and MED. The results of all four case studies support the use of interventions employing behavioural techniques for the treatment of multiple sleep problems such as bed refusal, sleep onset delay, night waking, and co-sleeping. Varying behavioural strategies were employed with each child depending on his or her individual circumstances and the maintaining factors identified in the assessments. Strategies included a positive bedtime routine, parental limit setting, rewards systems, and modified and unmodified extinction. Behavioural intervention was also successful in reducing the occurrence of nightmares in one child. The majority of the sleep behaviours targeted for intervention were eliminated or reduced to low levels of occurrence, with 9 out of 11 target behaviours rated as showing a substantial improvement. Improvements in sleep behaviours were maintained at a 2.5- to 3-month follow-up with the exception of co-sleeping in Case Study Two, which was rated as showing a moderate deterioration.

The behavioural interventions had high social validity. All of the parents indicated that they were very satisfied with the programme. It is important to note that both parents in Case Study One rated the programme as being very stressful while the other parents only rated the programme as somewhat stressful. This may reflect the severity of the child’s sleep problem as well as the level of stress for this particular family. All of the parents indicated that the amount of support they received was helpful. One mother attributed the frequent support to her completion of the programme. Overall, the feedback from the parents was positive, indicating that behavioural interventions are acceptable to families.

These results support the current literature that behavioural techniques are superior to pharmacological intervention (Dahl, 1992; Ramchandani et al., 2000). All four children in this research were unsuccessfully treated with sedative medication for their sleep problems in the past.

The research indicates that children with disabilities are at greater risk of developing sleep problems than typically developing children (e.g., Didden et al., 2002) and are also more susceptible to developing long-term and persistent sleep problems (e.g., Quine, 1991). All four children who participated in this study had had sleep problems since infancy, supporting the argument that sleep problems in this population are less likely to resolve on their own (Quine, 2001). This highlights the need for interventions for children with disabilities and their families.
Successful interventions for sleep difficulties in children with disabilities may have other associated benefits. For example, the parents of Jenny and Hamish reported improvements in behaviour, concentration, learning, and language, attributing these positive changes to the programme. Similarly to studies by Mindell and Durand (1993) and Wiggs and Stores (2001), the parents in this study commented that they were experiencing less stress and feeling more satisfied with their own sleep as a consequence of successful sleep intervention for their children. In addition, Jenny’s parents indicated that there were improvements in family relationships and functioning. This is consistent with Mindell and Durand (1993) who found that there was positive change in parental mood and relationships as a result of successful behavioural intervention for children’s sleep problems. Again these results emphasise the importance of interventions for children with disabilities as their cognitive functioning, behaviour, family functioning and relationships may already be compromised by their disability.

Although this study found behavioural interventions were successful in ameliorating sleep problems, the results indicate that interventions may take longer for children with disabilities. The interventions for children in this study were 6 weeks and 5 days, 17 weeks, 20 weeks and 1 day, and 22 weeks. There are several factors that may have contributed to the duration of the sleep interventions. Firstly, for Jenny and Molly, factors associated with intellectual disability may have extended the length of the interventions. Cognitive and communicative impairments may delay or restrict the acquisition of new sleep behaviours. Johnson (1996) suggested that a child’s ability to learn self-soothing skills necessary for sleep may be affected by intellectual disability and autism. Quine (1991) found that adaptive skill deficits, limited communicative ability, and incontinence in children with learning disability were associated with sleep problems. For Jenny, difficulties with toileting at one point, exacerbated sleep difficulties and limited her mothers’ ability to use planned ignoring during the night.

Secondly, heightened parental concern about the health and wellbeing of their child may have been another factor prolonging the interventions. For Jenny and Molly, their limited ability to communicate needs would have contributed to parental anxiety and attending behaviours. For Hamish and Lewis, parental concern about pain and MED may have underpinned parental responsiveness and permissive handling of their children’s behaviours at night-time. In some cases, the parents’ ability to ignore their children during the night was affected by their perception of their child’s ability to sleep through the night. Medical conditions such as epilepsy and asthma would have also increased parental concern and their need to attend to their child at night. Stores and Wiggs (2001) suggested that parent’s attitudes, parenting ability, and wellbeing,
are influenced by their child’s condition, resulting in some parents becoming overly permissiveness and inconsistent in their parenting behaviours.

Lastly, parental fatigue and stress were cited as major obstacles for parents implementing strategies consistent with the recommendations of the programme. Employing behavioural interventions may be time consuming as well as labour intensive, and parents of children with disabilities may have limited ability to commit to programmes because of their financial and social circumstances, as well as the demands associated with having a child with a disability. This may explain the incidents of parental non-adherence during the interventions and therefore emphasises the need for ongoing support for families throughout the programme.

