Sleep patterns and creativity in children and adolescents with and without high functioning autism (HFA): a descriptive study and an intervention trial

Claudia Cipolla (2010)

Note if anything has been removed from thesis:

Figs 3.3 p.91, 3.4 p.92, 3.5 p.94, 3.6 p.96. Appendices 4,6,7,12,13,14,15,16,17.

Sleep patterns and creativity in children and adolescents with and without high functioning autism (HFA): a descriptive study and an intervention trial

A thesis submitted in partial fulfilment of requirements for the degree of Doctor of Philosophy

Claudia Cipolla

Oxford Brookes University

Thesis submitted February 2010

THE FOLLOWING CONTENT HAS NOT BEEN SCANNED UNDER INSTRUCTION FROM THE UNIVERSITY:

FIGURE 3.3 (PAGE 91)

FIGURE 3.4 (PAGE 92)

FIGURE 3.5 (PAGE 94)

FIGURE 3.6 (PAGE 96)

APPENDICES FOUR, SIX, SEVE, TWELVE, THIRTEEN, FOURTEEN, FIFTEEN, SIXTEEN AND SEVENTEEN

This thesis is dedicated to my father, Giuseppe Cipolla.

ACKNOWLEDGEMENTS

I would like to say thank you to my supervisors Luci Wiggs and Morag McLean. I have greatly appreciated the support and guidance they have given me throughout this degree.

I also would like to express my gratitude to all children who participated in this research.

Finally to my partner Luis for his reassurance, optimism and great sense of humour!

LIST OF CONTENTS

LIST OF TABLES AND FIGURES

Chapter Four

Chapter Five

ABSTRACT

Sleeplessness is common in childhood especially in some clinical groups and it has negative effects on child and family functioning. There is limited data suggesting a link between lack of sleep and impaired creativity and more studies are needed especially with children with autism for whom creativity is compromised and risk of sleeplessness is high.

The adverse affects on children's neurobehavioral functioning caused by insufficient sleep might be reversed by removal of the sleep disturbance as a means of improving overall function of the child. The treatment of choice for childhood sleeplessness problems are behavioural approaches however they have not been sufficiently evaluated with school age typically developing children (TC) or with children with high functioning autism (HFA) and further, parents often have limited access to such treatments.

Therefore this study aimed to explore the relationship between sleep and neurobehavioral functioning and creativity of children (with and without HFA) and maternal mental health by means of a descriptive study (n=65) and to evaluate the efficacy of booklet behavioural intervention for sleeplessness problems, via a multiple baseline design (n=9), and its indirect effects on children's creativity.

Sleep was assessed by parent and child sleep reports as well as by actigraphy. Children's creativity was measured using the Torrance Test of Creative Thinking and newly developed tests of creativity. Measures of child neurobehavioral functioning and parental mental health were also employed.

The results suggested that sleeplessness was associated with impaired child neurobehavioral functioning and maternal mental health and with reduced creativity in TC only. The booklet-based behavioural intervention appeared to be effective for the treatment of sleeplessness in TC although no associated changes in creativity were consistently found. The booklet was less useful for children with HFA.

The thesis argues that sleeplessness impairs high level cognitive ability in children and affects maternal mental health. The results are clinically useful as they support the use of booklet-based behavioural interventions for sleeplessness in school age TC. The efficacy of such approach with children with HFA needs to be further explored. Nocturnal mental over-activity appeared responsible for the maintenance of sleeplessness in TC and HFA. This has important clinical implications when considering appropriate intervention approaches for this age group.

INTRODUCTION

Sleep is a reversible state of reduced awareness of and responsiveness to both internal and external stimuli in which persons appear quiescent, may shut their eyes, and lie down with little movement (Shneerson, 2005; Stores, 2001a). This reduced awareness which occurs during sleep has in the past misled people to conceive sleep as a passive phase in which the human brain is turned off, in contrast to the active state of wakefulness. Examples of 17th Century literature demonstrate that writers and poets shared the belief that sleep was a peaceful state similar to death, where mental activity was almost absent. This is illustrated by Cervantes' concept of sleep as "the cloak that covers all the human thoughts"; which resembles Shakespeare's "sleep that knits up the ravelled sleeves of care" (Iranzo, Santamaria, & de Riquer, 2004).

The turning point in sleep research arrived in the 1950's with electrophysiological studies which demonstrated brain activity during sleep and swept away previous old-fashioned beliefs. In this short period of time researchers discovered that sleep is a dynamic behaviour, not simply the absence of waking, but a special activity of the brain, controlled by elaborate and precise mechanisms (Hobson, 1989).

Sleep occupies roughly one third of our total life and is proportionately much more of a child's life. In fact, a child of school age spends more time asleep than engaging in any other activities, including playing, eating and interacting with others (Mindell & Owens, 2003). Given the large need to sleep in adults and even greater need in children, researchers suppose that sleep must serve some very important function for our wellbeing, although the simple question "why do we sleep?" is still unanswered. The various theories about the function of sleep have emphasised physical and psychological restoration, energy conservation, consolidation of memories, discharge of emotions, brain growth and other biological functions including the maintenance of the immune system (Stores & Wiggs, 2001a). No one theory is adequate across all species and stages of development and sleep is likely to serve multiple purposes (Rechtschaffen, 1998).

Researchers have tried to determine the function of sleep by examining experimentally the consequences of lack of sleep. A wide range of clinical and observational data supports the theory that loss of sleep seriously compromises cognitive functioning, motor ability and mood in adults (Allen, 2003; Bonnet, 2005; Horne & Pettitt, 1985; Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005; Limoges, 2005; Pilcher & Huffcutt, 1996) but to what extent these results can be applied to children remains unclear. A number of studies have suggested that small reductions of sleep alter neurobehavioural functioning (NBF) in children and memory, attention, vigilance, and reaction time seem to be negatively affected by lack of sleep (Allen, 2003; Dahl, 1996; Randazzo, Muehlbach, Schweitzer, & Walsh, 1998; Sadeh, Gruber, & Raviv, 2003). Furthermore, small increases in the amount of sleep (from as little as 30 minutes) seem to improve children's sustained attention and memory function (Allen, 2003; Sadeh et al., 2003). These results emphasise the fact that for an individual child small variations in sleep duration, which are more likely to occur in real life than total sleep deprivation, could have a clear impact on his or her NBF. Only one study failed to find any effect of restricted sleep on children's daytime functioning with the effects only becoming apparent following total sleep deprivation (Carskadon & Dement, 1981a; Carskadon, Harvey, & Dement, 1981c).

Despite the general consensus regarding the consequence of sleep loss on many aspects of psychological performance, its impact on creative thinking has not been well established. Creativity means different things to different people depending on the theoretical model used to define it. Creativity has been defined as divergent thinking (Guilford, 1950; Torrance, 1962), as a characteristic of the personality (Barron & Harrington, 1981), as a property of the cognitive processing (Runco & Sakamoto, 1999) and as an ability to solve problems (Gardner, 1993).

With regard to the relationship between lack of sleep and the consequences this has on the creative process, few investigations have been carried out. Two studies showed that total sleep deprivation (TSD) had a substantial negative effect on most aspects of divergent thinking in adults (e.g. "originality" and the generation of unusual ideas, and "fluency" in producing numerous ideas) (Horne, 1993) and only one experimental study with typically developing children (TC) suggested that high cognitive functions, such as verbal creativity, abstract thinking and concept formation, became impaired after a single night of restricted sleep (Randazzo et al., 1998). Moreover specific neuropsychological assessments have showed that tasks requiring prefrontal cortex functions (PFC) such as complex, divergent or creative thinking (especially verbal creativity) are far more affected by sleep deprivation than tasks requiring convergent thinking in children and adults (Dahl, 1996; Horne, 1993; Randazzo et al., 1998). The results found in these studies suggest that those PFC functions which have been referred to as "executive functions" (defined as a set of heterogeneous high mental abilities necessary for creative and adaptive learning), may be impaired in children suffering from sleep loss. All of these studies consider the effect of sleep loss on creativity but none of them investigate the opposing relationship. Only one recent study exists on the influence of creative talents on sleep patterns. The results of this study show that highly creative children have significantly more sleep disturbance than control children. Although these results are in the opposite direction, a relationship may exist that suggests creative ability may be associated with an individual's sleep patterns (Healey & Runco, 2006). This relationship is particularly true for many psychiatric or developmental disorders such as Bipolar Disorder, Attention Deficit Hyperactive Disorder, and Autism, which are all associated with potential sleep problems (Harvey, Mullin, & Hinshaw, 2006; Hoshino, Watanabe, Yashima, Kaneko, & Kumashiro, 1984a; Wiggs, 2001) and have an evident association with creativity (Cramond, 1994; Fitzgerald, 2004; Jamison, 1994).

Impoverished imaginative creativity has been referred to as a key symptom in the standard diagnostic criteria for autism (APA, 1994) and has been documented in a number of studies (Craig & Baron-Cohen, 1999, 2000; Craig, Baron-Cohen, & Scott, 2001; Scott & Baron-Cohen, 1996). Craig and Baron-Cohen (1999) found a deficit in children with high functioning autism (HFA) and Asperger syndrome (AS) using a standardised measure of creativity (the Torrance Test of Creative Thinking) (Torrance, 1974). This result supported the view that impairment in executive functions can be disastrous for the ability to generate novel responses. The executive dysfunction theory (Leevers & Harris, 1998) explains this lack of imaginative creativity found in children with autism, as an indication of executive problems in disengaging from routine actions and from reality, and predicted impairment both on imaginative and reality based creativity.

The imagination deficit theory (Scott & Baron-Cohen, 1996), in contrast, argues that children with autism are impaired only on imaginative creativity. Studies in which the Karmiloff-Smith technique of asking children to draw "impossible" people or houses were used, showed that children with autism were significantly worse than matched controls in their ability to introduce "unreal" changes to their representations of people or houses (Craig & Baron-Cohen, 1999; Scott & Baron-Cohen, 1996). This result was only evident in the AS group when they were required to use their imaginative creativity spontaneously (Craig et al., 2001). In line with these findings, children with autism and AS introduced fewer imaginary than reality-based elements in an experimental condition in which a verbal task was used (Craig & Baron-Cohen, 2000). Only one study failed to find an imaginative deficit in children with autism, arguing that the previously reported incapacity to draw impossible pictures might result from a misunderstanding of the task or limitations in the executive abilities required to draw an unusual picture for the first time (Leevers & Harris, 1998). The theory best explaining the role of the imagination deficit in the impoverished creativity observed in HFA and AS sufferers needs to be established. It is important then to question what is causing the abnormalities in the functioning of the imaginative creativity in children with autism. Is there any connection with sleep?

The limited research available suggests that lack of sleep impairs creative thinking in TC (Dahl, 1996; Randazzo et al., 1998) and no previous studies have directly assessed how this loss of sleep is related to imaginative creativity in children with HFA. Lack of sleep or sleeplessness (i.e. difficulty getting to sleep, problems staying asleep and waking too early in the morning) is common in TC and adolescents, as about 30% are affected (Kahn et al., 1989; Manni et al., 1997; Morrison, McGee, & Stanton, 1992) and contrary to popular belief, is not a transient problem which the children overcome growing up. An epidemiological study of 5 year old children suggested that sleep problems at this age were significantly associated with sleeping difficulties at age 6 months and with higher probability of sleeping difficulties at 10 years of age (Pollock, 1994). Also, the rate of reported sleep problems does not appear to decrease as children move into the pre-adolescence and adolescence stages (Bearpark & Michie, 1987; Kahn et al., 1989) with well documented serious compromising effects on cognition, mood and behaviour (Pilcher & Huffcutt, 1996).

However, sleep problems are even more common in children with autism spectrum disorders (ASD) with prevalence rates of anywhere between 50% and 90% (Richdale, 1999; Richdale & Wiggs, 2005;) and with sleeplessness (i.e. difficult getting to sleep; waking during the night) as the most commonly reported sleep problem. Research suggests that, in autistic children as well as in TC, these sleeplessness problems are usually caused by children's failure to learn appropriate sleep behaviours (Wiggs & Stores, 2004). The idea that aspects of sleep are learned behaviours is supported by the evidence that these common, persistent and often severe sleeplessness problems (which frequently are present in children with learning disabilities) are often treatable in TC and children with autism with behavioural interventions which use techniques to help them learn a desired new set of sleep behaviours (Richdale & Wiggs, 2005).

Behavioural interventions can be delivered in a booklet form, as a self-help intervention. Booklet-based intervention has practical advantages over face-to-face delivery as it is easy for parents to access and it can be administered to large groups with minimal cost. This method of delivery has been found to be as effective as face-to-face delivered treatments for young children with severe learning disabilities (Montgomery, Stores, & Wiggs, 2004) so it is supposed that this method could be effective with TC and children with autism too. However children with autism have a number of special characteristics (communication difficulties; resistance to change; high levels of anxiety, challenging behaviour and possible melatonin abnormality) which theoretically could make it difficult to implement behavioural treatment perhaps especially in a booklet form without additional support for the parents. There have been some attempts to treat sleeplessness in autistic children with behavioural methods which obtained promising results (Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005) but there is a need for a more defined evaluation of this type of treatment especially in view of the impact that sleep disturbance has on high cognitive functions in children and adolescents with and without autism.

Hence the current study aims to explore the association between sleep and high cognitive abilities such as creativity and to assess if a booklet delivered behavioural treatment is able to improve sleeplessness problems and in turn, creativity in TC and children with HFA.

CHAPTER ONE

Normal sleep and sleep disorders including developmental aspects

1.1 GENERAL ASPECTS OF SLEEP PHYSIOLOGY

A clear appreciation of normal sleep features provides a background for understanding those conditions in which normal characteristics of sleep are altered, and for interpreting certain consequences of sleep disorders. The following paragraphs will describe the physiological aspects of sleep including their development in children and adolescents. States of normal human sleep, sleep stages and sleep-wake cycles will be discussed.

1.1.1 States of normal human sleep

Normal human sleep comprises two separate states that occur in a peculiar sequence: non-rapid eye movement (NREM) and REM sleep. These two states have been defined on the basis of three fundamental measurements. First is brain wave activity measured by Eletroencephalogram (EEG), which provides the summary of electrical activity from several areas of the brain. This is considered the most revolutionary instrument in the history of sleep research given that the difference between the states of sleep and wakefulness was first identified through this (Berger, 1930). Second, the Eletroculogram (EOG) records eye movements during sleep, and finally muscle tone is recorded using an Electromyogram (EMG).

During wakefulness the EEG displays brainwave patterns that can be classified in two types; Beta and Alpha. Beta waves are the highest in frequency and lowest in amplitude and more desynchronous than other waves. In periods of relaxation, the EEG records Alpha waves which are slower, larger and more synchronous. During sleep, EEG records two different types of brain waves, called Theta and Delta. More details about them are given in the following section.

1.1.2 Sleep stages

On the basis of EEG measurements, NREM sleep is conventionally subdivided into four stages (Rechtschaffen & Kales, 1968). Stage 1 and 2 (light sleep) are characterized by Theta waves which are even slower in frequency and greater in amplitude than Alpha waves. In a healthy young adult, Stage 1 lasts 4-5% of total overnight sleep whereas Stage 2 covers 45-55% of one nights' sleep. Stage 3 and 4 (deep sleep) are referred to as Slow Wave Sleep (SWS) and are characterised by Delta waves which are the slowest and highest amplitude brain waves. Delta sleep is the deepest sleep which makes awakenings very difficult. Should sleepers be awoken they often show disorientation. Stage 3 and 4 take between 4-5% and 12-15% of overnight sleep respectively (ASDA, 2005).

While NREM sleep is usually associated with minimal or fragmentary mental activity, REM sleep (so named because "rapid eye movements" occur during this stage), by contrast, is defined by high level of EEG activation similar to wakefulness with Beta and desynchronised waves, loss of muscle tone and sudden body movements. Because of the combination of high levels of brain activity and absence of muscle tone, REM sleep was also named *paradoxical sleep*. In fact the paradox is that the EEG resembles that of waking, whereas behaviourally the subject remains asleep and unresponsive (Jouvet, 1962). In 2007, a new sleep stage classification was published. The review resulted in several changes, the most significant being the combination of stages 3 and 4 into Stage 3 (Iber, Ancoli-Israel, Chesson & Quan, 2007).

Sleep is a cyclical process which begins in NREM and progresses through deeper NREM stages with the first episode of REM sleep occurring approximately 80 to 100 minutes later. Thereafter NREM- REM cycles continue to alternate through the night at 90 to 110 minutes intervals, with stage 3 and 4 concentrated in the early NREM cycles and REM sleep episodes lengthened across the night.

1.1.3 Development of sleep stages

The strongest factor affecting the patterns of sleep stages across the night is based on developmental issues. Several studies show that REM and NREM sleep states change in structure and temporal organization during the first two years of development (Anders, 2004).

REM sleep is known as "active sleep" in newborns, and is characterized by eye closure, bursts of rapid eye movement, irregular respiration and frequent and large body movements (Hobson, 1989). Active sleep accounts for 50% or more of sleep at birth, decreasing to 20-25% in the first two years of life and staying at that level from then on. The high level of REM sleep in early development may suggest that REM sleep plays a role in the development of the central nervous system, though this has not yet been demonstrated (Stores & Wiggs, 2001a).

In contrast, NREM sleep that in infancy is referred to as "quiet sleep" is characterised by eye closure, relatively regular respiration and the absence of rapid eye movement. Quiet sleep, in which SWS is particularly prominent, increases from 50% at birth to 80% by adolescence. For almost 70% of the whole 24 hours, growth hormone (GH) secretion occurs in association with SWS (Van Cauter, Plat, & Copinschi, 1998). In infants, also present is so-called "ambiguous" or "intermediate" sleep which refers to a period of sleep in which active and quiet sleep are concurrently manifested. This irregular state is characterized by slow EEG patterns. The cyclical alternation of NREM-REM sleep is present from birth but has a period of 50 to 60 minutes in the newborn, compared with 90 to 110 minutes in the adolescence and adulthood in which sleep cycles occur 4-6 times per night (Horne, 1989).

As children grow older, they show a range of EEG patterns which begin to resemble adult sleep and the daily hours devoted to REM sleep is decreased. During adolescence, sleep stages do not change dramatically. However NREM sleep becomes the time when the pituitary glands release growth and sexual development hormones, suggesting a new developmental function for sleep.

1.1.4 The sleep-wake cycle

The alternation between sleep and wakefulness in humans is mainly regulated by a *circadian clock* and a *homeostatic process.* In humans the suprachiasmatic nucleus (SCN) of the hypothalamus is the site of a master circadian clock. The SCN not only generates circadian rhythms such as the sleep-wake cycle with a periodicity of about 24 hours (hence the expression circadian from *circa* and *dies* meaning about one day) but also maintains the temporal organization of the circadian rhythms to the external cues. The body clock is aligned to the 24-hour day by environmental cues (or *zeitgebers),* of which light is the most dominant. Synchrony is achieved by a process of 'entrainment' which corrects the difference between the body clock and the period of the environmental cycle. The body clock is entrained to light-dark cycles via suppression of melatonin secretion operated by the light.

Melatonin is produced by the pineal gland and its rhythmic secretion has an important role in the synchronization of the body clock to photoperiodic information. Normally, the production of melatonin (the so-called "hormone of darkness"), is inhibited by light and permitted by darkness. Its secretion peaks in the middle of the night, and gradually falls during the second half of the overnight cycle.

Normal sleep and wake cycles are not only under circadian control but also under the control of the sleep homeostatic process. The homeostatic process is determined by prior sleep and wakefulness In other terms, the need for sleep accumulates throughout the time of wakefulness and dissipates during sleep. The longer the subject stays awake or is deprived of a specific sleep stage, the greater will be the drive to recover the lost sleep or sleep stage. The component of sleep which is most controlled by homeostatic factors is SWS. Studies show a prominent delta sleep rebound during recovery sleep after deprivation (Berger & Oswald, 1962; Gillberg, Anderzen, & Akerstedt, 1991). In other words, the homeostatic process permits the restorative function of sleep which consists in the dissipation of factors accumulated during wakefulness.