Practical, ethical, and safety considerations were necessary for the children with disabilities in this study. Allowances were made for children’s special needs, such as learning needs, and also parental anxiety. Wiggs and France (2000) commented that standard extinction might be inappropriate for some children with disabilities because of safety concerns associated with some medical conditions. Graduated and modified methods were presented as more suitable in these circumstances. In addition, graduated strategies may be appropriate for parents who are anxious about starting an intervention or feel unable to completely ignore their child for the entire night. In the case of Molly and Lewis, the parents were hesitant to employ methods (that involved ignoring or a contingency for inappropriate behaviour) that were likely to cause child distress. In both cases, graduated methods were presented to the parents. Although the parents had indicated that they would use the graduated strategies, the parents instead modified the intervention strategies to suit their needs, which suggests that graduated behavioural strategies may also be difficult for parents to implement.

This study emphasises the need to adapt behavioural strategies in order to meet a family’s changing needs and the need to have a variety of strategies available. As discussed in Chapter One, learning to go back to sleep can be viewed as an important developmental milestone (Armstrong et al., 1994) and children must learn to initiate and reinitiate sleep on their own. Sleep incorporates behavioural components and is influenced by environmental and social interaction factors, and from a social learning perspective, the acquisition of appropriate sleep behaviours is vulnerable to learning influences (Blampied & France, 1999).

Children with disabilities and their families may have a heightened vulnerability to these learning influences, and may be more susceptible to falling into coercive behaviour traps like those described by France and Blampied (1999). It is acknowledged that child characteristics such as temperament (e.g. Hayes et al., 2001; Sadeh et al., 1994) and sleep state organisation may also contribute to the development of sleep problems in children. For children with disabilities, the physical characteristics of a disability may increase the likelihood of sleep
fragmentation, such as the abnormal EEG patterns in children with cerebral palsy (e.g. Shibagaki et al., 1985), seizures (Stores, Wiggs, & Campling, 1998), and pain (Lewin & Dahl, 1999).

Although children with disabilities may have a physiological vulnerability towards developing sleep problems, it is important to emphasise that behavioural interventions have been successful in treating sleep problems in children with disabilities (Bartlet & Beaumont, 1998; Didden et al., 1998; Piazza et al., 1997; Weiskop, 2001). A comprehensive assessment is necessary to ensure that there are no underlying medical conditions that may contraindicate behavioural intervention as well as to develop an appropriate intervention plan.

LIMITATIONS OF THE STUDY

There are several methodological limitations with this study. Firstly the experimental design was abandoned and replaced by case studies. This research did not employ an experimental control group and because of practical limitations the multiple baseline design was not used. The study was also limited by a small sample size. Secondly, there were no direct measures of sleep behaviours gathered. The study was based primarily on sleep diaries completed by parents and the reliability of these subjective reports could not be tested with the use of an infrared time-lapse video recording, which was a standard practice of the CSP. This was due to practical and safety concerns associated with older and mobile children in the study. Thirdly, the study included children with varying disabilities requiring very different interventions. The time restraints of the study resulted in the recruitment of children with various disabilities, rather than focusing on specific disability subgroups. This limited formal comparisons between participants.

The parents reported high levels of stress prior to the intervention. It would have been beneficial to have systematically evaluated stress as part of the study to assess the impact of the behavioural treatment. Information relating to improvements in child behaviour, learning, concentration and communication were also based on subjective information, and it may have been appropriate to gather information, pre- and post- intervention, to assess these changes in relation to the interventions.

IMPLICATIONS FOR FUTURE RESEARCH

This current study included children with varying types of disabilities and because of the limited sample size it is unclear whether different disabilities require separate treatment consideration. The literature on children with disabilities emphasises the need for sleep research in specific types of disabilities (Richdale et al., 2000; Stores et al., 1998; Weiskop, 2001; Wiggs & France, 2000). Most research investigating sleep has grouped children with differing
disabilities together and so future research addressing specific subgroups of disabilities is advocated.

Interventions in this study were protracted, and the longest intervention was around 22 weeks. Some parents in this study also reported that the intervention was stressful to implement, and in two cases the parents modified the programme. Research on the treatment of sleep problems in children with disabilities is still limited and more research in the area is required. Research should focus on minimising the stress on the child and family. Interventions that extend over long periods of time may be costly and impractical, so investigating the most effective intervention strategies for children with disabilities would be advantageous.

Further exploration of the underlying mechanisms that contribute to the development of sleep problems in children with disabilities may also be of benefit. Identification of specific initiating and maintaining factors for children with disabilities would be helpful in the development of prevention and intervention programmes. Because children with disabilities are at particular risk of developing persistent sleep problems, there is a need for preventative measures for this population. Future research on prevention programmes for sleep problems may be a cost-effective and practical way of disseminating information to large groups of parents.