The circadian process (process C) and the sleep homeostasis process (process S) interact to consolidate the sleep-wake cycle (Borbely & Achermann, 2005; Feinberg, 1974). When the homeostatic sleep drive becomes more forceful in response to sustained wakefulness and increased propensity to fall asleep with successive waking hours, the circadian clock guarantees maintenance of vigilance throughout the day with the peak of arousal occurring in daytime and in the early evening.

1.1.5 Developmental aspects of the sleep-wake cycle

The sleep-wake cycle changes from a polyphasic rhythm in the new born period, to a monophasic rhythm (in which sleep and waking consolidate respectively at night and in the daytime) by approximately 6 months of age. The daytime naps usually reduce from 4 to 6 in the neonate, to only 1 per day by one year of age. A study showed that the frequency of naps is extremely high in the first year of life, abolished in the school-age period and becoming habitual again in adolescence on weekends (Thorleifsdottir, Bjornsson, Benediktsdottir, Gislason, & Kristbjarnarson, 2002). The typical amount of sleep across the developmental stages is from 16-18 hours during the birth term, to 7-8 hours during later adolescence. However these average values do not consider individual differences in the need for sleep. Whereas in early years children (4-5 years old) generally sleep soundly at night and are alert the following day, during adolescence because of biological change (less SWS than at an early age, physiological delayed sleep phases, and the increase of a physiological need for more sleep) and social factors (the increase of social relationships, intense school and work schedules etc) sleep diminishes relative to sleep need, causing sleep deprivation and increased daytime sleepiness (Ishihara & Miyake, 1998). Also production of melatonin decreases with the developmental process; maximum night-time levels are achieved between 1 and 3 years of age and begin to decline with the onset of puberty. At this stage of development there is also a change with the timing of the secretion of melatonin which peaks later at night reflecting a progressive delay of sleep onset.

1.2 FUNCTIONS OF SLEEP

Despite decades of intense research the function of sleep still remains a biological and psychological enigma. This is not to say that there is a lack of theories concerning the function of sleep. On the contrary there are many, and each of them gives a contribution towards the understanding of why we sleep. More generally we can say that some theories suggest that sleep serves the function of homeostasis, which refers to the physiological tendency to maintain a constant set of internal conditions whereas others, including the developmental theories, sustain the heteroplastic function of sleep which is the capacity to change in response to the environment. Although homeostatic function is mainly concerned with recovery and energy conservation and heteroplasticity with information processing, the two functions can overlap (Hobson, 1989).

1.2.1 Homeostatic theories

The *energy conservation theory* of sleep is associated with homeostatic theories because it concerns the maintenance of thermal equilibrium. Low metabolic demands and decreased body temperature during sleep are ways for mammals to save internal energy. Evidence of the homeostatic function of sleep comes from findings of the fatal effect sleep deprivation can have upon energy and temperature regulation in rats. The subsequently dying rats showed a paradoxical weight loss despite increased food intake, as if they were literally burning up (Rechtschaffen, Gilliland, Bergmann, & Winter, 1983). On the contrary, though human sleep duration and metabolic rate seems to be correlated (Walker & Berger, 1980) the metabolic reduction during sleep seen in humans is only 8% or 10% which is insignificant.

Another explanation of the function of sleep is given by the *replenishment of neurotransmitters theory of sleep* (Hobson, 1989) which argues that the normal function of sleep is to restore levels of neurotransmitters after a day's activities. Most neurons decrease their activity slightly during sleep and a small minority actually cease firing. This cessation involves aminergic neurons thought to be crucial to memory, learning and attention. These aminergic neurons continue to synthesise new neurotransmitters during sleep and so more neurotransmitters will be available when morning arrives. This theory tries to explain why after having slept we feel mentally sharper, but it does not say why some persons (owls) are better at a cognitive level during night time and some others (larks) in the morning; this may mean that this theory is somehow incomplete or that circadian factors reduce or increase the effect of the neurotransmitters' synthesis.

The *restorative theory* is based on the hypothesis that sleep serves to restore physiological and psychological functions that are progressively degraded during prior wakefulness. This view of sleep function is intuitively reasonable because it is in line with the psychological and behavioural problems that we all experience with a loss of sleep.

In the recovery process, SWS seems to be very important (Oswald, 1980), especially as it has been linked with the release of growth hormones and protein synthesis which is needed for the upkeep and repair of all body systems, including the brain. Evidence comes from studies where acute physical activity increased the amount of time devoted to SWS (Shapiro, Bortz, Mitchell, Bartel, & Jooste, 1981; Youngstedt, O'Connor, & Dishman, 1997). However it remains unclear whether the increase in slow wave activity after physical activity is an indication that the central nervous system needs more recovery because of greater nerve activity (e.g. motor planning, processing visual stimuli etc.) or whether it is a side-effect of body heating.

In addition, a few studies have addressed the role of mental activity in determining subsequent sleep depth and their results are contradictory. Some studies show a significant increase in SWS after cognitively demanding activities (Horne, 1992; Horne & Minard, 1985) whereas others found no differences (De Bruin, Beersma, & Daan, 2002; Kobayashi, Ishikawa, & Arakawa, 1998; Takahashi & Arito, 1994). Evidence supporting the restorative hypothesis has also relied on the use of total sleep deprivation (TSD) (Horne, 1993) selective SWS deprivation (Ferrara, De Gennaro, Casagrande, & Bertini, 2000) and REM sleep deprivation (Dement, 1960).

1.2.2 Heteroplastic theories

One idea that has recently become very popular is that sleep has an important role in the learning process. This link between sleep (especially REM) and learning has been established by the so called *learning theory* of sleep. One way that REM sleep could aid the learning process is by reinforcing memory. However, Siegel (2001) argued that a major role for sleep in memory consolidation is not yet proven. In fact it is expected that an increase in learning requires an increase in consolidation and, in turn, in REM sleep. This hypothesis is supported by animal studies (Bloch, Hennevin, & Leconte, 1981) showing an increase in REM sleep duration after being exposed to a novel task. However, Siegel (2001) points out that this increase in REM sleep may be caused by stress, (which is known to increase REM sleep), caused by the learning task.

Although most studies have explored the possible role of REM sleep in learning and memory, some research has been done on the involvement of NREM sleep in learning and memory (Sejnowski & Destexhe, 2000) which suggests that an alteration of stage 2 sleep waves is involved in memory consolidation.

The *maintenance theory* proposes that after development and learning, sleep allows time for the brain to organise and store this information so that it will not be forgotten.

In order to remember this information, REM sleep provides an opportunity for rehearsing less common behaviours (Jouvet, Valatx, & Jouvet, 1961). Hobson (1989) suggested that our memory must be constantly renewed to avoid decay. One way to accomplish this renewal is to relearn the material in waking, but the information which we do not have occasion to practice might be refreshed by automatic brain activation seen in REM sleep. The author proposed that neurons fire in patterns which reproduce the original input, therefore allowing individuals to remember information acquired a long time ago and providing the ability to recognise objects after many years. Obviously, this assumption spurs many questions. For instance, what about the very low amount or absence of REM sleep in other species? Kavanau (1997) argued that the fact that some animal species such as dolphins are active during sleep presents evolutionary evidence for the involvement of REM in the memory maintenance. This was named "dynamic stabilization". Dolphins engage in unihemispheric NREM sleep meanwhile the other hemisphere remains active. These observations led Kavanau to believe that memory maintenance or dynamic stabilization occurs during sleep, but that some animal species can also maintain their memory during wakefulness.

Beside the maintenance theory which emphasises how sleep helps us to remember, there is the *reverse learning theory* of sleep developed by Crick and Mitchison (1983) which, in contrast, suggests that REM sleep serves to remove undesirable data from the memory. Thus, as it is important to retain information, it is equally crucial to erase certain memories in order to not be overwhelmed with too much information. Also, the fact that dreams are difficult to remember confirms the hypothesis that REM sleep is designed to erase rather than strengthen such information. For an explanation of the mechanism causing memory erasing, Crick and Mitchison (1995) conducted experiments with neural network models indicating that when overloaded with too much data, such information can be reduced by a process called 'reverse learning'. Perhaps it can be argued that the difficulties in suppressing irrelevant information observed in some developmental disorders such as autism are connected with the atypical sleep architecture which characterises this pervasive disorder. Thus, the hypoactivation of REM sleep (Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005), REM sleep disruption (Godbout, Bergeron, Limoges, Stip, & Mottron, 2000) and lower beta activity during REM sleep (Daoust, Limoges, Bolduc, Mottron, &

Godbout, 2004) observed in subjects with autism possibly impairs the reverse learning function which should remove irrelevant information from the memory.

1.2.3 Developmental theories of sleep function

Whatever the function of sleep is, it appears to be particularly important during early brain development. In fact, it can be argued that sleep is the primary activity of the brain during early maturation. Roffwarg et al. (1966) gave one of the most important contributions in the understanding of the role of sleep in the developmental process. They suggested that REM sleep was abundant in early life to provide the brain with stimulation necessary for development, especially for development of the visual system. Thus, the visual brain stimulates itself in REM sleep via a mechanism reflected in the EEG recordings as Pons Geniculate Occipital (PGO) waves. Thus, prenatal REM sleep could make the visual system neurons ready to respond to external images. However, scientists still do not know exactly how REM sleep may favour the development of visual systems through the dramatic change in the amount of REM sleep throughout the first two years of life which suggests that this must play an important role in achievement of visual competence.

Animal studies (Hobson, 1989) reveal that sleep is also important for the acquisition of motor skills. REM sleep provides an internal activation of the brainstem motor systems in cubs and controls rhythmic movements such as breathing, feeding and swallowing which are necessary for survival. Once again, it can be argued that REM sleep provides an opportunity for the brain to practice future behaviour. In this way, at birth we know how to breathe and to swallow because we have already learned these acts in the uterus during REM sleep.

Sleep must serve an important function not only in the early stages of life but also in later childhood and adolescence. Evidence of the significant role played by sleep throughout the development process come from sleep deprivation studies with school age children. These studies showed that small reductions of sleep amount, by at least 30 minutes, impaired neurobehavioural functioning (NBF). Children showed reduced alertness (Sadeh et al., 2003) and increased daytime fatigue (Allen, 2003). Also, decreased executive control, impulsivity, emotional ability (Dahl, Pelham, & Wierson, 1991) and diminution of goal-directed behaviour (Dahl, 1996) following inadequate night time sleep has been reported in children. However, one study by Carskadon and Dement (1981a) looked at 4 hours sleep restriction on a single night and no significant effect on cognitive functions were found. On the contrary, a second study (Carskadon & Dement, 1981b), in which 1 night of total sleep deprivation was examined, showed a marked tendency for impairment in cognitive functions during sleep deprivation. Given that children show cognitive and behavioural deficits which resemble the ones found in sleep deprivation studies with adults, these results confirmed that sleep fulfils some vital role in our waking lives, a role that enhances our ability to think, feel and interact since the early stage of life.

To understand the function of sleep, which itself is a complex matter, in adolescence is made even more complicated by the physiologic and psychological changes occurring during this stage of life. In fact, sleep patterns in adolescence emerge from a complex interplay of several distinct processes (physiologic and mental maturation, increased social interactions and peculiar circadian regulatory mechanisms) which play a key role in the transition from childhood to adulthood. Although the purpose of this thesis leaves aside these questions, it is noteworthy to mention that sleep contributes to physical growth and the development of secondary sexual characteristics observed in puberty, given that NREM sleep is the time when steroid hormones are released.

When the functions of sleep fail or are impaired, severe sleep disorders can occur with grave consequences for normal functioning. The purpose of the following section is to provide a brief and general description of the International Classification of Sleep Disorders second edition (ASDA, 2005) and also to reflect about its helpfulness and adaptability to the childhood population.

1.3 SLEEP DISORDERS: THEIR CLASSIFICATION AND MEASUREMENT

The purpose of the following sections is to provide a brief description of the current classification system of sleep disorders used by clinicians and researchers, and to discuss its adaptability to children. Sleep disorders are conditions of a physical and/or psychological nature that cause different types of sleep disturbance or problems. There are three basic types of sleep problems: difficulty getting to sleep or staying asleep; sleeping too much; and disturbed episodes that interfere with sleep. Before listing the main sleep disorders, it is important to mention that a clear distinction between sleep problems and their underlying causes is often not made clear, despite it being extremely important for adequate diagnosis and choice of treatment.

Sleep disorders can be assessed by using a number of methods. The decision of which method to use depends on the population of investigation and on the clinical or research purpose. Given the various differences between children and adults regarding the manifestation of sleep disorders, specialised methods have been developed to assess them in children. In the sleep literature these methods have been conventionally classified as objective and subjective methods of assessment.

A description of these methods of assessment of childhood sleep disorders in research will also be presented in the following paragraphs. Each method will be described with emphasis on its applications, strengths and weaknesses.

1.3.1 The International Classification of Sleep Disorders

The International Classification of Sleep Disorders Diagnostic and Coding Manual (ICSD-2) was published in 2005 by the American Academy of Sleep Medicine. The new edition replaces ICSD-1, published in 1990 and revised in 1997. This system classifies sleep disorders into eight major categories:

- Insomnia
- Sleep related breathing disorders
- Hypersomnias of central origin
- Circadian rhythm sleep disorders
- Parasomnias
- Sleep related movement disorders
- Isolated symptoms and normal variants
- Other sleep disorders

The ICSD-2 includes over 70 specific diagnoses within these eight categories as well as two appendices for classification of sleep disorders associated with medical or psychiatric disorders. Unlike ICSD-1, the ICSD-2 does not use a multiaxial system but focuses only on the diagnosis of sleep disorders. Some disorders are grouped according to a common complaint (e.g insomnia, hypersomnia, parasomnia, and sleep-related movement disorder) others are classified by presumed etiology (e.g circadian rhythm sleep disorders) and others are classified according to the organ system from which they arise. Separate sections deal with disorders that involve isolated symptoms, such as sleep talking, and longer and shorter than normal sleep duration. Appendices review sleep disorders associated with medical disorders, such as sleep-related epilepsy or headaches, along with psychiatric and behavioural disorders.

For each disorder, ICSD-2 describes essential and associated features, demographics, predisposing and precipitating factors, as well familial patterns. It also reports onset, course, complications and pathology. It includes polysomnographic and other objective findings and diagnostic criteria. Each section also addresses unresolved issues, further directions, and differential diagnosis, concluding with a concise bibliography.

Most pediatric presentations are incorporated into the text for individual sleep disorders but presentations unique to childhood are listed separately (behavioural insomnia of childhood, primary sleep apnea of infancy, obstructive sleep apnea pediatric, congenital central alveolar hypoventilation syndrome, sleep enuresis, restless legs syndrome, sleep related rhythmic movement disorder). The utility of the ICSD-2 or other older sleep classifications has been questioned in sleep research with children. Generally researchers do not use classification systems but typically construct their own criteria or definition of sleep problems, such as cut-off or frequency scores on a sleep questionnaire or diary, or based on parental reports that a sleep problem is presented.

Also children's sleep disorders differ from those in adults in many ways and these are not always made explicit in sleep classification systems. Firstly, sleep disturbances may result in overactivity and other forms of disturbed behaviour, in contrast to adults who are more likely to be sleepy and tired during the day; secondly, children's sleep problems have repeatedly been shown to be associated with adverse effects on family functioning (Gregory, Eley, O' Connor, Rijsdijk, & Polmin, 2005). Finally, most of the common children's sleep disorders are readily treatable or have a self limiting time-frame (Stores & Wiggs, 2001b).

In addition, a new code for childhood behavioural insomnia presented in the ICSD-2 that combines two ICSD-1 disorders (sleep onset association disorder and limit-setting sleep disorder) was downgraded and this has sparked consternation among some pediatric sleep specialists given the drammatic effect that insomnia has on children's daytime behaviour and family functioning.

A review of the objective and subjective methods of assessment of sleep disorders used both in clinical and research settings is shown below.

1.3.2 Objective methods of assessment of children's sleep disorders

Nocturnal polysomnography (PSG) is a diagnostic test considered to be the "gold standard" of sleep measurement. PSG is used most commonly in the sleep laboratory for patients suspected of having respiratory disturbances, nocturnal myoclonus, REM behaviour disorders, or narcolepsy. Patients spend a full night at the sleep laboratory attached to equipment which transmits electrophysiological signals from various regions of the body to computerised collecting stations which continuously record the signals throughout the night. PSG records multiple physiological parameters during sleep such as electroencephalogram (EEG), electro-oculogram (EOG) and electromyogram (EMG) which identifies the different sleep stages, helps to detect the above mentioned sleep disorders and also other physiological parameters (e.g. respiratory excursions of the thorax and abdomen etc.) (Spielman, Yang, & Glovinsky, 2005). Evaluation with PSG allows objective comparisons with normative data and with data gathered within various clinical populations. PSG can also be used to objectively evaluate treatment and to provide a helpful check on the accuracy of the sleep complaint. Sometimes, a large discrepancy between objective parameters and their corresponding subjective estimates characterises the core of the presenting problem (Frankel, Coursey, Buchbinder, & Snyder, 1976; McCall & Edinger, 1992). For instance, PSG allows diagnosis of "sleep state misperception disorder" in which a complaint of insomnia or excessive sleepiness occurs without objective evidence.

PSG has various limitations. Firstly, environmental factors such as noise, temperature or the presence of other persons, that exist in the natural environment and have an influence on sleep, can not be assessed in the laboratory; secondly, polysomnography in the sleep laboratory always includes one adaptation night in order to account for the "first night effect" (Agnew, Webb, & Williams, 1966; Riedel, Winfield, & Lichstein, 2001); thirdly, PSG recording is expensive so that only one or two nights of data are typically obtained, resulting in a limited sample of sleep which can be sufficient for diagnosis of some sleep disorders such as obtrusive sleep apnoea but not for assessment of others. For instance, PSG is not indicated as a first-line diagnostic tool in the assessment of insomnia (Spielman et al., 2005) as it would provide an inaccurate representation of the subject's sleep patterns which would be paradoxically improved because of the "reverse first night effect" (Hauri & Olmstead, 1989). PSG may also not be well tolerated by children (Wiggs & Stores, 1995). Finally, there is a limited availability of PSG in clinical services.

In contrast, *ambulatory polysomography* (APSG) is a portable device which has the advantage to record physiological parameters during sleep while individuals remain in their own beds at home. For instance, Minisomno, Vitaport, Embla, and Monet are ambulatory polysomographs which work on the basis of implanted computer systems. The recording units are small and can be worn on the body. Data is stored on a memory card or hard-disk. The recorder can even be connected through a computer network or a telephone modem to the sleep lab to control for technical problems such as loosening of electrodes or in case of a decline in power supply. Also, APSG can be used in the sleep lab for continuous 24 hourly sleep monitoring under the control of attending personnel (Penzel & Peter, 2007).

PSG criteria are not always useful for the diagnosis of sleep problems in children. In fact APSG has drawbacks especially for the investigation of settling and night wakings. Firstly, this method requires the cooperation of the child during the fitting and the night-time recording period to avoid detachment of the electrodes; secondly, children with sleeplessness who are usually upset during nocturnal wakings could find the presence of a recorder an additional source of distress. Thirdly, children with settling problems who leave their bed at night to visit the parents' bed could inadvertently disconnect the leads of the sleep recorder. Finally, although ambulatory recording is less distressing for children than admission to a sleep laboratory, the lack of behavioural nocturnal observation makes some records more difficult to interpret.