IMPLICATIONS FOR PROFESSIONAL PRACTICE

This research supports the use of behavioural strategies for the treatment of sleep problems in children with disabilities and suggests that successful intervention may have associated benefits for the individual and family. The study highlights however, the need for regular contact and support for parents implementing behavioural strategies with their children. Parents of children with disabilities may be under particular stress and pressure and their ability to institute a behavioural intervention may be restricted. Behavioural interventions can be stressful, time consuming and labour intensive, and regular support may prevent parents from withdrawing from the programme. The case studies furthermore emphasise the need to adapt intervention strategies to make allowances for a family’s changing needs. The treatment of sleep problems in children with disabilities may be very complex, and the consideration of individual family circumstances and needs is imperative. This emphasises the importance of a thorough assessment and the development of an appropriate intervention plan. Another implication for professional practice is the focus on preventative measures.

CONCLUSION

The four case studies illustrate that interventions employing a variety of behavioural techniques can successfully eliminate or reduce multiple sleep problems in children with
disabilities. These improvements were maintained at follow-up suggesting the long-term effectiveness of these interventions. The parents were satisfied with the outcome of the interventions and high social validity was reported.
REFERENCES


Stores, G. (2001b). Sleep patterns in the epilepsies. In G. Stores, & L. Wiggs (Eds.) *Sleep*


Appendix A.

University Ethics Committee Approval
19 March 2003

Risa Mizusawa  
Department of Education  
UNIVERSITY OF CANTERBURY

Dear Risa

The Human Ethics Committee advises that your research proposal "Sleep Problems in Children with Developmental Disabilities: a Family Intervention Approach" has been considered and approved.

Yours sincerely

[Signature]

Blossom Hart  
Secretary
Appendix B.

Assessment Questionnaire
Sleep Problems in Children with Disabilities: Family Intervention

Interview

Family Surname:                                  Date of Initial Interview:
Address:                                        Phone Number:

Referral:                                       Family Doctor Consulted: Y / N

Household Composition
Adults: Names: Age: Occupation:

Children:

Birth Order: 1 2 3 or subsequent

Significant Others- regarding child minding

Child having sleep problem: D, O, B.
Actual bedtime: Ideal bedtime:
Actual settling time: Ideal settling time:
Actual getting up time: Ideal getting up time:

Diary completed:
Number of days:

Average night waking over whole sleep time:

How does your child act when he/she wakes up during the night?

What do you do when he/she wakes up at night?

What seems to work the best?

Daytime sleep? Y / N Time of sleep and duration:
Current bedtime routine: (e.g. reading book, brushing teeth)

Does he/she share a room: Y / N Who with:
Does the child go to be on his/her own, or does he/she need to be rocked, patted or given some other kind of help from you?

Do you think that he/she is tired enough at bedtime to go to sleep? Why/why not?

**Describe the nature of the sleep problem:**

How is the sleep problem affecting other family members (parents/siblings)?

**Sleep History:**
Birth – 6 months:

6 months – 12 months:

12 months – 2 years

2 years – 5 years

5 years - current

**Clear precipitating event Y / N**
**Describe**

Have you noticed any changes in daytime behaviour since the development of the sleep problem?

**Child’s Developmental History:**
**Pregnancy:**

**Birth:**
Birth weight:                   Gestation age:                   Method of delivery:
Any Complications:

**Infancy:**
Any feeding problems:
Activity:
Crying:
Achievement of Milestones: e.g.  Crawling
                                    Sitting unassisted
                                    Walk alone
                                    Babbling
                                    First words
Early Childhood:
Were there any other special problems in the growth and development of your child? Y / N

Significant events over the child’s life (e.g. hospitalisation, bereavement):

Medical History and Current Health
Describe your child’s special needs

What is he/she like?

Strengths

Challenges

History of developmental problem(s)

Does your child have any other medical conditions? Y / N

What sources of support does the family have: (e.g. family, friends, GP, paediatrician, psychologist)

Is your child receiving any other treatment (e.g. medication, physical therapy, speech therapy)?

Current Health and Development:
Child’s Height:       (cm)       Child’s Weight:     (kg)

Tell me about your child’s feeding:

Where are you up to with toilet training?

Tell me about your child’s communication:
How well can he/she get about?

Does your child have needs that are not currently being met?

Other problems:    Child:   Y / N
                    Family:  Y / N

Describe:

Medication:
Has medication been used in the past:   Y / N
Name of medication  Type of medication  Side effects  Reason for discontinuing

Is medication used now:     Y / N
Name of medication  Type of medication  Side effects

Treatment of sleep problem
What has been done to handle the child’s sleep problem?

Previous programmes:

Suggested Programme

Rationale:
Appendix C.

Sleep Diaries
<table>
<thead>
<tr>
<th>Child's Name:</th>
<th>Recorded By:</th>
<th>Condition:</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>DAY SLEEP</strong></td>
<td>Date:</td>
<td></td>
</tr>
<tr>
<td>Time asleep</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&amp; duration</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>NIGHT SLEEP</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time to bed</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Behaviour</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time asleep</td>
<td></td>
<td></td>
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<tr>
<td>Night waking/s</td>
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<tr>
<td>1. Time</td>
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<tr>
<td>Duration</td>
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<tr>
<td>Behaviour</td>
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<td>What did you do?</td>
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<td>2. Time</td>
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<td>Behaviour</td>
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<td>Behaviour</td>
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<td>What did you do?</td>
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<td>4. Time</td>
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<td>Duration</td>
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<td>5. Time</td>
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<td>Duration</td>
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<td>What did you do?</td>
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<td>What did you do?</td>
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<td>7. Time</td>
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<td>Duration</td>
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<td>What did you do?</td>
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<tr>
<td><strong>Time awake</strong></td>
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**KEY** (for example)
c = crying
a = assisted to bed
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<tr>
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<td>(1½hrs)</td>
<td>2:30</td>
<td>7.45</td>
<td>7.30</td>
<td>7.15</td>
<td>7.50</td>
<td>7.35</td>
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<td>grizzly</td>
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<td>C</td>
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<td>7:45</td>
<td>7:50</td>
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<td>7:55</td>
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<td>4:00</td>
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<td>30mins</td>
<td>30mins</td>
<td>20mins</td>
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<tr>
<td>What did you do?</td>
<td>A</td>
<td>P</td>
<td>A</td>
<td>gave cuddles</td>
<td>P</td>
<td>A</td>
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<tr>
<td>What did you do?</td>
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<td>slept in cots bed.</td>
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<td>5:30</td>
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<td>Time awake</td>
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</tbody>
</table>

**KEY (for example)**
- c = crying
- a = assisted to bed
- P = sleep in Parents' bed.
Appendix D.

Programme Evaluation Questionnaire
Sleep problems in Children with Disabilities: Family Intervention

Evaluation Questionnaire

PLEASE CIRCLE YOUR ANSWER

1. How would you rate the quality of the help you received?
   1  2  3  4
   poor   fair   good   excellent

2. Did you get the kind of help (or service) you wanted?
   1  2  3  4
   no, definitely not   no, not really   yes, generally   yes, definitely

3. To what extent has the sleep programme met your needs?
   1  2  3  4
   none of my needs have been met   only a few of my needs have been met   most of my needs have been met   almost all of my needs have been met

4. If a friend were in need of similar help, would you recommend the programme to him/her?
   1  2  3  4
   no, definitely not   no, I don’t think so   yes, I think so   yes, definitely

5. How satisfied are you with the amount of help you received?
   1  2  3  4
   quite dissatisfied   indifferent or mildly dissatisfied   mostly satisfied   very satisfied

6. Have the services you received helped you to deal more effectively with other difficult child behaviour?
   1  2  3  4
   no, they seem to make things worse   no, they really didn’t help   yes, they helped somewhat   yes, they helped a great deal

7. In an overall, general sense – how satisfied are you with the service you received?
   1  2  3  4
   quite satisfied   indifferent or mildly satisfied   mostly satisfied   very satisfied
8. If your child had sleeping difficulties again, would you come back to the Canterbury Sleep Programme?

1  2  3  4
no, definitely not  no, I don't think so  yes, I think so  yes, definitely

9. How stressful did you find the sleep programme?

1  2  3  4
very stressful  moderately stressful  somewhat stressful  non-stressful

COULD YOU PLEASE COMMENT ON THE FOLLOWING ASPECTS

1. The instructions explaining the programme procedure.
   (E.g. where they logical, ambiguous, hard or easy to follow, confusing etc.)

________________________________________________________________________
________________________________________________________________________
________________________________________________________________________

2. The method of teaching your child to sleep alone.
   (E.g. was too slow, too difficult to follow, seemed rational etc.)

________________________________________________________________________
________________________________________________________________________
________________________________________________________________________

3. The telephone support given by the therapist.
   (E.g. was too frequent, not often enough, helpful, or not etc.)

________________________________________________________________________
________________________________________________________________________
________________________________________________________________________

4. Keeping daily records of the sleep pattern of your child.
   (E.g. was too tedious, useful, etc.)

________________________________________________________________________
________________________________________________________________________
________________________________________________________________________

5. Any other criticisms or comments

________________________________________________________________________
________________________________________________________________________
________________________________________________________________________
Appendix E.