Thus, in order to overcome this problem, *video/audio recordings* can accompany PSG. These methods are especially useful for identifying and describing parasomnias or other forms of sleep disturbances. Such recordings significantly improve the accounts provided by parents who may otherwise only be able to provide incomplete and distorted information (Wiggs & Stores, 1995). Audio monitoring has been used both to screen for sleep disordered breathing and to record vocalisation during nocturnal sleep, however Goldstein et al. (1994) found that a sound recording was only positive 50% of the time when compared to PSG. Videos are complementary to audio recordings especially in those circumstances such as room-sharing in which it is necessary to identify the source of the sound (Fuller, Picciotto, Davies, & McKenzie, 1998). Time lapse video recordings of infants have shown to correlate highly (r=79) with PSG in distinguishing between wakefulness and active and quiet sleep in infants (Anders & Sostek, 1976).

An *actometer* or *actigraph* is a device which seems to be particulary suitable to measure sleep and wakeful states in children. This is a small wristwatch-like device usually worn on the non-dominant wrist or ankle of the child, although there is close correspondence between recordings from either wrist or ankle (Zizi et al., 1996) or dominant/non-dominant side (Jean-Louis et al., 1996). Actigraphs record body movement during sleep, above a pre-set threshold, across many days and nights. A pulse is triggered for each movement exceeding the threshold and a cumulative score is recorded over a preset time interval or epoch that the majority of sleep researchers set at 1 minute. Then the data collected is usually downloaded to a computer for analysis.

Actigraphy is less sensitive to wakefulness than PSG (Sack, Blood, Percy, & Pen, 1995) and can overestimate sleep time if the subject is awake but still in bed. Further, obviously conventional sleep staging is not possible. However, they have been successfully used in clinical and research settings (Sadeh, Hauri, Kripke, & Lavie, 1995) with young children (Sadeh, Lavie, Scher, Tirosh, & Epstein, 1991) adolescents (Matty, Dahl, & Al-Shabbout, 1994) and with children and adolescents with autism spectrum disorder (ASD) (Allik, Larsson, & Smedje, 2006; Oyane & Bjorvatn, 2005). In addition, evaluations of its reliability and validity have been encouraging. In studies with infants less than 2 years old and with children aged 3-13, agreement rates between actigraphy and PSG have been found to be 95.3% and 89.9% respectively (Sadeh, Alster, Urbach, & Lavie, 1989; Sadeh et al., 1991). There are various actigraphs and associated software available commercially, such as the Basic Mini-motionlogger and Actiwatch-64, which have different sensitivities to the detection of movement and different algorithms to estimate sleep and wakefulness. Each system has its own strengths and weaknesses with respect to device reliability, recording modes, software and documented measure validity. No single system, however, has been validated for all ages or types of patients and reports comparing systems are rare, thus caution is required when comparing studies that have used different systems. However, the ability to provide prolonged recordings in a natural setting and their simple scoring procedure makes actigraphy an attractive alterative to PSG, especially for assessing the sleep of children who are less likely to tolerate laboratory monitoring and for those studies where sleep timing and continuity are the primary interest.

An evaluation of sleep disorders in children requires a multidimensional approach which combines different methods of assessment in order to obtain a complete understanding of the problem and to evaluate the appropriate treatment. A description of the most commonly used subjective methods of sleep assessment in research settings are described below.

1.3.3 Subjective methods of assessment of children's sleep disorders

Subjective methods of assessment of children's sleeplessness are generally based on parental account of sleep being a problem. *Sleep history* is important for both clinical and research purposes. Parents are usually the main source of information but also teachers and the children themselves, if they are old enough, could give an important contribution to the reconstruction of the child's sleep history.

Feber (1995) suggests using specific questions to emphasise the importance of establishing the precise pattern of the child's sleep-wake cycle in order to gather more information into the true nature of sleep problems. However, a full sleep history takes a fair amount of time. On the contrary, questionnaires to be completed by parents or children are subjective methods of assessment of children's sleep problems that are widely used in research and they are easy and quick to complete. There are several types of sleep questionnaires.

Screening questionnaires constitute a first level of evaluation and attempt to determine specific areas of need. To do this, sleep screening questionnaires employ brief, relevant questions for each sleep disorder (e.g. early waking, nightmares etc.) to identify their possible presence in a short time.

The "Simonds and Parraga Sleep Questionnaire" (1982) is a screening tool widely used in research. This tool was developed for epidemiological purposes and has been successfully employed to screen for sleep disorders and sleep behaviour in school-age children, especially with neurodevelopmental disorders (Simonds & Parraga, 1982; Wiggs & Stores, 1998b). The questionnaire is divided into two parts. The first part includes not only general questions about the child's age, height, weight and medical problems but also questions concerning the quality and quantity of sleep (e.g. "how long does it usually take for your child to fall asleep at night?"). The second part is concerned with sleep disorders such as sleepwalking, sleep terrors, and narcolepsy which have been defined in DSM-III (1980), and sleep behaviours which are sleep-related activities such as snoring and fear of the dark, which are not defined in the DSM-III (1980).

This screening questionnaire has satisfactory psychometric properties with test-retest reliability for individual items over a two week period ranging from .83 to 1.00 (Wiggs and Stores, 1995). This questionnaire also has the benefit of being easily intelligible to informants and to quickly identify the presence of sleep disorders but in the same way as all other screening questionnaires, it is useful only for a first level of evaluation and it should not be a substitute for a more comprehensive and detailed sleep assessment. The definition of sleep disorders used in this questionnaire is also based on an old edition of Diagnostic and Statistical Manual of Mental Disorders whereas a new edition is now available (APA, 2000).

Bruni et al (1996) developed the "Sleep Disturbance Scale for Children" (SDSC) in order to provide a standardised measure of sleep disturbance in childhood and adolescence based on the American Sleep Disorder Association (ASDA, 1997) . The authors preferred to use the old classification system rather than the new one because it seemed to be more adaptable to childhood and more clinical in its nature. The questionnaire consists of two sections: the first section gains demographic, behavioural and clinical data with specific questions about pathology that could affect sleep; the second section is made up of 26 items in a Likert type scale with values of 1-5, where higher numerical values reflect a greater clinical severity of the symptoms. The factor analysis yielded six factors which represented the most common areas of sleep disorders in childhood and adolescence and the test-retest reliability was adequate for the total (r=.71) and single item scores. The questionnaire appears to have several benefits: firstly, it can be used as a simple screening questionnaire; secondly, it can evaluate sleep disturbances of school-age children, both in large surveys and in research applications in clinical and non clinical population; thirdly, it can be an useful tool in analysing the relationship between sleep disturbance and other variables such psychological condition and cognitive performances; and finally, it is a valid measure of discrimination between control and clinical groups.

However this questionnaire is not without limitations. As with the majority of sleep screening questionnaires for children, the collection of information is limited to parents, whereas valuable data could be obtained from the children if a standardised version for children were available. Moreover further studies are needed to verify parental sleep reports and the validity of SDSC compared to sleep laboratory measures.

Other sleep disorder screening questionnaires for children are available for research purposes, such as the "Children's Sleep Habits Questionnaire" (CSHQ) (Owens, Spirito, & McGuinn, 2000) and the "Behavioural Evaluation of Disorders of Sleep Questionnaire" (BEDS) (Schreck, Mulick, & Rojahn, 2003). Whereas CSHQ has satisfactory psychometric properties, only limited psychometric data supports the use of the BEDS (Schreck et al., 2003).

Besides screening questionnaires, there are *questionnaires directed to particular sleep symptoms* such as the "Epworth Sleepiness Scale" (ESS) (Johns, 1991) which gives an evaluation of sleepiness in everyday situations. In contrast to other sleepiness scales that are not sensitive to minor changes in alertness such as the Stanford Sleepiness Scale (SSS) (Hoddes, Zarcone, Smythe, Phillips, & Dement, 1973) the ESS seems to have greater potential in making a distinction between clinically sleepy groups of patients (e.g. those with obstructive sleep apnoea or narcolepsy) and control groups. In addition the ESS seems to be an appropriate form of assessment not only for adults but also for children (Wiggs, 1995).

The Paediatric Sleep Questionnaire (PSQ) (Chervin et al., 2000) is also directed towards identifying specific sleep disorders in children for research purposes. In fact, it aims to investigate the presence of childhood sleep-related breathing disorders (SRBDs) and other symptoms such as snoring, sleepiness and related behaviour disturbances. The scales showed good test-retest reliability with .92 for the snoring scale, .83 for behaviour scale, .66 for the sleepiness scale, and .75 for the SRBD scale. Although the SRBD, snoring, sleepiness and behavioural scales appear to be valid and reliable instruments for use in clinical research, their applications in clinical practice should probably await studies of whether these scales distinguish between referred patients with SRBDs and those without SRBDs, and between patients with SRBDs and those with other causes of excessive daytime sleepiness.

The most common sleep disorders in childhood are those resulting from behavioural disturbances such as inability to settle to sleep alone or at an acceptable time (bedtime refusal) and repeated night waking often with disruptive behaviour and early morning arousal (Richman, 1987). These problems have been termed disorders of initiating and maintaining sleep (DIMS) (Bruni et al., 1996). Despite the high prevalence of DIMS in childhood (prevalence of sleeplessness in typical developing children and children with autism will be described in detail in section 2.1.2 and 2.2.2) and the fact that research has found that sleeplessness affects memory, attention, vigilance, and creativity (Allen, 2003; Dahl, 1996; Randazzo et al., 1998; Sadeh et al., 2003) there are few suitable measures to assess sleeplessness problems in childhood. A number of questionnaires have been utilised in research with children but these were generally designed to capture a broader range of sleep disorders (Simonds and Parraga, 1982). In addition, of the questionnaires that are available for children, few of them appear to have had a comprehensive psychometric evaluation (Morrell, 1999; Owens, Spirito, & McGuinn, 2000). A questionnaire that has been subjected to rigorous psychometric testing is the "Sleep Disturbance Scale for Children" (Bruni et al., 1996). As has been previously mentioned, this questionnaire is designed to capture a wide range of children's sleep disorders, however it contains a sub-scale relevant to DIMS which had been validated.

In an attempt to standardise assessment of sleeplessness, Richman and Graham (1971) combined items on the Behaviour Screening Questionnaire to produce a "Composite Sleep Disturbance Index" (CSDI) which reflects frequency and occurrence of settling and night waking problems. Since that time, use of a CSDI has permitted researchers to group childhood sleeplessness in broad categories as "mild", "moderate", and "severe" (Montgomery et al., 2004; Richman & Graham, 1971; Wiggs & Stores, 1998a) according to the frequency and duration of the sleep problems. However, assessment of these variables may not be conducted through only the use of questionnaires, but also by the use of sleep diaries and actigraphs.

It is important to say that most questionnaires have focused on parental reports and have not been well developed for use with children. Independent accounts from parents and children might reveal subjective and private matters such as fears associated with sleep about which parents can be unaware (Stores, 1996). In addition, most studies have concentrated on infants and young children's sleep problems, whereas relatively few studies have addressed these issues in school-aged children and adolescents.

Retrospective instruments such as those described above have the advantage of being able to summarise events occurring over a long period of time, but along with this quick summary comes the distortion inevitably introduced when recalling past events. Memories are often incomplete or selective; especially in the case of insomnia as there is a natural tendency to focus on the worst experiences and perhaps to amplify their importance. However these disadvantages can be minimised through the use of a prospective *sleep diary* which can provide much more accurate information. Parents or children themselves are asked to keep a daily record of parameters such as the time they went to bed, sleep-onset latency, frequency and total duration of wakings and total sleep time for at least one week. Also, other relevant events can be noted. They are encouraged to complete the diary at the time of the event they are recording.

Filling in a sleep diary directs the parents' attention to aspects of their children's sleep behaviour that might otherwise be overlooked. Some versions present the information in a graphical format that allows a large amount of data to be inspected rapidly, whereas others require more precise information. (Spielman et al., 2005). Diary records provide not only the basis on which treatment programmes may be devised, but also a baseline against which treatment responses may be measured. High internal consistency has been reported and sleep diaries are widely used for clinical and research reasons (Minde, Faucon, & Falkner, 1994).

1.4 SUMMARY

Normal human sleep can be divided into two separate states that occur in a peculiar sequence: REM and NREM sleep. These two states have been defined by means of three fundamental measurements: EEG, EOG and EMG. On the basis of EEG measurements, NREM sleep is conventionally subdivided into stages 1 to 4 indicating increasing depth of sleep. Several studies related to the physiological maturation of sleep stages show developmental changes in the micro and macro structure and timing of sleep.

Several theories of the function of sleep suggest that adequate quantity and quality of sleep is necessary for optimal daytime functioning. Deficits seen in behaviour, learning and creativity following sleep disruption emphasise the need to investigate disordered sleep. Sleep disorders are conditions of a physical and/or psychological nature that cause different types of sleep disturbance or problems.

Sleep problems can be evaluated by different methods which have been conventionally classified as objective and subjective. Depending on the population being investigated and on the clinical or research purpose of the investigation, the choice of method to be used will vary. However, in general, objective and subjective measures are used in conjunction because they allow a more complete sleep assessment. Given the differences between children and adults regarding the manifestation of sleep disorders, specialised methods have been developed to assess sleep in children. However, for clinical research purposes, the measures need to be uncomplicated, psychometrically satisfactory and accepted by children and their parents.

Prevalence, causes and general effects of sleeplessness in childhood and its treatment will be discussed in detail in the next chapter.

CHAPTER TWO

Sleeplessness in typically developing children and in children with Autism

2.1 SLEEPLESSNESS IN CHILDHOOD AND ADOLESCENCE

Sleeplessness is the inability to sleep which appears to be the most common and intractable childhood sleep problem and to which most of the research literature is devoted. Longitudinal studies have demonstrated that sleep problems first presenting in infancy may become chronic, persisting into the pre-school and school-age years. This section aims to describe some of the sleeplessness definitions that have been provided on the basis of different methodologies which assess parents' and children's perception of sleeplessness. Also prevalence and causes of sleeplessness will be discussed.

2.1.1 Definitions of sleeplessness

Several attempts have been made to define sleeplessness in children. The ICSD (ASDA, 2005) contains criteria for the diagnosis of several sleep disorders, including common causes of sleeplessness in children; though only a minority of published reports use the ICSD criteria. Perhaps this can be ascribed to the limited adaptability of this classification to children, as has been previously mentioned in section 1.3.1. Interestingly, some authors have categorised children as "good" or "bad" sleepers on the basis of parental reports of sleep being a problem. Although such an approach has the merit in detecting a subjective evaluation of sleep, the variability of personal, family and cultural factors which may influence these judgements make it difficult to compare groups of children or extend these evaluations to the general population of children. For instance, considerable variation has been found between countries in sleep onset times and the amount of time slept in childhood and adolescence (Sverrisson & Kristbjarnason, 1990; Tynjala, Kannas, & Valimaa, 1993) which make it hard to establish an absolute value representative of the satisfactory amount of hours a child should sleep. Thus, whereas going to bed at midnight could be considered a serious sleeplessness problem by a family of English nationality, this is maybe considered a reasonable bedtime for children in Sicily. In other words, the definition of sleep problems in young children is highly influenced by the environmental and cultural context in which they occur.

Further research should be focused on children's own definitions of what a sleep problem is. Subjective definitions of sleeplessness are mainly based on parents' evaluations whereas little is known about the personal perception that children have of their own sleep patterns. For example, sleeplessness may only be a problem for parents but not for children if wakefulness consists of engaging in a pleasurable activity such as watching TV or reading (Wiggs, 2007). Williams et al. (2007) suggested that children consider staying awake until late as a sign of maturity and also underestimate the consequences of inadequate sleep. However, a study showed high correlation between subjective and objective sleep indices reported by school age children suggesting that children's sleep questionnaires can be applied to a survey for screening sleep habits (Gaina, Sekine, Chen, Hamanishi, & Kagamimori, 2004).

Validation of subjective sleep assessments for children is not well established. For instance, there are a number of child sleep questionnaires for parents with good psychometric properties (e.g. The Children's Sleep Habits Questionnaire by Owens et al., (2000) and Sleep Disturbance Scale for Children, by Bruni et al., (1996) which do not have a validated self-report version for children. Consequently more knowledge of children's definition of sleeplessness is necessary to optimise treatments and interventions which perhaps would be more effective if relevant to the needs of the child.

Sleep problems of settling, night waking and early morning waking appear to be very common forms of sleeplessness in children. These types of sleeplessness can appear in isolation or in combination. With respect to settling and night waking problems, a number of studies have tried to quantify them in terms of the length of time taken to sleep, numbers of wakings during the night, length of time of each waking and the total number of occurrences of these difficulties each week. Richman and Graham (1971) combined items on the Behaviour Screening Questionnaire to produce a "Composite Sleep Disturbance Index" (CSDI), which reflected frequency and occurrence of settling and night waking problems. This formed the basis of the first time operationalised definitions which were used to classify childhood sleeplessness.

Richman and Graham (1971) classified both mild and severe sleep problems as: mild settling problems which occur once or twice a week and the child settles in less than an hour; severe settling problems which occur three or more times a week and the child takes over an hour to settle; mild night waking which occurs once or twice a week and the child wakes for a few minutes; severe night waking which occurs three or more times a week for more than a few minutes and the child is disturbed or goes into the parents' bedroom.

However, Richman (1981) in a successive study clarified the definition of night waking to describe both children who woke briefly but frequently each night and those who woke only once but for a longer time period. Since that time, use of a CSDI has allowed researchers to group childhood sleeplessness in broad categories such as "mild", "moderate", and "severe" (Montgomery et al., 2004; Richman & Graham, 1971; Wiggs & Stores, 1998a) according to the frequency and duration of the sleep problems, and has also allowed comparisons with other works. Assessment of these variables may not be only by subjective reports (e.g. sleep diaries or questionnaires) but also by objective recordings of the child's sleep patterns (e.g. actigraphs). Correspondence between these objective and subjective assessments of sleep measures is generally good (Sadeh, 1990) but not always without some differences (Acebo et al., 2005).

Early waking, which as indicated above is a feature of sleeplessness in childhood, has usually not been included in paediatric sleep problem research and has often not been given a clear definition (Clements, Wing, & Dunn, 1986). However, the Simons and Parraga Sleep Questionnaire (1982) is perhaps the only paediatric sleep questionnaire with the advantage of having a specific question about child's early waking time ("does your child usually awaken before 5 a.m. in the morning?"). Setting a time limit for defining early waking is appropriate if the reduction of total sleep time is a consequence. In fact early waking may not in itself be indicative of insufficient sleep. For instance, children with advance sleep phase syndrome who fall asleep unusually early could wake up before 5 a.m. with no reduction of total sleep duration.

It should be noted that although research provides definitions bedtime problems, night and early wakings generally include parameters related to some combination of frequency, severity and chronicity, the research criteria used in the literature to define sleeplessness problems are not always consistent across studies. For this reason, standard research definitions of sleeplessness in young children need to be used consistently in future research. Furthermore an increment in the use of objective assessment tools, such as actigraphy, would be highly beneficial.

2.1.2 Prevalence of sleeplessness in typically developing children and adolescents

Sleeplessness is one of the most frequently reported sleep problems which affects 30% of typical developing children (TC) and adolescents with different degrees of severity (Kahn et al., 1989; Manni et al., 1997; Morrison et al., 1992; Owens, Spirito, McGuinn, & Nobile, 2000). However it is not possible to estimate the true prevalence of sleeplessness in such a young population because of the lack of epidemiological studies in childhood and adolescence (Blader, Koplewicz, Abikoff, & Foley, 1997). Contrary to popular belief, sleeplessness is not a transient problem which decreases across all ages, but on the contrary it risks becoming a more severe and persistent problem if not efficiently and promptly treated.

An epidemiological study showed that sleeping problems at the age of 5 were significantly associated with sleeping difficulties at the age of 6 months. But most interestingly, this study predicted that children with sleep problems at 5 years of age were more likely to develop a sleep problem at 10 years of age (Pollock, 1994). This study confirmed the results of previous research which showed that 50-70% of 1 year old children with sleep problems will continue to present those problems at age 2 (Jenkins, Owen, Bax, & Hart, 1984) and nearly two thirds of these children will be described as having chronic sleep problems five years later (Richman, Stevenson, & Graham, 1982). Also, other more recent studies arrived at similar conclusions: 25-50% of 6-12 month old children were found to have difficulties with settling to sleep or waking in the night, and about 25-30% of children between 3 and 5 years old were described as having persistent sleeplessness problems (Mindell, 1993; Mindell & Owens, 2003). Once again, the percentage of children with sleep problems seems to not reduce as the children get older.

With respect to specific sleeplessness problems, as has been previously mentioned, settling problems and nocturnal waking are the most frequent sleep problem complaints in childhood. However, it is a common perception that night waking declines with age whereas bedtime resistance increases. Nocturnal wakings have been reported in 20% to 30% of 1 to 3 year old children (Thunstrom, 1999; Zuckerman, Stevenson, & Bailey, 1987) and 6% of 5 to 12 years olds (Blader et al., 1997; Owens, Spirito, McGuinn et al., 2000) whereas bedtime resistance has been described as increasing in the first five years of life from 14% during infancy to 50% at 5 years old (Beltramini & Hertzig, 1983). Older children are more likely to experience sleep onset difficulties or delays rather than resistance in going to bed (Wolfson & Carskadon, 1998). About 25% of parents reported that their school age child has significant difficulties in falling asleep (Stein, Mendelsohn, Obermeyer, Amromin, & Benca, 2001). Several studies have also reported significant difficulties in falling asleep (Stein et al., 2001), diminution of nocturnal sleep duration and increased daytime sleepiness (Thorleifsdottir et al., 2002) in school age children.

It is important to know, however, that all of these studies are cross-sectional and differ considerably in sampling procedures and age representation. In addition, the few long-term studies that have been carried out were focused mainly on pre-school children (Kataria, Swanson, & Trevathan, 1987; Lam, Hiscock, & Wake, 2003; Zuckerman et al., 1987). In addition, a study about children's sleep behaviour, perhaps one of the few in which children were followed longitudinally in the first 10 years of life, challenged the view that night waking gradually decreases with advancing age whereas bedtime resistance increases (Jenni, Fuhrer, Iglowstein, Molinari, & Largo, 2005). This study showed that night wakings tended to persist over time. Between the age of 2 and 7 years, almost 40% of all children woke up at least once per week over a period of 3 years. On the contrary, bedtime resistance and sleep-onset difficulties tended not to persist; 12% of all children had occasional bedtime resistance, and only 5% had sleep-onset difficulties for a period of 3 years. In one of the few studies to employ both objective methods of sleep measurement (actigraphs) and subjective rating by parents (questionnaires), Sadeh, Raviv and Gruber (2000) examined the sleep of 140 children aged between 7 and 12 years. Results showed not only that older children had more morning drowsiness and unplanned naps, but that two wakings per night still occurred. Thus, as found by Jenni et al. (2005), the number of wakings experienced by older children was similar to that of infants and toddlers.

Whatever the trend of these sleeplessness problems, high rates of sleeping difficulties are still reported in adolescence. In a recent study, Laberge et al (2001) surveyed mothers of 1146 children aged 10-13 years and found that nearly 60% of the children had difficulty falling asleep and 26% were described by the mothers as having had frequent nocturnal awakenings at one point between age 10 and 13. But, although the prevalence of difficulty falling asleep and nocturnal awakenings reported in this study are similar to those reported by early adolescents in other populations (Bearpark & Michie, 1987; Klackenberg, 1982; Tynjala et al., 1993) surprisingly a decrease in these problems with this age was found. This result stands out from the majority of sleep studies in adolescence in which classic sleep disturbances are reported to be more frequent in older than in younger adolescents (Carskadon, 1982). Table 2.1 summarises the results of some of the most representative studies about the prevalence of sleep problems in TC.

Table 2.1 Prevalence of sleep problems in typically developing children

Studies	**Subjects N**	**Age range (years)**	**Sleep problems (%)**
Owens et al. (2000)	424	5-9	37
Kahn et al. (1989)	972	8-10	43
Laberge et al. (2001)	1146	10-13	60
Morrison et al. (1992)	943	13-15	40

2.1.3 Causes of sleeplessness in typically developing children and adolescents

The factors underlying sleeplessness vary across the developmental sequence. During the newborn period, certain medical conditions are considered to be a primary cause of infant's sleeplessness. *Allergies* may interfere with night time sleep due to the introduction of cow's milk into the diet (Andre Kahn et al., 2001). *Infantile colic* and *otitis* are also considered to be causes of night-time waking and to be associated with intense nocturnal crying. Finally *gastro-oesophageal reflux* which is characterised by regurgitation of stomach content into the oesophagus is another cause of sleep disruption during the newborn period. Other non-medical causes of sleeplessness in infancy are frequent night-time feeding and discomfort from wet nappies (Wiggs & Stores, 2001).

Although substantive literature addresses infantile sleep problems, little is known about the causes and whether the waking is harmful for the baby. Toddlers and pre-school children often display *separation anxiety at bedtime*. Studies have demonstrated an association between insecure maternal attachment during the day and distress at nocturnal separation (Benoit, Zeanah, Boucher, & Minde, 1992). In addition the development of self-consciousness between 1 and 3 years of life which leads to *fear of being alone during the night* and also the *inability to self-sooth* are frequently the cause of delayed bedtime and settling problems. The loco motor abilities, which in TC develop between age one and three, might facilitate to seek parental proximity at night and the consolidation of bad sleep habits. A longitudinal study in which 493 children were followed in the first ten years of life found that bed-sharing and night waking peaked at 4 years old (Jenni et al., 2005). The author argued that the explanation for this result is based on specific developmental changes in separation-attachment processes, self-recognition, night time fears and locomotion which may contribute to the age trend of night waking and bed sharing during early childhood. Settling to sleep at night may also be difficult because the child's *daytime naps* are frequent, long and too close to bedtime. *Advanced sleep phase syndrome* can cause early morning insomnia and the inability to stay awake in the evening in early childhood. Early meal and nap times may also facilitate the child's tendency to fall asleep unusually early.

During childhood the main and very common cause of sleeplessness is *inappropriate sleep behaviour*. Resistance in going to bed with repeated requests of attention and recurrent awakenings associated with crying often occurs between one and three years of age (Anders, 2004). These problems will persist throughout childhood if appropriate sleep behaviours are not learnt. Therefore, poor sleep habits may contribute to increasing settling difficulties in childhood, including not being able to sleep in a dark room, lengthy and inappropriate bedtime rituals, and being unable to sleep without the mother's presence. Such habits are likely to increase the time taken to settle to sleep and to fall asleep at night if not removed. Also, frequent awakenings during the night can be the consequence of bad sleep habits. The idea that sleep is a behaviour, that when not properly learnt causes difficulties, is supported by a number of studies which show that even children with mental impairment can learn successfully appropriate sleep behaviour and overcome sleeplessness (Richdale & Wiggs, 2005).

There is a growing amount of literature which supports the notion that *media exposure* impairs sleep patterns in school aged children (Tazawa & Okada, 2001; Van Den Bulk, 2004). For instance, a recent study showed that computer game consumption and television viewing resulted in significantly reduced amounts of slow-wave sleep as well as prolonged sleep-onset latency and reduced sleep efficiency (Dworak, Schierl, Bruns & Strüder, 2007).

With respect to adolescence, both *exogenous and endogenous factors* may contribute to the insurgence of sleep deprivation problems. Exogenous problems include change in social roles, work demands and peer influences (Carskadon, Vieira, & Acebo, 1993; Fallone, Owens, & Deane, 2002; Klimes-Dougan, Hastings, Granger, Usher, & Zahn-Waxler, 2001). For instance, part-time employment while attending high school is common in adolescence. There is evidence to suggest that those adolescents who work for a large number of hours tend to get an insufficient amount of sleep compared with adolescents who work fewer hours (Dornbusch, 2002).

Furthermore cross-cultural research reinforces the view that less total sleep time among adolescents is associated with the inability to concentrate on schoolwork, mood disorders and substance abuse (Carskadon, 2002). In addition, sleeplessness in adolescents can be the result of lack of consistency in the routine family functioning. Hence an irregular sleep-wake schedule (e.g. irregular mealtimes, bedtime and waking up times) can combine to disrupt circadian rhythms, to which adolescents are especially prone, with negative effects showing in their daytime functioning (Stores, 2001b). In fact, adolescents demonstrate a delayed circadian phase preference compared to children and adults (Laberge et al., 2000). This condition is characterised by a physiological difficulty in getting to sleep until very late and by great difficulty in getting up in time for school or work as well as sleepiness during the day. Phase preference was long attributed to exogenous psychosocial factors (Anders, Carskadon, Dement, & Harvey, 1978; Kirmil-Gray, Eagleston, Gibson, & Thoresen, 1984) but more recently has been linked to changes in biological rhythms, possibly related to the onset of puberty (Carskadon, Acebo, Richardson, Tate, & Seifer, 1997; Laberge et al., 2000; Waldhauser & Steger, 1986). Onset of puberty with circadian factors such as diurnal rhythm of cortisol, circadian variations in temperature and melatonin levels count as endogenous factors which cause sleeplessness in adolescents (Born & Fehm, 1998; Born, Muth, & Fehm, 1988; Capaldi, Handwerger, Richardson, & Stroud, 2005; Duffy, Rimmer, & Czeisler, 2001).

It is important to highlight that there are specific child and adolescent populations who are particularly prone to disturbance of sleep. These include young people with neurodevelopmental disorders such as children with autism (Wiggs & Stores, 2004).

2.2 SLEEPLESSNESS IN CHILDREN AND ADOLESCENTS WITH AUTISM SPECTRUM DISORDER

The following section provides a brief introduction to the main features of autism. It is necessary for a better understanding of the issues that will be examined. Prevalence and causes of sleeplessness in this population will be also discussed.

2.2.1 Autism Spectrum Disorder

Autism is a pervasive development disorder (PDD) that was first documented by Kanner (1943). The criteria listed in the Diagnostic and Statistical Manual Text Revision or DSM-IV TR (APA, 2000) to determine whether an individual can be said to have autistic disorder are the following: a) impairment in reciprocal social interaction; b) impairment in verbal and non verbal communication; and c) restricted repetitive and stereotyped patterns of behaviour, interests and activities. Delay or abnormal functioning in these areas have onset prior to age three for a diagnosis of autism.

Although widely and increasingly employed, DSM-IV (APA, 2000) is not the only diagnostic system that has been used by researchers and clinicians. Another major diagnostic system frequently used is the tenth edition of the International Classification of Diseases (ICD-10). The criteria used in this system shows remarkable similarity with those of DSM-IV. Autism is normally diagnosed in childhood with predominance in male subjects. Early studies into what proportion of the population might have autism gave a prevalence rate of about 4 per 10,000 (Lotter, 1966; Wing & Gould, 1979). However, Rutter (2005) in a review of more recent epidemiological studies of autism cited a prevalence of somewhere between 30 and 60 cases of autism per 10,000 of the population.

Many experts believe that autism may not result from a single cause. For instance, there is strong evidence to suggest that autism can be caused by genetic factors, however, it is unclear which genes are responsible for its insurgence and experts believe that multiple genes might be the cause of it (Freitag, 2007). Many other causes have been proposed, such as the exposure of children to vaccines; these proposals are controversial and the vaccine hypotheses have no convincing scientific evidence (Rutter, 2005).

A condition closely related to autism is Asperger syndrome or AS (Asperger, 1944) which is also a PDD (DSM-IV lists under the heading of PDD Autism, Asperger syndrome, Rett's syndrome, childhood disintegrative disorder, pervasive developmental disorder not otherwise specified which are all disorders usually first diagnosed in infancy, childhood, or adolescence) and is differentiated from Autism by the absence of a clinically significant delay in language or cognitive development before the age of three. For this reason it has been argued that Autism and AS belong to the same condition but have a different degree of severity. Thus, taken together, they constitute a continuum of states known as autism spectrum disorders (ASD) to which this thesis will refer when speaking of both conditions.

Apart from the main characteristics of Autism, a number of secondary features may co-occur, such as aggression, hyperactivity, inattention, psychosis and sleep difficulties.

2.2.2 Prevalence of sleeplessness in children and adolescents with autism spectrum disorder

Although sleeplessness is common in TC and adolescents, higher rates are reported for those with autism and Asperger syndrome. Studies have suggested that between 44% and 89% of people with autism experience some sleep difficulties (Patzold, Richdale, & Tonge, 1998; Polimeni, Richdale, & Francis, 2005; Richdale, 1999; Richdale & Prior, 1995; Richdale & Wiggs, 2005; Wiggs & Stores, 2004) although rates change according to the age range of the population affected, whether the problem was reported to be in the past or present, and also based on the type of sleep difficulties. Sleep problems in children with autism seem to be more prevalent in younger children (Richdale & Prior, 1995) and to be generally related to difficulties initiating and maintaining sleep. In a retrospective study of 88 children with autism in Tokyo, sleep disturbances were reported in 56% of the individuals and 90% of those problems started in infancy (Taira, Takase, & Sasaki, 1998). Studies showed that children with autism are likely to experience severe sleep disturbances during childhood, particularly when they are under 8 years of age, and also that they continue to have problems with sleep onset and maintenance when older (Richdale & Prior, 1995; Wiggs & Stores, 2004). However, age has been inconsistently associated with sleep difficulties in children with ASD. Some studies reported sleep as being more problematic in younger children than older children with autism (Inanuma, 1984; Takase, Taira, & Sasaki, 1998) which opposes other studies that show older children having more reported sleep problems than younger ones (Honomichl, Goodlin-Jones, Burnham, Gaylor, & Anders, 2002). Furthermore, others have found no age differences, suggesting that problems continue throughout childhood (Patzold et al., 1998; Schreck & Mulick, 2000). The reason these studies came to different conclusions might be due to the use of different samples. For instance, Inanuma (1984) and Takese et al. (1998) used a sample of autistic children whereas others such as Honomichl et al. (2005) sampled children with PDD.

When current and past sleep problems were compared between children with typical development and ASD, it was found that significantly more children with ASD than children in the control group had a sleep problem both currently and in the past. 63.2% of children with ASD and 22.9% of TC had current sleep problems whereas 76.3% of children with ASD and 37.1% of TC had past sleep problems (Patzold et al., 1998). Also, Allik (2006) found a difference between TC and children with ASD on the basis of past and present sleep problems. More difficulties initiating sleep and more daytime sleepiness was found during the previous six months in 32 children with HFA than in the 32 controls. Furthermore, the prevalence of current sleep problems were significantly higher among the children with ASD than in the control group (19 children with ASD vs. 3 controls). In addition, Richdale and Prior (1995) reported that 55.6% of children with HFA had a severe sleep onset and maintenance problems in the past compared to 15.4% of the control group. With regards to present sleep problems, they reported that 44.4% of children with HFA had a current sleep problem against 26.9% of control children. Also, HFA children slept less and woke earlier than the comparison group. Altogether, 89% of HFA children had a current sleep problem and/or had one in the past.

Regardless of whether or not sleep problems were reported to be present or past, it has been argued that differences between TC and children with ASD may be due to the tendency that parents of children with ASD are inclined to overestimate sleep problems (Schreck & Mulick, 2000) In fact, 54.5% of children with autism were regarded by their parents as having abnormal sleep patterns; on the contrary, objective sleep measures derived from actigraphy did not find differences between autistic and non-autistic children, with the exception of earlier morning waking which occurred more in children with autism (Hering, Epstein, Elroy, Iancu, & Zelnik, 1999). However, Wiggs and Stores (2004) showed that sleep patterns measured objectively did not differ between autistic children with or without reported sleeplessness, though the sleep quality of all children seemed to be compromised when compared with the normal value.

With respect to the types of sleeplessness, as has previously been mentioned, the most frequently reported type in children with ASD appears to be difficulty in initiating and maintaining sleep with some studies reporting also early morning waking (Richdale & Prior, 1995) Several studies have estimated the prevalence of settling problems or initiating sleep in children with ASD to be between 26% and 82% (Hoshino et al., 1984a; Richdale & Prior, 1995; Taira et al., 1998). Patzold and colleagues (1998) showed that children with ASD had significantly longer sleep latency than the control group but woke during the night on approximately the same number of nights as did the control groups; rates were reported to be 23.3% of night waking in children with ASD and 20.8% in the control group. Despite the fact that this study showed no difference between groups in the number of night waking, children with ASD appear to have a longer period of night waking. In fact 18.5% of HFA children have been reported to spend more than an hour awake on one or more nights whereas no control children have been recorded as doing so (Richdale & Prior, 1995).

In addition, another study found that after difficulty with falling asleep, the most common problem in children with autism was awaking during the night with the rate reported to be 21.6% (Taira et al., 1998). Also, a study which compared children with autism with TC and with children affected by a different clinical condition found that the highest percentage, 73%, of sleep problems was in the autism group and the most frequently described problems were settling at bedtime or co-sleeping (Cotton & Richdale, 2006).

The fact that all these studies arrived to similar conclusions supports the notion that such sleep problems are specific to ASD (Patzold et al., 1998) and are not related to IQ level (Richdale & Prior, 1995); except for waking at night which has been found to occur much more frequently in autistic children with mental retardation (Williams, Sears, & Allard, 2004). It is not clear why children with autism and mental retardation have more frequent night waking. This might reflect an issue of discomfort (e.g. need to go to the toilet, feeling cold) with which children with intellectual problems are not able to solve on their own but need parental help. Polimeri et al (2005) reported the highest prevalence of sleep problems, 73%, in children with ASD compared to the control group but no significant differences on severity or type of sleep problems. Settling problems were reported in 72% of TC; 69% of children with autism and 68% of children with Asperger Syndrome (AS); night waking problems were found in 53% of TC, 51% of children with autism and 45% of children with AS; early morning waking and co-sleep were found respectively in 14% and 25% of TC, 38.5% and 29% of children with autism and 25.5% and 36% of children with AS.

With respect to adolescents with ASD, high rates of sleep problems are still reported. Elia et al. (2000) completed a polysomographic study which showed a significant reduction in total sleep time (1 hour and 13 minutes) between children with autism aged 5 to 16 and a control group. In a further study of individuals with autism aged 12-24 years, sleep efficiency (sleep as the percentage of time in bed) was reported to be 81%, compared to 92% for controls (Diomedi et al., 1999). Although, a study with persons with ASD aged 15-25 years reported, on the basis of objective actigraphic measurement, 80% of sleep problems such as too little sleep, early morning waking and difficulties in falling asleep, surprisingly parents and caregivers reported only minor sleep problems (Oyane & Bjorvatn, 2005). This suggests an adaptation process, or a change in nature of behaviour during wakefulness such that it doesn't affect carers as much, rather than an actual reduction in sleep disturbances in adolescence. Table 2.2 shows some of the more representative studies reporting prevalence of sleep problems in children with ASD.

Table 2.2 Prevalence of sleep problems in children with autism spectrum disorder

Studies	**Subjects N**	**Age range (years)**	**Sleep problems (%)**
Patzold et al. (1998)	38	4-12	76
Wiggs and Stores (2004)	69	5-16	64
Polimeri et al. (2005)	105	2-17	73
Richdale and Prior (1995)	39	2-19	75

2.2.3 Causes of sleeplessness in children and adolescents with autism spectrum disorder

Several hypotheses have been offered to explain sleep disturbances in children with autism. Physiological studies have revealed that *serotonergic neurotransmission abnormality* could be associated with sleep disturbance in people with ASD (Nir et al., 1995; Segawa & Nomura, 1992). However, it is important to note that the role of serotonin (5-HT) in sleep regulation is not firmly established. As Jouvet (1999) argued, early studies indicated that 5-HT is a neurotransmitter responsible for transmitting nerve impulses in the brain inducing sleep and tranquillity, whereas other studies indicate that serotonergic neurons also play a role in inhibiting sleep. In the majority of cases, elevated levels of serotonin in autistic individuals has been reported. Studies in which 5-HT was measured found higher levels of 5-HT in High Functioning Autistic (HFA) children than in children without autism (Singh, Singh, & Warren, 1997). Although the relationship of 5-HT to autism has been investigated, the implications remain unclear due to conflicting findings. In fact, low levels of Tryptophan (an acid precursor to serotonin) have also been documented in autistic children (Hoshino et al., 1984b).