Parental Information Sheet
Sleep problems in Children with Disabilities: Family Intervention

A guide for parents:

My name is Risa Mizusawa and I am a postgraduate student at the University of Canterbury, currently training to become a child and family psychologist. As part of the requirements for a Master of Education (in Child and Family Psychology) I am undertaking a research project that involves helping children and their families with persistent sleep problems. I have chosen to focus my research on children (aged 2-12 years) with developmental disabilities.

My supervisor Dr. Karyn France is the principal investigator of the Canterbury Sleep Programme. If you have any questions you may contact her during working hours on or after hours on . My second supervisor Dr. Garry Hornby, can also be contacted if you have any questions. His contact number during working hours is ext. . However, you may contact me on or .

The study will use approaches that have been demonstrated to reduce sleep problems in children with disabilities. Furthermore, the approaches that will be used aim to minimise child and parental distress. The intervention will be tailored to each individual family, and will address your child’s special needs.

The purpose and aim of the study:
The purpose of this particular study is to determine what treatment works for children with disabilities with persistent sleep problems. The study aims to reduce sleep problems, such as settling and night waking, by implementing individualised treatment plans. Treatment options will be discussed with you, and you will be free to choose whether to implement a treatment. Parents will receive support during this time and I will ensure regular contact by phone and home visits.

What the study involves for you and your child:
You will attend an initial assessment interview (most likely to be conducted in your own home), so I can begin to understand the nature of your child’s sleep problems. This usually takes around 1 hour. This may be videotaped or audiotaped so if needed, it can be discussed with my supervisors.

I will regularly meet with you and your child over a period of several weeks to formulate a plan. The plan will be developed in consultation with my supervisor. You may also be asked to discuss the treatment with your family GP. I will also see you at a later date to evaluate how things are going for you, your child and your family.

You will be asked to keep a sleep diary. This involves keeping a written record of your child’s behaviour around bedtime, any night waking, and what you did about
this. You will be given materials to use for this purpose. Generally, you will keep this every night until the sleep problems are resolved.

You may also be asked to have a video recorder set up close to the child at night (inside the bedroom or in the hallway) if appropriate, for several nights. This allows us to record your child’s sleep behaviour directly. However, if a video is not suitable for your situation then, 2 people (yourself and partner, or another family member) will be requested to record sleep diaries on the same night for approximately 6 nights for purposes of the study. I will discuss this with you.

You will be asked to fill in a short questionnaire about your participation at the end of the study.

Any information that you provide is confidential to the study. The results of the study with regard to your child will be discussed with you. The information will be written up, your details combined with the results of other peoples’ treatment. Academic papers submitted for a Master of Education may be written up for publication in a professional journal. Your confidentiality will be maintained, and pseudonym names will be used.

The audiotape of the first interview, and the videotape (if any) of your child’s sleep, will not be used except by my supervisors and myself. However, any safety issues arising will be handled according to professional ethics.

The contact persons for this programme are:

Risa Mizusawa
Home:
Cell:
Signature:

Dr. Karyn France
Work:
A/H:
Signature:
Appendix F.

Consent Form
Sleep problems in Children with Disabilities: Family Intervention

Consent Form:

I/we have read the information sheet and understand the description and purpose of Risa’s research. On the basis of this, I agree that my child and myself/s will participate in the study.

I am aware that this study is part of a Master of Education degree at the University of Canterbury. I consent to Risa writing up her findings and submitting a report, with the understanding that anonymity will be preserved. I understand that the report will be submitted to the University of Canterbury and will be lodged in the University’s library.

I give consent to the interview being videotaped or audiotaped, as well as to the use of a video recorder to record my child’s sleep behaviour.

I understand that we are free to withdraw, with our child, from the study at any point, including withdrawal of any information I have provided.

Child’s Name: ______________________

Parent’s Name: _____________________

Parent’s Signature: ________________ Date: ______________

Address: __________________________

_______________________________

Phone No: ________________________
Appendix G. Criteria for the 5-Point Scale Used for Visual Analysis

1. **SUBSTANTIAL IMPROVEMENT:**
   Data showing that following intervention there was an elimination of the undesirable behaviour, or reduction to a very low level of occurrence.

2. **MODERATE IMPROVEMENT:**
   Data showing that following intervention there was a clear reduction in the behaviour, but not sufficient to be considered substantial.

3. **NO CHANGE:**
   Data showing that following intervention there was no change in behaviour.

4. **MODERATE DETERIORATION:**
   Data showing that following intervention there was a clear increase in the undesirable behaviour, but not sufficient to be considered substantial.

5. **SUBSTANTIAL DETERIORATION:**
   Data showing that following intervention the undesirable behaviour was occurring 100% of the time or increased to a very high level of occurrence.
Appendix H. Information Sheet on the Sleep Programme: Jenny

Preparing the Room for the Sleep Programme
It is important that we prepare Jenny’s bedroom so that it is less stimulating for her and safe.
Removing toys from the bedroom and unnecessary furniture.
Securing the wardrobe.
Securing her bedroom door.