Another group of studies have suggested that *abnormalities in melatonin regulation* can be related to sleep problems in children with autism(Chamberlain & Herman, 1990; Nir et al., 1995; Rapin & Katzman, 1998; Ritvo, Ritvo, Yuwiler, Brothers et al., 1993). As mentioned in section 1.1.4, melatonin seems to act as a time signal in the regulation of sleep-wake cycles and seasonal rhythms. Alterations in the melatonin rhythm may be responsible for sleep onset and maintenance problems in autism. More specifically those with sleep onset problems may have a melatonin rhythm with peaks later in the night whereas reduced rhythm amplitude may cause night waking and early-morning waking (Richdale, 1992).

Chamberlain and Herman (1990) first drew a theoretical model suggesting that autism may reflect dysfunction of the pineal-hypothalamic axis resulting in a hypersecretion of pineal melatonin. Nir et al (1995) compared a group of 10 young adults with autism and a control group with respect to the level of melatonin. Results showed a disturbance in the functional rhythm of melatonin. Autistic patients had significantly smaller amplitude of change. The serum level of melatonin in the patients was higher than in the control group during the day and lower than in the control group during the night. Similar findings were reported by Ritvo et al. (1993) who measured levels of melatonin overnight and first-voided urine samples from people with autism and healthy controls. Those with autism had significantly higher melatonin concentrations in the first urine sample, but none in the overnight sample.

A further area of interest with regard to sleep difficulties in children with autism is the sleep EEG studies; however investigations in this field are limited. Diomedi et al (1999) reported abnormalities of sleep phases and the presence of REM sleep components in non-REM sleep in mentally retarded autistic children.

Polysomnographic recordings of sleep in children and adolescents with ASD have revealed a decreased total sleep time and a greater number of muscle twitches compared with healthy controls; also persons with ASD showed REM sleep latency and a lower proportion of stage 1 sleep than those with Fragile X syndrome and mental retardation (Elia et al., 2000). Laboratory sleep recordings for two consecutive nights were obtained from 16 young adults with ASD and from 16 control participants. Objective data showed that persons with autism presented longer sleep latency, frequent nocturnal awakenings, increased duration of stage 1 sleep, decreased slow wave sleep and fewer EEG spindles (Limoges et al., 2005). These conclusions are similar to those found by Goudbout et al. (2000) who observed two interesting characteristics in people with AS in the age range 7-53: abnormalities of REM sleep, and low incidence of sleep spindles (these can also been seen in association with learning disability). Sleep spindles are thought to represent a sleep protective mechanism by which the inputs to the brain are filtered. This is congruent with selective attention atypicalities found in people with ASD.

The sleep difficulties experienced by children with autism may be related not only to physical abnormalities but also to severe *social problems*. The sleep-wake cycle is a circadian rhythm and is primary regulated by a light-dark cycle. However, sleep and wake time can be also guided by social cues. Deficient perceptions of social and environmental cues may impede the ability of autistic children to appropriately synchronise their sleep/wake rhythm. Children with social-communication problems may find it particularly difficult to use such cues to entrain their rhythm (Richdale & Prior, 1995).

A further hypothesis regarding sleep difficulties in children with autism may be a relation to *arousal factors* (Richdale & Prior, 1995). Literature suggests (Patzold et al., 1998) that children with autism are generally more energetic and less sleepy at bed time and also HFA children are reported to wake more spontaneously at a fairly typical time despite a shorter time sleeping and night waking. This suggests that children with autism may be physiologically hyperaroused. Also, early studies showed that the occurrence of stereotyped behaviours in autistic children served an arousal-reducing function since environmental complexity and novelty generate an increase in physiological arousal in those children (Hutt & Hutt, 1968). This suggests an association between disturbed night sleep and the need to modify stimulation through stereotypical behaviour (Johnson, 1996). Allik et al. (2006), drawing on their findings, speculated whether cognitively over-aroused mental activity such as certain intrusive repetitive thoughts about sleep or school work, delayed the initiation of sleep in HFA and AS children.

Fear and anxiety may also be a cause of sleep problems, particularly in the older and higher functioning children with autism or Asperger syndrome. Autobiographical reports by such children refer to feelings of anxiety and fear (Volkmar & Cohen, 1985). Therefore anxiety may significantly contribute to sleep problems in more able children with autism since they may be more likely to think deeply about any fear (Richdale, 1999). Children with autism and AS have also been found to have significantly higher scores on the anxiety subscale of the Developmental Behavioural Checklist and State-Trait Anxiety Inventory than comparison children (Limoges et al., 2005; Patzold et al., 1998). Also, anxiety-related behavioural features such as verbalized fears or physiological signs of anxiety at bed time, were prominent in parents' reports (Wiggs & Stores, 2004). These findings are supported by Allik et al. (2006) who found that more parents of children with ASD reported their child to have anxiety at bed-time than in the comparison group.

As in the general population, the most common sleep disorders underlying the symptoms of sleeplessness appear to be *behavioural in origin*. This means that sleeplessness is in the majority of cases is caused or maintained by the fact that children have not learnt appropriate ways, or have learnt inappropriate ways of getting to sleep or staying asleep (Wiggs & Stores, 2004). Wiggs and Stores (2004) explored the sleep of 69 children aged 5 to 16 years with ASD by the use of detailed sleep histories, sleep questionnaires, diaries and actigraphy. They found that sleeplessness was often the result of behavioural sleep disorders. This is confirmed by Patzold et al's (1998) results which showed that children with autism displayed more unusual bedtime routines requiring particular conditions to fall asleep (i.e. parents holding the child, lying down with the child, positioning blinds and curtains a certain way) which increased the risk of sleep problems in this population compared to those in the control group.

Children with autism might have obsessive interest for *electronic media* (e.g television programs or video games) and so they might stay awake until late to watch or play them. Excessive television viewing has, in fact, been associated with autism (Waldman, Nicholson, Adilov, 2006) and, as for TC, it might be one of the underlying factors in causing or maintaining sleep problems in this population.

Allik et al. (2006) compared 32 children with AS/HFA aged 8-12 with 32 age and gender matched TC regarding sleep and associated behavioural characteristics. The result suggested that insomnia was a common distressing symptom frequently associated with *coexistent behaviour problems* in children with AS/HFA. They also investigated if children with insomnia (difficulties initiating and maintaining sleep) differ from children without insomnia regarding autistic symptoms and general behavioural problems. The authors found that children with insomnia in the AS/HFA group had more autistic, emotional, hyperactive symptoms than those children without insomnia. This result resembles the finding of Schreck et al. (2004) that sleep problems predicted more intense symptoms of autism. In this study, the sleep problem best predicting the diagnostic characteristics of autism was the reported amount of hours per night the child slept. The fewer hours slept the more severe the reported symptom of autism (e.g. stereotypical behaviour, social difficulties).

2.3 EFFECT OF SLEEP PROBLEMS ON CHILDREN AND PARENTS

The following section reviews the evidence regarding the effect of sleep problems on daytime functioning and creativity in TC and in children with autism. The impact that such disturbances can have on parents' mental health is also discussed.

2.3.1 The impact of sleep loss on neurobehavioural functioning (NBF) of typical developing children and children with autism spectrum disorder

The effect of sleep loss in adults has been the focus of considerable research. Several studies have found that in adults, sleep deprivation can result in deficits in cognitive performance such as attention, concentration, memory and reaction time (Kingshott, Cosway, Deary, & Douglas, 2000) and a few studies have found that lack of sleep impaired high mental abilities such as creativity in adults (Horne, 1988). In contrast, minimal efforts were made in exploring these issues among TC (Sadeh, Raviv, & Gruber, 2000). This neglect is intriguing considering that a high prevalence of sleep loss has been documented by use of objective sleep measures in school age children (Sadeh et al., 2000).

One of the few longitudinal studies examining the relationship between sleep and cognitive outcomes in children found that those who had more rhythmic sleep patterns at 7 and 19 months of age had better mental development scores at 24 months and better language scores at 36 months (Dearing, MacCartney, Marshall, & Warner, 2001). This result suggests that sleep patterns early in life may have an underlying role in the subsequent cognitive and behavioural development. The relationship between sleep problems and daytime functioning over time has also been investigated in an study carried out by Gregory and O'Connor (2002) with 450 children aged from 4 to 5 years who were followed over an 11 year period. It was found that sleep problems correlated with behavioural problems including anxiety, depression, aggression and attention problems.

There is also a small number of sleep deprivation studies which have examined the effect of lack of sleep on neurobehavioural and cognitive functioning in school age children. For instance, Copes and Rosentswieg (1972), found descreases in psychomotor performance following total sleep deprivation in children. Carskadon, Harvey and Dement (1981a, 1981b) studied the effects of sleep restriction (1 night of 4 hours of sleep) and sleep deprivation (1 full night) and found performance decrements only after the full night of sleep deprivation. This finding suggests that sleep restriction may not always lead to detectable cognitive impairment as does total sleep deprivation. However, Ishihara (2002) found that when sleep was shortened for two hours in three children aged 7, 9, and 13 years sleepiness the following day increased significantly in all three children. Also Randazzo et al. (1998) reported decrements in performance of complex cognitive tasks following a single night's restriction of 5 hours in bed. The results of these studies demonstrate not only that even small amounts of sleep loss can result in significant impairments and disturbance of daytime functioning but also that difficult tasks such as those involving the use of creativity are more affected by lack of sleep than simple tasks.

Another perspective on the relationships between sleep and NBF is based on sleep fragmentation studies in which a person is experimentally awakened multiple times for a brief duration. It has been suggested that the adverse effects of sleep fragmentation result from the increase of less restorative sleep and the relative decrease in deeper and more restorative sleep stage (Wesensten, Balkin, & Belenky, 1999). Sadeh et al. (2002) showed the adverse effects of multiple night wakings and reduced sleep quality on subsequent daytime functioning in school age children. The authors examined sleep using objective sleep recording (actigraph) for five consecutive nights in a sample of 135 children aged 7 to 12 years. NBF was evaluated using a battery of standard neuropsychological tests (e.g. finger tapping task, simple reaction time, memory and learning tasks) and child behaviour was assessed using a parent completed checklist. In this study, it was found that fragmented sleep was associated with both difficulty in behavioural inhibition and sustained attention.

Thus, lack of sleep in children seems to be associated with impairment of a child's daytime functioning. Examining sleep patterns in children aged from 7 to 9 years has indicated that reduced objective sleep time is associated with teacher and parent reports of negative behaviour (Aronen, Paavonen, Fjallberg, Soininen, & Torronen, 2000) impaired working memory functioning (Steenari et al., 2003) daytime sleepiness and depressed mood (Wolfson & Carskadon, 1998) and deficit in school performance (Hofman & Steenhof, 1997). However only a few studies have looked at the effect of sleep extension on children's daytime functioning. In a recent study (Sadeh et al., 2003), the sleep of 77 children aged 9 to 12 years was manipulated with children being randomly allocated to a sleep restriction condition (where sleep was shortened by at least 30 minutes of total regular sleep time) or a sleep extension condition (where sleep was extended for at least 30 minutes of total regular sleep time). Results of this study showed that children in the extension group condition had significant neurobehavioural improvement on several tasks on the following day compared to the sleep restriction group, which showed impairment on tasks such as reaction time and memory testing. However Allen (2003) has argued against the validity of the findings by Sadeh et al. (2003), suggesting that a more conservative statistical analysis of the data would show no significant differences in NBF following sleep restriction. Nevertheless Allen (2003) partially replied to Sadeh's study (2003) by reporting significant improvement in NBF in children aged 6 to 11 who experienced sleep extension for 30 minutes. These studies have important clinical implications, suggesting that an increase in sleep time by as little as 30 minutes is beneficial for children's NBF.

Despite the research relating sleep deprivation to cognitive impairment and inappropriate behaviour in TC, the influence of sleep problems for children with autism has not been sufficiently investigated. As explained in section 2.2.3, the limited research available suggests a few general relationships between sleep problems in children with autism and their behaviour. Hoshino et al. (1984a), by use of parental sleep questionnaire, examined the sleep patterns of 75 children with autism aged 3 to 11 years and divided them into poorly mentally developed and well mentally developed children on the basis of a developmental test. The author found that those children who were poorly developed often showed a high rate of sleep disorder, with a long duration period of sleep disturbance compared with well-developed autistic children. These facts suggested that a) sleep disturbance of autistic children might be closely related to the severity of their problem or prognosis, and b) sleep problems aggravate clinical symptoms of autism such as isolation or hyperactivity. Patzold et al. (1998) reported similar conclusions and suggested that problematic daytime behaviour in children with a PDD may be associated with the presence of sleep difficulties. In fact the authors found that children with PDD had significantly more anxiety, communication problems and disruptive behaviour than the non-autistic control group. In recent years Hoffman et al. (2005) examined the relationship between sleep problems detected by "The Children's Sleep Habits Questionnaire" (CSHQ), (Owens, Spirito, & McGuinn, 2000) and the level of autism through "Gilliam Autism Rating Scale" (GARS) (Gilliam, 1995). Parent's reports of their children's sleep problems were related to scores obtained on the GARS. In particular, sleep disordered breathing was shown to be the best predictor for children's stereotyped behaviour, social interaction problems and overall level of autism. The other factor contributing to the prediction of GARS domain was children's parasomnias which was the primary predictor in the analyses for developmental disturbances. The fact that lack of sleep exacerbates autistic symptoms was also showed by Schreck et al. (2004) who found that reduced hours of sleep was the primary variable predicting children's overall autism scores. These results are consistent with those of Hoffman et al. (2005) in demonstrating a relationship between children's sleep problems and the autistic behavioural problems.

The foregoing relationship among sleep problems for children with autism and daytime inappropriate behaviour suggest that additional research is required to delineate direct connections among the specific sleep problems and the specific daytime behaviour.

A similar relationship between the presence of sleeplessness and elevated rates of daytime problems has been reported in many others groups of children with various developmental disorder (Wiggs, Montgomery, & Stores, 2005) and is corroborated by teacher reports (Wiggs & Stores, 1999) suggesting that this relationship is not due to a reporting bias.

Despite good evidence that sleep problems can have a negative impact on children's development, studies examining these relationships in typical and atypical childrena share common limitation. First, the majority of these studies are correlational in nature. There is limited information about the direction of causation of sleep problems and daytime behaviour. In addition, the relationship between sleeplessness and daytime behaviour is not as direct as one might imagine. On one hand, it appears that sleep problems in TC causes cognitive and behavioural difficulties during the day, while on the other hand it might be that other factors besides an objective lack of sleep (e.g., behavioural confrontations with the parents in the evening and the ways in which parents deal with this) might cause sleep difficulties in children. Wiggs and

Stores (1996a) asserted that it is difficult to determinate if the sleep problems of children with daytime behavioural difficulties cause the behaviour problems, maintain existing problems or exacerbate problems already present. For instance, it is not clear if the high rate of sleep problems found in children with autism is due to a delayed development of good sleep patterns or inappropriate daytime behavioural functioning. Secondly, studies have looked at heterogeneous categories of children in terms of important variables. For example, studies have included children aged from a few months to those in their teens and also have addressed groups of children with various neurodevelopment disorders. Thirdly, sleep deprivation or restriction studies in children have typically lasted for a few nights at most and lack of external validity given that they did not consider the impact that the sleeplessness problem has in real life such as on the family unit. Fourthly, the majority of studies have relied on subjective data regarding sleep and behaviour, mainly involving parental reports. Fifthly, studies have rarely taken into account children's report of their own sleep problems symptoms. Finally, there is very little research about the relationship between lack of sleep and high mental ability such as creativity in TC and there are no studies that have investigated this link in children with HFA.

2.3.2 Creativity and sleep in typical developing children and children with autism spectrum disorder

Creativity can be defined in different ways depending of the perspectives used to investigate it. Creativity has been defined as divergent thinking (Guilford, 1950; Torrance, 1962) as a characteristic of personality (Barron & Harrington, 1981) as a property of cognitive processing (Runco & Sakamoto, 1999) and as an ability to solve problems (Gardner, 1993). With respect to the developmental aspects of creativity, children's creativity seems to be associated with imagination and play. Creative play is of great importance in the development because it stimulates curiosity, flexibility and promotes problem-solving behaviour that leads to learning.

Impoverished imaginative creativity has been referred to as a key symptom in the standard diagnostic criteria for autism (APA, 1994) and a study found a deficit in children with autism using a standardised measure of creativity (Craig & Baron-Cohen, 1999). Different theories have tried to explain the creativity deficit seen in autism. The *executive dysfunction theory* (Leevers & Harris, 1998) suggested that the lack of imaginative creativity found in children with autism is due to executive problems in disengaging from routine actions and from reality and predicted impairment, both on imaginative and reality based creativity as opposed to the *imagination deficit theory* (Scott & Baron-Cohen, 1996) which suggested that children with autism are impaired only on imaginative creativity.

There is little doubt from historical and anecdotal accounts that imagination plays a central role in creative functioning, and research in creative cognition has provided experimental evidence of this (Finke, 1990). However there is a need for novel measurement to assess creativity by examining the use that children make of their imagination. There is, in fact, a need for new tests to evaluate both verbal and graphic creativity in typical and atypical children.

Although there are no doubts that sleep plays an important role in the cognitive process, few studies have attempted to determinate what are the empirical links between sleep and creativity in adults and TC. Furthermore, no current studies have looked at the relationship between creativity and lack of sleep in people with autism. Horne (1988) investigated whether divergent thinking was impaired in adults by 1 night of sleep loss and found that sleep loss had a substantial impact on most aspects of divergent thinking compared to the control condition. Increased perseverance was clearly apparent with fixation on previously successful strategies when subjects attempted solution to new problems.
Interestingly, it has been shown that REM sleep in particular is an enhancing factor for divergent thinking and so this is beneficial in situations in which originality and creativity are called for. For instance, Lewin and Glaubman (1975) found that REM deprivation was detrimental in those tasks in which creativity and originality were required. Gulford's Utility Test (1967) was employed with scoring providing a good evaluation of the amount of divergent thinking, flexibility, originality and creativity, but beneficial in memory tasks learnt by rote. Even though Lewin and Glaubman's experiment was carefully balanced, with equal number of wakings, the same length of time being awakened and the total amount of sleep in both experimental and control nights, the order effect was not balanced with the REM deprivation night always preceding the control night. Thus, it can be speculated that poor results of the creative

measures following the REM deprivation nights were due to the order effect but not to the removal of REM sleep *per se*. However, in 1978 this experiment was replicated and extended, with similar experimental conclusions. In addition to balancing for order effect, new cognitive tasks were employed and subjects were sampled from different age groups. The study thus reconfirmed the hypothesis that REM sleep contributes to divergent thinking and supported the claim that it encouraged the individual's adaptation to new situations (Glaubman et al., 1978).

Based on the assumption that dreams occur especially during REM sleep, some studies have also provided empirical support for a link between creativity and sleep-dreams in adults (Domino, 1976, 1982). However a current review of dream function has de-emphasised the role of dreaming in an individual's creative process (Moffitt, Kramer, & Hoffman, 1993). Early studies (Domino, 1976, 1982; Sladeczek & Domino,1985) and current research (Pagel & Kwiatkowski, 2003) which support the hypothesis that sleep-dreams enhance creativity can be criticised for two reasons: a) because sleep and dreams were evaluated exclusively by subjective measures; and b) because of the difficulty of assessing dreams.

To overcome these problems a recent experimental study has used an objective measure of sleep demonstrating a clear association between creativity and REM sleep (Walker, Liston, Hobson, & Stickgold, 2002). This study investigated the difference in cognitive flexibility (this is thought to be employed in problem solving and creative thinking), using anagram word puzzles, during waking trials and following awakenings from nocturnal REM and NREM sleep which were identified through polysomnographic (PSG) recording. Anagram performance significantly differed across the sleep-wake cycle with overall performance (such as number of anagrams solved) following REM awakenings greater than that from NREM. Thus, it can be speculated that there are changes in cognitive flexibility between REM and NREM awakenings. Studies have in fact demonstrated that aminergic neurotransmitters, which are high in situations of stress and anxiety and cause impairment in cognitive flexibility (Toren et al., 2000), are inhibited during REM sleep (Beversdorf, Hughes, Steingerg, Lewis, & Heilman, 1999). Interestingly, participant performance after awakening from REM sleep was not better than participants who stayed awake during the day, indicating that in REM sleep, there is an alternative (but just as effective) mode of problem solving that differs from the mechanism available while awake. Thus, REM sleep state, lacking in aminergic dominance, is more conducive to fluid reasoning and hence better ability to perform cognitive tasks requires flexible thought. One might argue that the advantage in performance observed following REM awakenings compared to NREM awakenings is simply due to a difference in general brain activation between these two conditions. However in Walker et al.'s (2002) study, the average reaction time (this is known to be a robust indicator of arousal level) did not differ between REM, NREM sleep and wakefulness while the number of anagrams solved was significantly different.

It could be also argued that a true evaluation of the relationship between creativity and REM sleep is not possible since certain amounts of the sleep state properties will have been lost as a consequence of waking. However in Walker et al.'s (2002) study tests were administered immediately following awakenings from specific sleep stages when the brain still displayed similar neurophysiologic properties to the prior sleep state (sleep inertia). In addition, there are early studies that support the hypothesis that creative ability is more effective following REM than NREM sleep (Been, 1997; Broughton, 1982) despite one recent review that argued against the concept of such flexible thinking during REM sleep (Revonsuo, 2000).

Other studies have examined the relationship between creativity and total or partial sleep deprivation regardless of sleep stage parameters. For instance, a number of studies show that subjects deprived of either REM sleep or SWS before (Johnson, Naitoh, Moses, & Lubin, 1974) or after (Lubin, Moses, Johnson, & Naitoh, 1974) nights of total sleep deprivation, showed no significant difference in mood, anxiety and cognitive tests upon awakening. Thus, total sleep time and its quality, rather than specific stages, seems to be of particular importance for the restoration of superior cognitive functions (Johnson, 1973). These conclusions confirmed a previous study in which tasks requiring complex and divergent thinking, under the control of the prefrontal cortex (PFC), showed the most pronounced sleep deprivation deficits (Horne & Pettitt, 1985). A recent study also (Wagner, Gais, Haider, Verleger, & Born, 2004) showed that sleep, by restructuring newly acquired mental representation of a task, facilitated a process of insight. In this study participants were asked to reduce a string of digits, using two simple rules, to a single digit (Number Reduction Task). Out of three groups of participants (those who slept, those who stayed awake during the day, and those who stayed awake during the night), those who slept eight hours were twice as likely during retesting to gain insight into a hidden rule built into the task.

When looking at the literature review about the link between sleep and creativity it is striking that very little attention has been paid by researchers to this relationship in children. While there is some sleep research that has investigated the effect of lack of sleep on neurobehavioural functioning (e.g. attention, memory, behaviour) in TC and in children with autism (see section 2.3.1) there are currently only two studies that have specifically looked at the association between sleep loss and creative ability in TC (Randazzo, et al.,1998; Healey & Runco, 2006) and there is no published work with either adults or children with autism on this topic. This is surprising given the considerable importance of creativity in the adaptation of children to new situations and in the development of autonomy and independence (Albert and Runco, 1988).
Randazzo et al (1998) showed that verbal creativity and abstract thinking in children between the ages of 10 and 14 years were affected by one night of restricted sleep with only 5 hours in bed. The performance of the sleep restricted group was significantly worse in the areas of fluency, originality and average on the verbal version of the 1990 Torrance Test Creative Thinking (TTCT) test, while TTCT figural subscale scores for the control group were higher than scores for the sleep restricted group but not at a level of significance. The results found in this study are consistent with the interpretation of deficits in the executive/frontal lobe function following sleep deprivation in adults (Horne & Pettitt, 1985). Thus, a) damaged executive function which enables individuals to engage in creative learning (Wagner et al, 2004), and b) impaired creative problem solving and verbal processing which are heavily dependent upon the frontal lobe are associated with sleep deprivation in children (Dahl, 1996). However, Randazzo et al.'s (1998) study did have some methodological problems. As for all human sleep deprivation research, the experimental manipulation was not blind. This might have caused the outcomes to be influenced by either the placebo effect or observer bias. In addition, no objective measure of sleep prior to the experimental night was made, although subjects reported compliance with instructions to obtain at least 10 hours in bed on the previous three nights.
One study with TC (ages 10-12) in New Zealand reached a different conclusion (Healey & Runco, 2006). This research looked at the incidence of sleep disturbances

in 30 highly creative children when compared with 30 control children. Creativity was measured using the TTCT figural form (1974) and measures of sleep problems were obtained using the Child Depression Inventory (Kovacs, 1992) and Revised Child Manifest Anxiety Scale (Reynolds & Richmond, 1985). The authors showed that there was a significant difference between the two groups, with the creative children reporting more sleep disturbance, therefore suggesting that creative ability may indeed affect an individual's sleep patterns. More specifically, out of the 60 children tested on the standard creativity test, 17 of the highly creative children indicated that they had higher levels of sleep disturbance compared to only 8 of the control children. Healey and Runco (2006) is the only study with children to suggest that the creative process contributes to the development of insomnia, since planning and creative problem solving might put children at risk of developing sleeplessness. There are several anecdotal accounts with adults about creative insomnia (for instance film maker Alan Berliner made a documentary on his lifelong insomnia and its complex role in his creative process (HBO, 2007) and there is one experimental work with adults which found no significant association between sleep disturbance and creativity (Sladeczek & Domino, 1985).

However, there are a number of limitations in the study carried out by Healey and Runco, (2006) that need to be considered. Firstly, very elementary measures of sleep disturbance were used. Secondly, the creative measures provided a measure of creative potential rather than creativity *per se*. Thirdly, the small sample size limited the power of these findings. Finally, it was possible that a variable such as anxiety could be responsible for the relationship between sleep and creativity found in this study.

Therefore, given that: a) the absence of literature on this topic especially with different categories of children such as those with autism; b) the absence of studies that have used more robust measures of sleep such as valid and reliable sleep questionnaires, sleep diaries and actigraphic measures of sleep; c) the contradictory results found between the only two studies that have investigated the relationship between creativity and sleep in TC; and finally d) the great importance that sleep and creativity have on the developmental process of children, more research is needed to overcome these limitations.

2.3.3 The impact of child sleep problems on parents' mental health

A clear link between sleep problem occurrence in TC and parental stress has been established. An investigation of the effects of children's sleep disturbances on parents showed problems ranging from fatigue and frustration to marital discord (Armstrong, O'Donnell, McCallum, & Dadds, 1998). This is perhaps most evident in puerperal mothers. A study compared the rating of maternal distress of 47 mothers who had presented for admission to a mother/baby hospital due to their infant's sleep problem, with 50 control mothers who had infants of a similar age. The mothers of children with sleep problems obtained significantly higher scores compared to the control group on the postnatal depression scale (Lam et al., 2003). Similarly a recent study reported that scores on maternal depression were significantly higher in mothers of TC who had a sleep problem as compared to those who had a child without a sleep problem (Gregory et al., 2005; Hiscock & Wake, 2001b). This finding has also been replicated in a many other studies where a higher incidence of depression and anxiety and stress have been identified in the mothers of sleepless children (Reijneveld, Van der wal, Brugman, Hira Sing, & Verloove Vanhorick, 2004).

Periods of sleep deprivation related to parenthood are common in different cultural contexts and occasionally have serious averse effects. In a large study of Dutch parents of 1-6 month old infants, 6% admitted shaking and hitting their child during nocturnal crying episode (Reijneveld et al., 2004). In an smaller study of American mothers, 70% admitted to fantasies of aggression and 20% reported that these fantasies were about killing the child (Levitzky & Cooper, 2000). Also in a large study of Swedish parents of 5 -6 year old sleepless children, 80 mothers out of 361 and 32 fathers out of 272 reported parenthood related sleep difficulties (Smedje, Broman, & Hetta, 1998).

In mothers, the most prominent complaints were early waking, not feeling rested in the morning and day time sleepiness, whereas in fathers, sleep difficulties included difficulties initiating sleep, night waking and not feeling rested in the morning. These studies indicated that some parents of children with sleep problems have a chronic lack of sleep that might possibly cause or contribute to dysfunction in family life.

However associations between parental stress and children's sleep problems have not always been found. For instance, a study reported that maternal stress was associated with settling problems and night waking of children attending mainstream school

(Lam et al., 2003) as opposite to another study where an association between sleep problem of TC and maternal stress was not found (Doo & Wing, 2006).

Similar associations between child sleep patterns and parental mental health have been found in parents of children with autism. In a cross-sectional survey with children with PDD aged 2 to 8 years, parents whose child had sleep problems experienced a higher level of stress than those whose child had no sleep problem (Richdale, 1999). Thus sleep problems are an additional difficulty on families with children with PDD (Patzold et al., 1998) who are more likely to exhibit daytime behaviour problems and daytime sleepiness that may interfere with their behavioural development (Johnson, 1996). In fact, stress levels of parents with children with PDD have been found to be higher than in parents whose children have no developmental delay (Koegel et al., 1992). Parents of children with PDD are particularly concerned about the future of their child including their cognitive impairment and the acceptance of their children in the community (Moes, Koegel, Schreibman, & Loos, 1992). Mothers may be at a greater risk of the negative effects of children's sleep problems compared to fathers. For example mothers of children with autism have been found to have higher stress level than fathers, as they are more often the primary caregiver (2006). Allik (1998) found that in children with autism, mothers' mental health was related to child behaviour: both hyperactivity and conduct problems as well as prosocial behaviour. However paternal health was not found to be associated with any of the child behavioural measurements.

One factor that may mediate the relationship between parental mental state and children's sleep problems is parent coping style. For children with developmental disorder, Wiggs and Stores (1998b) found that coping styles were important in parents' perception of their child's sleep. Parents who had a child with sleep problems but who did not consider the child's sleep to be a problem had lower stress a higher perceived control over the child than parents who recognised their child's sleep as a problem. This suggests that parents who have higher stress and external locus of control may be more affected by their child's sleep issue.

However the relationship between paediatric sleep problems, child daytime behaviour and parents' wellbeing are not fully understood. For instance, a recent study by Wiggs and Stores (2002) found that there were no significant differences between autistic children with and without sleeplessness problems in terms of mothers' levels of sleeplessness, their mental state, and their views on aspects of childhood sleeplessness including their management. Also the two groups were not different in term of autistic symptoms such as anxiety, impairment in language, and social skill. The objective sleep parameters of both groups was impaired in terms of quality and quantity compared to normal values but did not significantly differ from each other. This result suggested that the pattern of maternal and child factors associated with sleeplessness in autistic children is different to that reported in TC and children with learning disabilities. The fact that there was no significant difference between the mental state of mothers of children with and without sleeplessness problems indicates that this may be less important for this clinical group than other child aspects.

The association between parent mental health and sleeplessness problems in typical and atypical children needs to be further investigated. It remains, in fact, unclear whether this relationship is causal, bi-directional or related to other factors. Studies with larger sample size, more objective sleep measures and a deeper analysis of paternal factors are needed.

In conclusion, research evidence shows that childhood sleeplessness impacts negatively on parents and that children with sleeplessness problems are at increased risk for behavioural, emotional, and cognitive disturbances compared to children without sleep problems. In addition, high cognitive ability such as creativity might be compromised by lack of sleep although data are limited. Given the prevalence of sleeplessness in TC and children with HFA, the lack of information about the relationship between sleep and creativity in these children and the impaired creativity characterising autism spectrum disorder, it would be important for future work with TC and children with HFA to assess the links between sleep and children's neurobehavioural functioning and include assessment of creative function.

2.4 TREATMENT OF SLEEPLESSNESS PROBLEMS: GENERAL APPROACHES

The choice of which treatment to use depends of the underlying sleep disorder. For example, sleeplessness problems can arise secondary to a comorbid medical condition, such as epilepsy, result from a sleep disorder with physiological causes such as obstructive sleep apnoea or more psychological causes such as the fact that the child has not learnt appropriate sleep behaviour. Thus, these different causes of sleeplessness problems require diverse kind of interventions which also need to be tailored to the requirements of each child.

Evidence suggests that *medications* are commonly prescribed as a treatment of sleeplessness problems in children (Bramble, 2003). However, the use of sedative-hypnotic medications for sleeplessness may be useful for very short-term relief but it is certainly not a long term solution (Ramchandani, Wiggs, Webb, & Stores, 2000). In addition, hangover effects, paradoxical responses and parental resistance to its use are common (Kuhn & Weidinger, 2000). Melatonin has been found to be helpful for TC and adolescents (Jan, Freeman, & Fast, 1999) and also for children with ASD (Giannotti, Cortesi, Cerquiglini, & Bernabei, 2006) both with sleep-wake cycle disorders. However, the concerns surrounding the use of melatonin must be carefully considered given that long term effects of giving melatonin to children are not well-known (Armour & Paton, 2004).

Chronotherapy or manipulating the time of sleep, is also used in the treatment of children with irregular, delayed or advanced sleep phases who need to be distinguished from children who refuse to sleep or wake up, as the treatment will be different, though the manifestation of the problem is similar. For children with irregular sleep phases it will be necessary to set a consistent routine (e.g. meals, bed time, wake up time) and for those with advanced or delayed sleep phase syndrome it will be generally necessary to move the sleep phase progressively, either forward or backward, until the desired sleep time is reached. Chronotherapy may also involve either morning or evening bright light exposure in order to respectively advance and delay sleep onset. This method has been used successfully with children with intellectual disability and autism in the treatment of sleep-wake cycle disorders (Piazza, Hagopian, Hughes, & Fisher, 1998).

Cognitive treatment for bedtime refusal and night-wakings is based on working with parents to change their sleep-related interactions with their child and thus leading to a behavioural change in their child's sleep patterns (Sadeh, 2005). This method is often used in conjunction with behavioural strategies (the different behavioural techniques are described fully in the next section) and considers parents' cognition essential for the resolution of the child's sleep problems. In fact many parents experience great difficulties modifying their bedtime interaction with their child because they perceive any sort of behavioural restriction as neglectful, abusive or inappropriate. In addition these negative interpretations are often associated with very strong negative emotions such as guilt, shame and anger that serve as negative reinforcers for any change in parenting behaviour (Morrell, 1999). Combined cognitive-behavioural techniques including self-control training, desensitisation, muscle relaxation, breathing exercises, positive imagery training, and positive self-statements have been shown to be effective in the treatment of night time fears in children (Giebenhain & O'Dell, 1984; Graziano & Mooney, 1980, 1982; Ollendick, Hagopian, & Huntzinger, 1991). However, the efficacy of these methods has been demonstrated particularly in case studies and in a limited number of controlled studies. In addition, it is difficult to determine the efficacy of any specific component because most researchers have used a combination of techniques.

There is a lot of evidence that the most common childhood sleeplessness problems are best treated by 'teaching' children good sleeping habits. These types of treatment are called *behavioural treatments.* Behavioural interventions are based on the principles of learning theory which involves shaping and modifying children's behaviour in small steps by rewarding appropriate, and ignoring undesirable, conduct. Behavioural treatments for sleep problems include extinction, graduated extinction, positive bedtime routines, scheduled awaking. Techniques have been delivered by therapists face to face or in written form in a parent self-help booklet. These methods have been empirically validated for the treatment of bedtime problems and night wakings in TC. A recent review of 52 studies investigating behavioural interventions for these problems in children aged 0-5 years reported that 94% of studies showed clinically significant effects (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006).

Success does not appear to be limited to TC from the general population. A number of case studies (Wolf et al., 1964; Howlin 1984; Weiskop et al., 2001) and also multiple baseline design studies (Weiskop et al., 2005) have showed that behavioural interventions are effective in the treatment of sleep problems in children with autism.
In addition, parent education has been shown to be an effective behavioural method to prevent children's sleep problems (Kerr, Jowett, & Smith, 1996; Mindell et al., 2006; Moore, Meltzer, & Mindell, 2007). Via written material or in educational groups, parents are taught how to help their children to develop self-soothing skills, how to

handle the child during sleep initiation and how to respond to night-time waking. Almost all these prevention programmes give the recommendation that children should go to bed when they are drowsy, but awake, to help them develop independent sleep (Mindell et al., 2006). Interestingly, Wolfson et al (1992) found that 72% of 3 week old infants, whose parents attended a sleep educational group before and after birth, slept all through the night compared to 48% in the control group.

Despite behavioural interventions being found to be effective in the treatment of childhood sleeplessness, more research needs to be carried out to determine the efficacy of behavioural interventions in the treatment of sleep problems in older children with and without autism.

2.5 BEHAVIOURAL TREATMENT FOR SLEEPLESSNESS IN CHILDREN WITH AND WITHOUT AUTISM SPECTRUM DISORDER

Behavioural treatments for sleep problems which are thought to be due to the child not having learned appropriate sleep habits or for those where the child has explicitly learnt inappropriate sleep habits are receiving increasing attention. As mentioned previously there are several behavioural techniques to help reduce sleep problems in children which are based on the notion that inappropriate sleep habits in childhood can be improved by identifying the factors that reinforce them and substituting these with more appropriate sleep behaviour. The efficacy of behavioural approaches for sleeplessness have been suggested with TC (Mindell et al., 2006) and children with autism (Schreck, 2001) as well as with other developmental disorders (Wiggs & France, 2000).

The following sections describe the principal behavioural techniques in the treatment of childhood sleeplessness.

2.5.1 Extinction

The common term used in the media to describe extinction is the "cry it out" or "ignoring" approach (Ferber, 1985). Extinction, or systematic ignoring, is one of the earliest behavioural interventions for bedtime problems and night wakings in children and continues to be recommended today (Mindell et al., 2006; Morgenthaler et al., 2006). Extinction involves putting the child to bed at a consistent time and ignoring the child's negative behaviour, such as yelling and crying - with the exception of any concern that the child is hurt, ill or in danger. It is believed that after the child protests for a while, the exhaustion and the absence of parents' presence, will lead to sleep onset and self-soothing (Sadeh, 2005).

The first study of systematic extinction was conducted by Williams (1959) with a toddler whose crying decreased after consistent ignoring by the parents. Since that time extinction has been demonstrated as an effective treatment for settling and night-waking problems in clinical research (France et al., 1991; Meltzer & Mindell, 2004; Reid, Walter, & O'Leary, 1999; Rickert & Johnson, 1988). Extinction has been found to be effective in conjunction with medication or placebo, or alone with improvements maintained at 6 and 30 months (France, Blampied, & Wilkinson, 1991).

Successful extinction programmes with autistic children have also been documented (Schreck, 2001; Wolf, Risley, & Mees, 1964). Weiskop et al. (1999) explored extinction for night waking, settling problems, co-sleeping and early waking in children with autism aged 4 to 7 and found that systematic ignoring totally eliminated co-sleeping, decreased night waking and improved sleep-onset and night settling.

However, the problem with extinction procedures is that it is stressful for parents. Perhaps parents might lack consistency. Parents must ignore their child's crying no matter how long it lasts. If parents responds after a certain amount of time, the child will only learn to cry longer the next time (Mindell et al., 2006). Also parents, although advised about the likelihood of an "extinction burst", which is a worsening of the child's negative behaviour when treatment is implemented, may perceive it as evidence that the intervention is not working (Moore et al., 2007) and may intermittently attend to their distressed child making the extinction programme fail (Rapoff, Christophersen, & Rapoff, 1982). Many parents are unable to ignore crying long enough for the procedure to be effective and also are often unwilling to use this technique (Hall & Nathan, 1992).