Jenny may like a nightlight in the bedroom. This may need to be put in a place where she cannot reach so she is less likely to play with it.

Child Safety
Your child’s safety is most important. Stop the programme if your child is ill, or in danger of hurting herself.

Bedtime Routine
Establish a regular bedtime routine with Jenny. For example, getting changed for bed, brushing teeth, “quiet time” such as a bedtime story.
Remind Jenny that she will receive a reward (a stamp on her hand) if she stays in her bed all night. Emphasise that “daytime is for cuddles and play, night-time is for sleeping”

Bedtime / Wake time
Try to keep to the scheduled bedtime and wake times as much as possible.

<table>
<thead>
<tr>
<th>Negotiated Bedtime:</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Negotiated Wake Time:</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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</table>

The Parental Presence Approach Combined with Trimeprazine
- It is important that you think through and decide how you are going to deal with Jenny’s crying when you are in the same room as her prior to starting the programme.
- **Trimeprazine:** Give Jenny her medication at bedtime. Give the dose that was prescribed by your doctor for that night. Give the full dose of trimeprazine for the first 2 nights and decrease the dosage by 1/5 every second night of the programme, or as recommended by your doctor.
- Say good night to your child and lie in the other bed in sight of Jenny.
- If your Jenny cries, pretend to be asleep (do not respond to her).
- Remain in the bed until she falls asleep.
- Once your child is asleep, you may leave the room.
- If your child awakens before your own bedtime enter her room and lie down in the other bed and pretend to be asleep.
- Make sure your child sees you entering the room (i.e., give a little cough to indicate your presence). Continue sleeping there until your child falls asleep, or for the rest of the night after your bedtime.
- If Jenny awakens during the night, make sure she is aware of your presence (i.e. give another cough).
- If you must attend to your child, do so with minimal contact. It is important not to make a big fuss.
- Follow this routine (parental presence) for approximately the first 7 nights of the programme.
- After 7 nights of the parental presence, return to your normal sleeping arrangements. Completely ignore your child’s inappropriate sleep behaviour (unless she is ill or in danger). Ensure that you remain within hearing.

I will make contact with you each day to find out how you are going and answer any questions or concerns you may have.

Contact Details:

Risa Mizusawa  
Ph Home

Dr Karyn France  
Ph Home
Appendix I.

Letter to the Family GP: Jenny
February 2003

Dr.
Christchurch

Re:
DOB:

Dear Dr.

X and her parents were referred to the Canterbury Sleep Programme. X presents with a history of persistent night-time sleep disturbance. Attempts at alleviating the problem in the past have shown that he is likely to be resistant to intervention. In these cases, we like to work with the family’s GP. We have found the most appropriate treatment is a combination of a “parental presence” programme in conjunction with trimiprazine given in decremental doses over the first 10 days of the programme. A full dose is given in the first night and decreased (by 1/5th) every second night until there is no medication on the 11th night. This usually reduces the amount of crying the parent has to handle. In conjunction, the parent will remain in the room for the first week of the programme feigning sleep while X is crying. This approach also decreases the amount of crying.

Recent discussions with General Practitioners and experience with children indicates that the 30mg/5mls dosage used in the France, Blampied and Wilkinson (1991) article results in some children being too heavily sedated. The idea is for X to continue to awaken so learning will occur but to be sedated enough that protracted crying and anxiety is avoided.

We have suggested to X’s parents that they organise an appointment to see you to confirm that X is medically fit for the programme and to ask you if you think X is suitable for the trimiprazine/parental presence programme. If you do, we would appreciate you prescribing the medication at the level you think is most appropriate.

The original article is enclosed for your information. In this case, Risa Mizusawa, a 5th year Child and Family Psychology student will be working with Mr. and Mrs. X to carry out the programme under my supervision. Please feel free to contact us should you have any queries.

Yours sincerely

Senior Lecturer.
Principal Investigator Canterbury Sleep Programme
Appendix J. Information Sheet on the Sleep Programme: Molly

Preparing the Room for the Sleep Programme
It is important that we prepare Molly's bedroom so that it is less stimulating for her and safe.
For example:
Removing toys from the bedroom.
Securing windows so that she is unable to open them.
Securing her bedroom door.

Sometimes a child may like a nightlight in the bedroom. This may need to be put in a place where she cannot reach so she is less likely to play with it.

If you are concerned about your child's safety, you may wish to use a “baby monitor” or intercom system to listen to your child at night. This should also be placed away from the child’s reach.

Child Safety
Your child’s safety is most important. Stop the programme if your child is ill, or in danger of hurting herself.