2.5.2 Graduated extinction

Graduated extinction is a variant of systematic ignoring method. This method involves ignoring the child's negative behaviours for a specific duration of time, before briefly checking on the child or gradual removal of reinforcement (by gradual removal of parents' physical presence). Usually the interval between checks is tailored to the child's age as well as the parents' judgement of how long they can tolerate the child's crying. This period of time can be fixed (e.g. every 5 minutes) and/or can be increased over a night or over a week. The checking procedure itself involves the parents comforting their child for a brief period usually between 15 seconds to a minute, avoiding eye contact and any sort of response to the child's attention-seeking behaviour. This aspect is very important to avoid the risk of teaching the child that crying leads to parents' presence. Thus, the parents' presence must be as "boring" to the child as possible and increasingly more effort must be required by the child to achieve this unrewarding situation (Wiggs, 1996).

Both randomized control studies (Adams & Rickert, 1989; Moore, Friman, Fruzzetti, & MacAleese, 2007) and case reports (Van Houten & Rolider, 1984) have found graduated extinction effective. Lawton et al. (1991) reported improvement at 2 months follow up and that parents considered that this technique was easy to implement. In addition, the combination of graduated extinction with medication, has been associated with significant improvement in the number of awakenings and the time to fall asleep at bed time in children (Bruni et al., 1998). Interestingly, a recent study evaluates the use of a "bedtime pass", which could be traded for a visit from the caregiver to a child with bedtime problems. After the card was used parents were instructed to ignore the child. The results of this study showed that the children settled to bed in shorter time compared to a control group (Moore et al., 2007).

Graduated extinction has been also used in the treatment of sleeplessness in children with autism or pervasive development disorder. A case study of a 2 year old girl and a 12 year old boy with severe bedtime disturbance (crying, screaming, self-injury behaviour) showed graduate extinction to lead to a significant decrease in bedtime tantrums, although neither child reached the zero level of night time disturbance (Durand, Gernert-Dott, & Mapstone, 1996). A form of graduated extinction is "stimulus fading" which has been defined by Owens et al. (1999) as "the process of gradually removing or changing a prompt for a behaviour to decrease reliance on it". This technique was successful in addressing sleep problems in a 6 year old boy with autism. The child needed the mother's presence to fall asleep and during night wakings. The mother's proximity to the child at bedtime was gradually decreased over a period of eight weeks, from sleeping with the child in the same room, to finally returning to her own bed. This result was maintained after 6 months (Howlin, 1984).

Although extinction techniques and their variations appear to be successful in the treatment of sleeplessness problems and in trying to eliminate negative behaviours, they do not provide positive behaviours to take their place (Adams & Rickert, 1989). They are therefore ideally combined with teaching the child more positive alternative behaviours.

2.5.3 Positive bedtime routines

Positive bedtime routines involve the institution of a sequence of pleasurable and calming activities preceding bedtime in order to establish a behavioural chain leading to sleep onset (Morgenthaler et al., 2006). Thus, instituting a fixed sleeping place and "rituals" which always precede the onset of sleep, creates positive bedtime associations and promotes independent sleep. This method involves collaborating with parents in order to establish a few quiet activities (e.g. bath, change into pyjamas, brush teeth, read books, turn out lights and go to sleep) lasting maximum 20 minutes (Mindell et al., 2006; Moore et al., 2007). Positive bedtime routines might be also used with other techniques such as "faded bedtime" with children who tend to fall asleep late at night. This procedure involves changing the child's bedtime to be consistent with their natural tendencies. Average baseline bedtimes are calculated and 30 minutes are added to it to guarantee that the child falls asleep within 15 minutes. When a smooth bedtime ritual has been established and associated with rapid sleep-onset, the child's bedtime is moved 10 to 15 minutes earlier every few nights, until the desired bedtime is reached.

The literature that has demonstrated the efficacy of faded bedtime is less extensive than the studies looking at extinction or graduated extinction. Two studies found positive routines with faded bedtime to be beneficial in the treatment of bedtime problems, or frequent night waking (Galbraith & Hewitt, 1993; Milan, Mitchell, & Berger, 1981), whereas another study did not find any significant differences between extinction and positive routines with faded bedtime, though both techniques were found effective when compared to controls where no treatment was implemented (Adams & Rickert, 1989). Faded bedtime appears to be beneficial also for the treatment of night waking and excessive day time sleepiness in children with autism (Piazza & Fisher, 1991).

A similar method is "faded bedtime with response cost" in which the child is taken out of bed for prescribed periods of time, when he/she does not fall asleep. Also bedtime is delayed and day time sleep is not allowed. Only once the behavioural chain is well established and the child is falling asleep quickly, the bedtime is moved earlier by 15 to 30 minutes over successive nights. It is believed that sleep restriction enhances the physiological drive to sleep and facilitates the capacity to fall asleep. Indeed, research in school-age children has shown that delaying bedtime and the resultant sleep restriction leads to shorter sleep latency at bedtime (Randazzo et al., 1998; Sadeh et al., 2003).

Faded bedtime with response cost seems also to be effective in the treatment of early waking, night waking and delayed sleep onset in children with autism aged 5 to 8 (Piazza, Fisher, & Sherer, 1997). Although there is evidence on the efficacy of extinction, graduated extinction, positive routine with faded bedtime (with and without response cost) for use with TC (Mindell et al., 2006), only extinction has sufficient evidence for a possibly efficacious intervention for sleep problems in children with autism (Schreck, 2001).

2.5.4 Scheduled awakings

This method is used in the treatment of night wakings. The method consists of calculating the average time of occurrence of the child's nocturnal awaking and instructing the parents to awake the child a short time before the expected awaking will occur. The child is then quickly encouraged to continue sleeping. The period of time between scheduled awakings is gradually increased with the intent to establish an undisturbed sleep period lasting the whole night (Sadeh, 2005).

Scheduled awakings have been used successfully in the treatment of spontaneous night awaking in a case study (McGarr & Hovel, 1980) and in a multiple baseline design (Johnson, Brandley-Johnson, & Stack, 1981) with TC, as well as in the treatment of possible sleep terrors in children with autism (Durand, 2002). Rickert and Johnson (1988) found that scheduled awakings were significantly more effective than a control group with no treatment, and as effective as extinction, however extinction worked more quickly.

Although clinical interventions have shown the effectiveness of this method, the underlying mechanism by which scheduled awakings affect arousal disorders in children is still unclear. It has been suggested that the waking causes a reorganisation of the child's deep sleep, which eliminates the undesired awaking or that the procedure conditioned the child to self-arouse (Durand, 2002).

However, the major limitation of this method appears to be the fact that it is more complex than other methods to implement. Parents are in fact required to implement the treatment at least once every night (Moore et al., 2007) and it may be difficult to convince them to wake a sleeping child (Mindell, 1999). Furthermore, this method can be applied only to children without settling problems although it was found that scheduled awaking has higher parental adherence rate than extinction (Rickert & Johnson, 1988).

2.5.5 Booklet-delivered behavioural treatment for sleep problems

A recent review of randomized or quasi-randomized controlled trials of self-help, media based behavioural therapy (e.g. audiotape, videotape, book, manual) for parents and caregivers of children and adolescents with behavioural problems other than sleep, have shown that these methods were more effective than no treatment (Montgomery, 2001). Although most behavioural treatment studies for sleep problems in children have been based on a face-to-face approach as the main mode of delivery, a limited number of studies have explored a more economic mode of delivery, such as interventions based on an informational booklet only.

Montgomery et al. (2004) carried out a randomized controlled trial with two active treatment groups: a conventional face-to-face and a booklet delivery of behavioural advice. Both interventions aimed to treat settling and night waking problems in children with learning disabilities aged 2-8. Results confirmed the effectiveness of conventional behavioural treatment for sleep problems and interestingly, suggested that brief delivery of this treatment using the booklet did not reduce its effect, with benefits maintained at six months. In addition, only minimal differences were found in an index of satisfaction between the treatment groups with 75% of parents in the conventional group and 68% in the booklet group reporting to be satisfied with the intervention. Eckerberg (2002) compared the effects of graduated extinction based on advice and support to one based on a written booklet only and found that both interventions reduced protesting and sleep latency, reduced the number of night wakings and extended sleep duration. Furthermore, the study did not find differential effects of the treatment format. Seymour, Brock, During and Poole (1989) failed to show any effects of adding telephone support to their trial of a booklet containing specific advice about behavioural techniques. The booklet, with or without the telephone support, showed superior outcomes compared with a waiting list; however, the results were achieved faster in the group with therapist contact. After 4 weeks of treatment there were no significant differences between the 2 interventions and positive outcomes were maintained at a 3-month post treatment follow-up. Similarly, Weymouth et al. (1987) evaluated the efficacy of an advice booklet. The authors performed three studies in which participants received a booklet and full clinical support with a therapist, or a booklet and reduced contact with a therapist, or only the booklet. Overall, they found that parents could be successful in the treatment of their child with the use of a self-administered booklet, with some parents requiring additional clinical support.

However a randomized controlled study (Scott & Richards, 1990) did not find significant differences in terms of nocturnal waking frequency between a group who received a special advice booklet and support visits, another group who received the booklet only and finally a group who did not receive any form of intervention at all. The authors concluded that support and advice do not lead to changes in sleep problems in young children because they are very resistant to any sort of change. Although the treated groups reported no improvement in terms of a child's night waking and parental level of distress, parents of the treatment groups were positive about both the support they had received and the booklet.

The results of these studies are promising for several reasons. Firstly, these results suggest that parents can successfully utilise information on sleep related behavioural interventions with no need for face to face professional help. For instance, it has been reported that parents have used intervention methods based on books, parenting magazines and television with a high rate of success (Johnson, 1991). Secondly, such treatment has important cost implications. Given the increasing demands on healthcare providers in most countries, services might be inadequate to meet these needs. In fact sleep problems often contribute to other medical problems such as hyperactivity and impairment of parents' mental health. Therefore, booklet-based interventions might be a solution to this increasing healthcare request, because treatment in booklet format is easy to access and it can be administered to large groups with minimal costs (Montgomery et al., 2004).

However, several factors may affect the result of a booklet based behavioural intervention for sleep problems. For example, parents whose child has severe sleep problems may require more individualised attention than written information can provide (Mindell, 1999); also families with a low level of literature may not find it appealing to use written information. However, a study involving sleep disorder patients, with limited literacy skills, found very minor differences between videos and booklets for improving knowledge and concluded that simple written information was effective (Murphy, Chesson, Walker, Arnold, & Chesson, 2000); parents' motivation is another aspect that could affect efficacy. Parents with low level of motivation may need more active support and encouragement than a booklet can provide.

Although it is widely recognised that behavioural treatments are the method of choice for children's sleeplessness, (Mindell, 1999; Mindell et al., 2006; Ramchandani et al., 2000) most studies have included pre-school age children and the methods of delivery of behavioural advice is an area which needs to be addressed by future research. For example, studies have looked at the efficacy of written information for parents with children aged 0 to 5 years old whereas booklet-based interventions have not been assessed in older TC nor with children with HFA.

2.6 STUDY RATIONALE AND RESEARCH QUESTIONS

Sleeplessness is the inability to sleep which appears to be the most common and intractable sleep problem in TC and children with HFA. The relationship between sleeplessness and neurobehavioural functioning in school age TC and children with HFA has not been sufficiently examined and only a few studies have investigated the relationship between sleeplessness and creativity in TC and no study exists currently on children with autism. Futhermore the investigations into the links between childhood sleeplessness and parental mental health have tended to focus on young children.

Thus, the first phase of the current study addresses the following questions:

- Is there a relationship between creativity and sleeplessness in school age children and adolescents, with and without autism?
- Is sleeplessness of school-age children and adolescents with and without autism associated with daytime functioning?
- Is parental mental state associated with lack of sleep in school-age children and adolescents with and without autism?

It appears that the negative effect of sleep deprivation on cognitive performance can be reversed by treating sleeplessness with behavioural therapy (Owens et al 2002; Kuhn & Elliott 2003) although whether enhanced sleep improves creativity is unknown.

Behavioural interventions can be delivered in the form of a booklet, as a self-help intervention. Booklet-based behavioural treatment has the benefit of being easy for parents to obtain and to administer to large groups with minimal costs. This method of delivery has been found to be as effective as face-to-face delivered treatments for young children with severe learning disabilities (Montgomery, Stores & Wiggs, 2004), but the efficacy of booklet-based interventions has not been assessed with school age-children or with children with autism.

Hence, phase two of this study aims to answer the following questions:

- Is a booklet-delivered behavioural treatment able to improve sleeplessness problems in school-age TC?
- Is a booklet-delivered behavioural treatment able to reduce sleeplessness problems in children with HFA?
- Does a successful booklet-delivered behavioural treatment for sleeplessness problems improve creativity in children with and without HFA?
- How do parents of typical and autistic children evaluate this booklet-based intervention?

The next chapter will describe the methods of the studies used to address these questions.

CHAPTER THREE

Methods

3.1 METHODS

This chapter aims to give a clear description of methods used in the next two chapters, including details of participants, design, procedure and tools used to measure sleep, creativity, behaviour, spatial and verbal abilities of typical children (TC) and children with high functioning autism (HFA) and parental mental state. Furthermore, a booklet-delivered behavioural treatment for sleep problems used in chapter 5 will be explained.

3.1.1 Participants

This research project investigated TC and HFA children with and without sleeplessness problems. TC were recruited through two mainstream primary schools and one mainstream secondary school in Oxford and children with HFA via Oxfordshire Autism Support Service and Oxfordshire Autistic Society for Information and Support (OASIS). Given the low response rates for both groups, recruitment was further enhanced by an advertisement on Oxford Brookes University and National Autistic Society webpages, and also presentation to OASIS parent groups. It is estimated that around 900 families were invited to take part in the study and 119 agreed to participate. Children, whose information was gathered from questionnaires completed by parents, were considered eligible to take part in this study on the basis of the following inclusion criteria:

For the autism group	HFA according to the *International Classification of Disease* (WHO, 1992) as applied by local specialised childhood autism services (i.e. parent report of formal diagnosis)

	and confirmed by further assessment using the *Autism Screening Questionnaire* (ASQ) (Berument, Rutter, Lord, Pickles, & Bailey, 1999);
For the typical children	Normal functioning, absence of HFA, or other significant clinical condition ascertained by the ASQ and a semi-structured interview with parents;
For the sleeplessness groups	No clinically significant sleep disturbance as assessed by the *Children's Sleep Habits Questionnaire* (CSHQ) (Owens, Spirito, & McGuinn, 2000); Current sleeplessness was defined as parent reported settling problems or night waking (two or more times per week, for more than 30 minutes) or early waking (waking before 5am more than one or two times a week). Further sleeplessness had to be considered problematic by parents. The sleep measure used for identifying children with sleeplessness problems was the *Composite Sleep Disturbance Index* (CSDI) (Wiggs & Stores, 1998a);
For the no sleeplessness group	No sleeplessness or other clinically significant sleep disturbance as assessed by the *Children's Sleep Habits Questionnaire* (CSHQ) (Owens, Spirito, & McGuinn, 2000) and *Composite Sleep Disturbance Index* (CSDI) (Wiggs & Stores, 1998a);

For all children	Age 4 to 16; Residents within reasonable travelling distance.

Families who agreed to take part but whose child was not eligible for this study were telephoned and explained the reason for their exclusion. They were also thanked and informed that in the event that their child was eligible to participate in a new study, they would be contacted. The original intention was that subjects that fulfilled the criteria were to be divided in four groups:

-Typical developing children and adolescents with current sleeplessness (TC+S);

-Typical developing children and adolescents without current sleeplessness (TC-S);

-High functioning autistic children and adolescents with current sleeplessness (HFA+S);

-High functioning autistic children and adolescents with current sleeplessness (HFA-S).

However, no children with HFA-S were identified therefore only three groups were included.

3.1.2 Design

The research was organised in two phases. In Phase One (Visit One and Two) a descriptive study was conducted: a) to gain descriptive details (such as age, cognitive abilities, autistic traits and schooling) of the four groups; b) to assess the prevalence and type of sleep habits and sleep problems in these groups; c) to make an evaluation of verbal and figural creative abilities; and d) to reveal patterns and connections between these variables with sleep. In addition, Phase One described the link between children's sleep and parental mental state. Descriptive study design provides a picture of the variables being examined as they occur naturally and involves neither manipulation of variables nor attempt to establish causality. The description of the variables and the relationship among them is a critically important factor for acquiring more understanding in the area of sleep and creativity in which little research has been conducted.

In Phase Two (Visit One, Two, Three) a multiple baseline design was used: a) to evaluate the effectiveness of a treatment for childhood sleeplessness problems; and b) to assess whether a successful booklet-delivered behavioural treatment for sleeplessness problems in turn improves creativity in children with and without autism. A multiple baseline approach uses a varying time schedule that allows one to determine if the application of a treatment is truly influencing the change in behaviour. For instance, the length of time of the initial baseline was varied (either one or three weeks) before applying the booklet behavioural intervention to determine if the change in sleep behaviour corresponded with the introduction of treatment. Only HFA+S and TC+ S participated in this phase (see Figure 4.1).

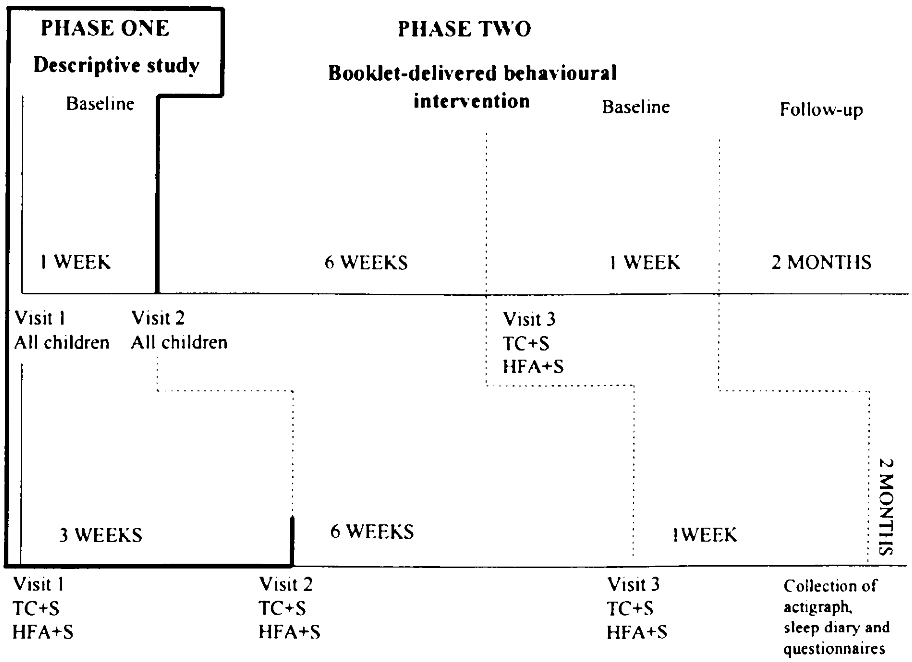

Figure 3.1 Descriptive study and multiple baseline design for demonstrating the effect of the booklet-delivered behavioural treatment for sleeplessness problems.