Bedtime Routine
Establish a regular bedtime routine with your child. For example, getting changed for bed, brushing teeth, securing windows, bedtime story.
Use prompts such as:
“Molly, time for bed” “let’s get ready for bed” “night-time is for sleeping”

Bedtime / Wake time
We recommend setting Molly's bedtime to a later time until we are further into the programme. This will reduce difficulties with settling initially. Bedtime will be gradually set earlier as we progress with the programme.

<table>
<thead>
<tr>
<th>Negotiated Bedtime:</th>
</tr>
</thead>
<tbody>
<tr>
<td>Negotiated Wake Time:</td>
</tr>
</tbody>
</table>
Sleep Programme Options

1) Standard Extinction
Put Molly to sleep, and secure the bedroom door behind you. Do not attend to your child until the morning, at the negotiated time. Ignore any behaviour such as banging on the door or crying.

Regularly check your child for safety by listening through the door, and/or if you can observe her from outside the house through the window. Do not open her door to check on her.

Only attend to your child if she is ill or in danger of hurting herself.

2) Graduated Extinction
Put Molly to sleep and secure the bedroom door behind you. If she wakes attend to her after the appropriate period of time. When you attend to your child do so with minimal contact. For example, assist her back to bed, but do not talk or cuddle her. Leave the room and secure the door behind you.

<table>
<thead>
<tr>
<th>Night #1</th>
<th>Night #2</th>
<th>Night #3</th>
</tr>
</thead>
<tbody>
<tr>
<td>1st time</td>
<td>1st time</td>
<td>1st time</td>
</tr>
<tr>
<td>5 mins</td>
<td>10 mins</td>
<td>15 mins</td>
</tr>
<tr>
<td>2nd time</td>
<td>2nd time</td>
<td>2nd time</td>
</tr>
<tr>
<td>10 mins</td>
<td>15 mins</td>
<td>20 mins</td>
</tr>
<tr>
<td>3rd time</td>
<td>3rd time</td>
<td>3rd time</td>
</tr>
<tr>
<td>15 mins</td>
<td>20 mins</td>
<td>25 mins</td>
</tr>
<tr>
<td>4th time</td>
<td>4th time</td>
<td>4th time</td>
</tr>
<tr>
<td>20 mins</td>
<td>25 mins</td>
<td>30 mins</td>
</tr>
<tr>
<td>5th time</td>
<td>5th time</td>
<td>5th time</td>
</tr>
<tr>
<td>25 mins</td>
<td>30 mins</td>
<td>30 mins</td>
</tr>
</tbody>
</table>

4th Night start with 20 minutes, 5th night start with 25mins etc.

Maximum time before attending: 30minutes

Attend to your child at any time if she is ill or in danger of hurting herself

I will make contact with you each day to find out how you are going and answer any questions or concerns you may have.

Contact Details:

Risa Mizusawa
Ph Home:

Dr Karyn France
Ph Home:
Appendix K. Information Sheet on the Sleep Programme:
Hamish and Lewis

Reward System:
e.g. 15 mins of individual time doing anything they want with mum or dad
    Choosing their meal for dinner
    Playing a board game
    Reward box - full of inexpensive fun toys and stickers

Star Charts:
Make a star chart for each child together as a family, and place a star or sticker on
the chart in the morning, to reward sleeping through the night.
Lewis may like a sticker or stamp on his hand as well as on his star chart

Cuddles before bed:
Set aside “15 mins of cuddles” for Hamish and Lewis before their bedtimes. It is
good to turn the TV off and cuddle with them on the couch or engage in a quiet
activity before going to bed.

Bedtimes/wake times:
Bedtimes should reflect child’s age.

<table>
<thead>
<tr>
<th>Lewis’s bedtime:</th>
<th>Lewis’s wake time:</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hamish’s bedtime:</td>
<td>Hamish’s wake time:</td>
</tr>
<tr>
<td>Jamie’s bedtime:</td>
<td>Jamie’s wake time:</td>
</tr>
</tbody>
</table>

Descriptive Praise:
Instead of saying just “good boy”, say things like “Hamish, I really like the way you
are going to bed like I asked”

Clear Instructions:
Keep a calm voice, go down to child’s level, and make sure you have eye contact:
“Lewis, its 7.30pm, its your bedtime now. Get into bed now please”
Give the child a few seconds to respond to instructions
If he does not do as asked take him to bed, with minimal fuss, and remain calm.

Lewis:
• 15 minutes of cuddles before bedtime
• Bedtime routine: eg brush teeth, get changed, story
• Give bedtime instructions “Lewis, its your bedtime now, go to bed please”
• Take Lewis to his room, and put him in bed
• If he comes out of his room: Take him to his room without talking and hold his
door
Hamish:
  • Work towards a “fear busting and monster taming certificate”

I will make contact with you each day to find out how you are going and answer any questions or concerns you may have.

Contact Details:
Risa Mizusawa
Ph Home:

Dr Karyn France
Ph Home:
Appendix L. Summary of the Case Study Results

A summary of the results for all of the participants in the study is given. The visual analysis results for the target behaviours are summarised in Table 3 for all four children. The parent ratings on the Programme Evaluation Questionnaire are shown in Table 4 to illustrate the social validity of the programme. Reliability of the sleep measures and the reliability of the visual analysis are also reported (Table 5).

**Table 3: Summary of the visual analysis for all of the participants**

<table>
<thead>
<tr>
<th>Child</th>
<th>Sleep Onset Delay</th>
<th>Night Waking</th>
<th>Co-Sleeping</th>
<th>Nightmares</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>BL vs. INT</td>
<td>BL vs. FU</td>
<td>BL vs. INT</td>
<td>BL vs. FU</td>
</tr>
<tr>
<td>Jenny</td>
<td>--</td>
<td>--</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Molly</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Hamish</td>
<td>--</td>
<td>--</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Lewis</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

Visual analysis using a 5-point scale (Weiskop, 2001) where 1= substantial improvement, 2= moderate improvement, 3= no change, 4= moderate deterioration, and 5= substantial deterioration; (refer to Appendix G for the criteria). Where, BL= baseline, INT= the last 2 weeks of intervention, and FU= Follow-up.

**Table 4: Summary of the social validity for all of the participants**

<table>
<thead>
<tr>
<th>Qual. of Help</th>
<th>Meet Parents’ Needs</th>
<th>Gen. Satisfaction</th>
<th>Stressfulness</th>
<th>Recommend</th>
</tr>
</thead>
<tbody>
<tr>
<td>1= poor</td>
<td>1= none of needs met</td>
<td>1= not satisfied</td>
<td>1= very stressful</td>
<td>1= definitely not</td>
</tr>
<tr>
<td>4= excellent</td>
<td>4= all of needs met</td>
<td>4= very satisfied</td>
<td>4= non stressful</td>
<td>4= yes definitely</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Jenny</th>
<th>4</th>
<th>3</th>
<th>4</th>
<th>1</th>
<th>4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mother</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Father</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Molly</th>
<th>4</th>
<th>3</th>
<th>3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mother</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Hamish &amp; Lewis</th>
<th>4</th>
<th>3</th>
<th>4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mother</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Father</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Where, Qual. of Help= quality of help received, Meet Parents’ Needs= the extent the programme met the parents’ needs, Gen. Satisfaction= general satisfaction with the programme, Stressfulness= how stressful the programme was to implement, and Recommend= whether the parents’ would recommend the programme to a friend.
Table 5. Summary of the reliability of parental sleep diaries and visual analysis for all of the participants

<table>
<thead>
<tr>
<th>Child</th>
<th>Reliability of the Sleep Diaries</th>
<th>Reliability of the Visual Analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Father's diary</td>
<td>Researcher's notes</td>
</tr>
<tr>
<td>Jenny</td>
<td>69%</td>
<td>99%</td>
</tr>
<tr>
<td>Molly</td>
<td>--</td>
<td>84%</td>
</tr>
<tr>
<td>Hamish</td>
<td>100%</td>
<td>97%</td>
</tr>
<tr>
<td>Lewis</td>
<td>100%</td>
<td>98%</td>
</tr>
</tbody>
</table>

Visual Analysis

See Table 3. Baseline and the last 2 weeks of intervention were compared and the majority of target behaviours (9 out of 11) for the children were rated as showing a substantial improvement. Comparisons between baseline and follow-up also rated these behaviours as showing a substantial improvement.

There were two exceptions. For Molly, the comparison between baseline and the last 2 weeks of intervention for co-sleeping was rated as showing no change. When baseline and follow-up was compared, the rating moderate deterioration was given. For Hamish, the occurrence of nightmares between baseline and the last 2 weeks of intervention was rated as showing no change. The comparison between baseline and follow-up however, was rated as showing a substantial improvement.

Social Validity

See Table 4. All five parents in the study reported that the quality of help they received was excellent and that they were generally very satisfied with the programme. Four parents out of five parents reported that all of their needs were met with programme, while one parent indicated that most of her needs were met. Two parents commented that the programme was very stressful, while the other three parents felt that it was somewhat stressful. Four parents indicated that they would definitely recommend the programme to a friend if they had a similar problem with a child and one parent indicated that she would probably recommend the programme to a friend.
Reliability

See Table 5. Reliability between the mothers’ and fathers’ sleep diaries was variable. For Jenny, the reliability between these diaries was 69%. For Hamish and Lewis the reliability was calculated at 100%. Reliability between mothers’ sleep diaries and notes taken by the researcher during daily telephone calls was high, between 84 and 99%. The reliability between co-raters in the visual analysis was high (100%).