3.1.3 Procedure

Phase one: Descriptive study

Head teachers of all the previously mentioned schools for typical children in Oxfordshire were approached by letter (Appendix One) and asked if they would like to help with the study by distributing letters of invitation, (Appendix Two) information sheets (Appendix Three) and screening questionnaires comprising of the *Composite Sleep Disturbance Index* (CSDI) (Wiggs & Stores, 1998a), the *Children's Sleep Habits Questionnaire* (CSHQ) (Owens, Spirito, & McGuinn, 2000) and *Autism Screening Questionnaire* (ASQ) (Berument et al., 1999) (Appendix Four) to all their pupils aged between 4 and 16 years. All schools agreed to participate. Invitations were distributed by class teachers to the children who took them home to their parents. Replies were then returned by the children to the class teacher and collected from school by the first investigator two weeks later. One month later a further letter and a reply slip were distributed in the same manner, thanking parents who had responded and reminding those who had not yet replied that we would be grateful for their completed forms (Appendix Five). At the same time, first contact was established with Oxfordshire Autism Support Service and Oxfordshire Autistic Society for Information and Support (OASIS) which unhesitatingly agreed to help. Invitations were packaged with stamped addressed envelopes and sent to OASIS members and parents of children attending autistic bases in Oxford, Chinnor and Thame. Those who returned the screening questionnaire were contacted by telephone to further explain the study, to answer any queries and to arrange a time for a home visit.

Two visits were made to each family. Between these two visits passed one or three weeks (for those who did not intend to participate to the intervention phase, one week only elapsed between the two visits whereas those children who had sleep problems (TC+S and HFA+S) and took part to the intervention phase, were randomly assigned to the one or three week baseline groups). Details about the time frame are illustrated in Figure 3.1.

Visit One

Parents were briefly interviewed in order to gather family descriptive information, medical information, school details of their children and were also asked to complete *The Strength and Difficulties Questionnaire* (Goodman, 1997) to assess their child's behaviour and the *General Health Questionnaire* (GHQ) 12-item version (Goldberg & Williams, 1991) to assess parental mental state (Appendix Six). When more than two weeks between the first contact and Visit One elapsed, the CSHQ was administered again. Children were also given a questionnaire (the child version of CSHQ and a modified version of the *Sleep Disturbance Questionnaire* (MSDQ) (Espie, Inglis, Harvey, & Tessier, 2000) which assessed the frequency of attributions made by children about the causes of sleeplessness) (Appendix Seven).

Families were given an *actigraph* (wristwatch-like monitor to detect movement) for the children to wear over 5 consecutive nights (during the first week for children without sleeplessness and the first or third week for children with sleeplessness problems) and a *sleep diary* (Appendix Eight) for the parents to complete (for 7 days for children without sleeplessness and for 7 or 21 days for children with sleep problems); instructions for the use of these tool were discussed with the families. Also, during Visit One the *British Ability Scales II Edition* (BASII) (Elliot, 1983) (*Tests of verbal ability and visuo-spatial skills)* was used to gather information about the children's abilities for descriptive purposes.

Visit Two:

One week later for children without sleep problems and one/three weeks later for the children with sleeplessness, the family was visited again and the actigraph, diary and parent questionnaire were collected. Also, creativity tests were used to make an evaluation of the children's ability to generate many original ideas and also mental images. The following were administered: *Torrance Tests of Creative Thinking* (TTCT) (Torrance, 1974) (Figure 3.3) *Combining and flexibility task,* (Craig et al., 2001) (Figure 3.5) and a *Story telling task* (Figure 3.6) created specifically for this study. In addition a short term memory task (Weschler, 2004) and a vigilance task (Gale & Lynn, 1972) were also administered to the children.

<u>Phase two: Booklet-delivered behavioural intervention</u>

After visit two the parents of children with sleeplessness problems (typical children and autistic children) aged 4-11 were sent a letter of invitation (Appendix Nine) and an information sheet (Appendix Ten) to take part in a booklet-delivered behavioural intervention for childhood sleeplessness. Parents were then telephoned to give more details about the intervention and answer any questions. During the phone call those who agreed to participate gave the name of their child's GP who were sent a letter (Appendix Eleven) to notify them of the child's participation in the study and to ask if there were any medical or other reasons why the child should not take part. Then, parents were posted an illustrated booklet containing information about behavioural techniques as described in section 3.1.4. The intervention was implemented for a six-week period in which the parents were asked to keep a sleep diary. When parents felt that this would not be possible, they were asked to complete the sleep diary only during the third week of treatment to minimise workload and ensure systematic collection of the data they provided. Parents were telephoned weekly to monitor progress, encourage, discuss any problems and remind them of the information in the booklet if necessary. They were also told that they could call the researcher at any time although this rarely happened.

Visit Three

After the six-week period, in order to measure the effect of the treatment on children's sleep and creativity, a post-intervention assessment was made as at the baseline assessment (actigraphy, sleep diary, sleep questionnaire, creative testing). Parents were also given a *questionnaire to evaluate the booklet* (Appendix Twelve) and to gather their views of the treatment. One or three weeks later the questionnaires, diary and actigraph were collected.

Follow up

A postal follow up assessment consisting of a 1 week sleep diary (Appendix Eight) was completed by mothers 3 months after the end of the treatment phase. This sleep diary was used to evaluate the efficacy of the behavioural intervention over time.

Details about the timeframe of the four visits and follow up and the measures used are shown in Figure 3.1. and Table 3.1 respectively.

Table 3.1 Assessment schedule at screening period, Visit One, Visit Two, Visit Three, Visit Four and Follow-up (FU)

	Screening period	Visit One	Visit Two	Visit Three	FU
Semi-structured interview		X			
Composite Sleep Disturbance Index (CSDI)	X	X			
Children's Sleep Habits Questionnaire (CSHQ) Parents' version	X	X			
Children's Sleep Habits Questionnaire (CSHQ) Children's version		X			
Modified Sleep Disturbance Questionnaire (MSDQ)		X		X	
Sleep diary		X		X	X
Actigraph		X		X	
Autism Screening Questionnaire (ASQ)	X				
Strength and Difficulties Questionnaire (SDQ)		X			
General Health Questionnaire (GHQ)		X			
British Ability Scale second edition (BASII)		X			
Torrance Test of Creative Thinking (TTCT)			X	X	
Combining and flexibility task			X		
Story telling task			X	X	
Questionnaire to evaluate the treatment				X	

3.1.4 Booklet: description of its content

A brief, easy to read, advice booklet entitled "*Encouraging Good Sleep Habits in Children*" was given to parents experiencing problematic night time situations with their children (Appendix Thirteen). The information was based, with permission, on a booklet written by the Child Psychology Department of Dudley Road Hospital, West Birmingham and expanded by Dr Luci Wiggs to address the needs of children aged 4-11 with and without autism. Thus, the information, the illustrations and the phrasing in the booklet used in the current study were modified so as to be more appropriate to the sleep issues of older typical children and children with autism. As described in section 2.5.5, booklet-delivered behavioural treatment for sleep problems has been used successfully with typical children aged 0 to 5 years old (Seymour et al., 1989; Weymouth et al., 1987) and children with intellectual disability (Montgomery, Stores, & Wiggs, 2004) but no published empirical evaluation exists with older typical children or children with autism.

The advice booklet used in this study consisted of 16 pages of text with black and white cartoons to make it more appealing. Although the booklet's reading level was not directly assessed, the written information on which the current booklet was based were previously evaluated using a standard method called the Flesh Readability Test (Flesch, 1948) indicating that this was readable by someone educated up to age 13 (Montgomery et al., 2004).

The techniques described for resolving problems were behavioural and focused on the child's sleep problems and the parents' response to them covering the following topics:

-*Introduction to behavioural techniques in general*: This section provided a description of how behaviours can be triggered by events that precede them and an explanation of how behaviours can be encouraged or discouraged by providing or removing appropriate reinforcement. Management strategies such as ignoring, consistency and reward systems were also explained as described in section 2.5.

-*Bedtime routine*: This paragraph described the importance of a clear routine which included relaxing activities (e.g. bath, reading a story and listening to music) not lasting more than 30 minutes and the exclusion of lively activity. A bedtime routine was also described as important because it helped children to wind down before bedtime and because it taught them the order of events leading up to sleep. The importance of defining an end-point as something that children could learn to associate with falling asleep was also emphasised (see section 2.5.3).

-*Specific techniques for changing undesirable behaviour*: A number of techniques were described to help children with settling and night waking problems, as follows:
a) extinction which consists of putting the child to bed and ignoring his negative behaviour (crying, calling out for attention) as described in section 2.5.1;
b) graduated extinction which consists of ignoring the child's negative behaviour for a fixed period of time (5 minutes) before checking briefly on him. If the application of this technique disturbs other people in the family who are sleeping, the booklet suggests that this method could be applied by parents staying in the child's bedroom until the child goes to sleep and then gradually increasing the distance between them and child as described in section 2.5.2
c) positive reinforcement for encouraging desirable sleep behaviour was also discussed. All the techniques described so far need to be accompanied by explicitly rewarding appropriate behaviour. The most important reward is parent praise although the use of star chart, sweets or toys were described as useful. In the booklet was also suggested the implementation of a graded approach to reward such that a child has to perform increasingly demanding behaviour in order to be rewarded.
d) the booklet also contained a section that aimed to raise parental awareness of the problem of child sleeping in their bed and encouraged parents to return the child to his bed using the techniques described above.

A paragraph of the booklet also referred to *chronotherapy* or manipulation of sleep times as described in section 2.4. This was a full new section especially relevant for parents with older children and children with autism. Studies have shown that alterations in the melatonin rhythm (section 1.1.4 describes how the body clock is entrained to light-dark cycles via suppression of melatonin secretion operated by the light) may be responsible for sleep onset and maintenance problems in school-age children (see section 2.1.3). In fact with the onset of puberty there is a change with the timing of secretion of melatonin which frequently peaks later at night causing difficulty in getting to sleep until very late (Ishihara & Miyake, 1998).

Another group of studies (see section 2.2.3) have also suggested that sleep problems in children with autism are due to abnormalities in the melatonin regulation (Chamberlain & Herman, 1990; Nir et al., 1995; Rapin & Katzman, 1998; Ritvo et al., 1993). More specifically children with sleep onset problems may have a melatonin rhythm which peaks late in the night, contrary to those who experience night waking and early-morning waking who may have a melatonin rhythm which peaks early in the night (Richdale, 1992). Thus, in the booklet it was suggested that those children with advanced or delayed sleep phases might benefit by moving their sleep routine either forward or backwards gradually by about 15 minutes each night. In the booklet it was also argued that when a desirable sleep time is reached, it is important to stick to it. Parents were encouraged to establish daytime routines which maximise the exposure to bight light in the morning (eat breakfast by the window or sitting in the garden) of those children with delayed sleep phase, whereas they were encouraged to minimise the exposure to morning light (such as wearing sunglasses) for those children with advanced sleep phase.

3.2 ASSESSMENTS

The following paragraphs will give a description of the subjective and objective sleep measures, standard and newly developed creativity tests and the assessments used to measure the child's neurobehavioural function and the parent's mental state.

3.2.1 Screening measures

This questionnaire (Appendix four) contained three sections. Section one covered details for contacting the families, questions about the child's current medical condition, present or past sleep problems and sleep medications being taken. Section two included items for detecting the presence of sleeplessness as will be described in section 3.2.3 (*Composite Sleep Disturbance Index* or CSDI) and the *Children's Sleep Habits Questionnaire* (CSHQ) (Owens, Spirito, & McGuinn, 2000). The latter is a standardised tool for screening for sleep disorders in school-aged children and was used: a) to identify those children who were not eligible to take part in this research study (for instance children with possible sleep disordered breathing); and b) to describe and discuss those sleep problems most frequently reported by families.

CSHQ was completed by mothers but a self report version for children was also used in the current study. Only when children were referred by parents as being sufficiently old, did they fill in the questionnaire.

The reason why CSHQ was adopted is because it has been previously used to assess sleep disturbance in TC aged 8-10 (Gregory, Willis, Wiggs, Harvey, & STEPS, 2008) in adolescents (Beebe et al., 2006), children with pervasive development disorders (PDD) (Honomichl et al., 2002), and autism (Hoffman et al., 2005). In addition, satisfactory test-retest reliability of CSHQ subscales has been reported for both normal and sleep-referred clinical population (r=.62 - .79) (Owens et al., 2000).

This sleep questionnaire consists of 33 items and parents were asked to indicate the frequency with which these sleep-behaviours occurred on a 3 point Likert scale (rarely=0-1 night per week; sometimes=2-4 nights per week; usually=5-7 nights per week). The scale gave both an overall score and eight subscale scores (bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness) reflecting key sleep domains that encompass the major medical and behavioural sleep disorders. Higher scores on the total and on the eight subscales reflected greater problems. A cut-off total CSHQ score of 41 correctly yields a sensitivity of .80 and specificity of .72 (Owens, Spirito, & McGuinn, 2000) and was applied in the current study to identify children with sleep disorders.

Section three included the *Autism Screening Questionnaire* (ASQ) (Berument et al., 1999). This instrument is based on the Autistic Diagnostic Interview Revised (ADI-R) (Lord, Rutter, & Le Couteur, 1994) and is a well establish diagnostic tool for screening for autism, as well as being used in several research studies (Barret, Prior, & Manjiviona, 2004; Bishop & Norbury, 2002; Bolte, Crecelius, & Poustka, 2000; Charman, Howlin, Berry, & Prince, 2004). It has good discriminative validity between autistic spectrum and other disorders (Berument et al., 1999), can be used with all age groups, and is an especially efficient screening tool for children of over 4 years of age (De Giacomo, Lamanna, Martinelli, Spadone, & Margari, 2008). Finally, it can be administered and scored in few minutes.

The ASQ consists of 40 questions on reciprocal social interaction, language and communication and repetitive and stereotyped patterns of behaviour. In addition the scale includes a question about self-injurious behaviour and a question about the individual's current language functioning. A score of 1 is given for the presence of abnormal behaviour and a score of 0 for its absence. The total score ranges from 0 to 39 (the item on current language level not being included in the summary score) and a total score of 15 or more is the standard optimal cut-off for differentiating children with pervasive developmental disorders from other diagnoses with a sensitivity of .96 and specificity .80 (Berument et al., 1999).

The threshold score of 15 was also used in this study to identify children with autism.

3.2.2 Basic descriptive information

A semi-structured interview was carried out with parents to collect information on the following aspects:

- child's age, gender, birth order;
- child's physical condition;
- child's current and past medication;
- child's school placement;
- family's structure and social-economical status.

This can be seen in Appendix Fourteen.

The British Ability Scale Second Edition

The British Ability Scale Second Edition (BAS II) (Elliot, 1997) was used in the current study to gather descriptive information about general mental ability and spatial and verbal ability of the participants and to ensure that differences between the groups in terms of creativity were not due to differences in cognitive skills.

The BASII was chosen for the current study because it has the advantage of being a flexible instrument. Firstly, it allows the assessment of mental abilities at all ages from pre-school to the end of secondary school: secondly, it enables accurate assessment of both gifted and developmentally delayed children. In fact, the BASII has been used to assess autistic children's mental skills (Keen & Ward, 2004). Thirdly, it uses a form of adaptive testing where children start at a level that is judged to be appropriate for them. Thus, level of performance on the first few items is then used to determine which other items children are presented with; fourthly it provides important separate assessment of verbal and spatial ability; fifthly, it has careful and detailed attention to ethnic and gender related fairness; and finally, it has well established qualities of reliability and validity.

The BAS II is composed of three scales, as follows:

The <u>cognitive (or core) scales</u> designed to measure clearly identifiable verbal, non-verbal reasoning, spatial abilities all of which contribute to obtain a General Conceptual Ability measure (GCA);

The <u>diagnostic scales</u> (subordinated to the cognitive scale) provide additional information on specific abilities;

The <u>achievement scale</u> measures basic literacy and numeracy skills.

Since the current study intends both to obtain a general measurement of spatial and verbal ability for descriptive purpose and to ensure that differences in terms of creativity between the groups were not due to cognitive deficit, the *verbal core scales* and *spatial core scales* of the cognitive battery were the only measures used. The cognitive battery contains an early years battery (for ages 3:6 to 5:11) and a school age battery (for ages 6:0 to 17:11). Pre-school subtests 'verbal comprehension' and 'naming vocabulary' contributed both to generate the cluster verbal score whereas the subtests 'pattern construction' and 'copying' yielded the cluster spatial score. For the school age, subtests 'word definition' and 'verbal similarity' contribute to generating the verbal cluster score while 'recall of designs' and 'pattern construction' produced the spatial cluster score. Additional preschool and school subtests which contribute respectively to pictorial reasoning and non- verbal reasoning cluster scores were not administered to the children because of a lack of direct relevance to this investigation and because of time restrictions

Thus, verbal, spatial cluster scores and General Mental Ability score (GMA) were the only measures obtained (the GCA was pro-rated from the verbal and spatial core scale and renamed GMA). In accordance with the guidelines (Elliot, 1997) the individual subtest scores were calculated as T-scores with a mean of 50 and standard deviation of 10, GMA and verbal and spatial cluster scores were calculated as standard scores with mean 100 and standard deviation 15. See descriptive categories labels in Table 3.2.

Discrepancies between verbal and spatial scores and between those and the overall GMA score were also analysed in order to evaluate the differences between the groups in terms of cognitive profiles. For the pre-school and school children, verbal and spatial (V-S) discrepancies were identified on the basis of a minimum 18 and 16 – point difference respectively required for significance at the .05 level of probability. In terms of differences between the spatial cluster and the overall GMA score, (S-G) a minimum difference of 10 points for significance at the .05 level was required to identify a discrepancy in both pre-school and school children. Whereas a minimum difference of 10 points in pre-school children and 9 points in school age children was required to identify discrepancies between verbal cluster and GMA (V-G) at the .05 level of significance as specified in the handbook.

Table 3.2 Descriptive classification labels for GMA and verbal and spatial cluster scores

GMA and cluster scores (mean=100; sd=15)	**Categories**
130 and above	Very high
120-129	High
110-119	Above average
90-109	Average
80-89	Below average
70-79	Low
69 and below	Very low

3.2.3 Sleep measures

Subjective sleep measure

This research study used a comprehensive sleep evaluation protocol so as to capitalise on the strengths of several sleep measurements. Parent- and self-report questionnaires were adopted to assess children's sleep.

Composite Sleep Disturbance Index

The main sleep measure for identifying children with sleeplessness problems was the Composite Sleep Disturbance Index or CSDI. As described in section 2.1.1 this measure has a) permitted researchers to group childhood sleeplessness in broad categories (Montgomery et al., 2004; Richman & Graham, 1971) according to the frequency and duration of the sleep problems, b) allowed comparisons with other works, and c) shown change in intervention study (Montgomery et al., 2004; Wiggs & Stores, 1998a ;Wiggs & Stores, 2004). CSDI is based on allocating scores according to the severity of settling, night waking and early waking problems as reported by parents. Settling problems and night-wakings were measured in terms of frequency (never, less than once a month, about once/twice a month, one/two nights a week, three/six nights a week) and duration (few minutes, up to half an hour, up to one hour, one/two hours, more than two hours) whereas early waking was measured in terms of frequency only (never, less than once a month, about once/twice a month, one/two nights a week, three/six nights a week). The total CSDI score ranged from 0 to 10.

The CSDI was derived from five questions from the screening questionnaire (see Appendix Four, section two). These questions (How often does your child have problems settling at bedtime? How long does it take them to settle to sleep? How often does your child wake in the night? How long it usually take to resettle him/her? How often does your child wake before 5 am in the morning and remain awake?) were used to categorise children with no sleep problem and those with mild or severe problems (a score of 0 corresponded to absence of sleeplessness; a score of 1 and 2 respectively corresponded to a mild and severe form of sleeplessness). Children with at least multiple mild or severe sleep problems were qualified for inclusion in the study on condition that these problems were accompanied by parents' concern as expressed in question number 1 ('In your opinion does your child have a sleep problem?'). Also children were not included in Phase Two if the sleeplessness problem was of a type which would not be helped by the behavioural intervention. Details of the scoring criteria can be seen in Table 3.3.

Table 3.3 Scoring of the Composite Sleep Disturbance Index (CSDI)

Problems	Scores	Criteria
Settling frequency	0	From never to one/twice a month
	1	One/two nights a week
	2	From three nights a week to daily
Settling duration	0	Few minutes
	1	Less than an hour
	2	From one hour to more than two hours
Night waking frequency	0	From never to once/twice a month
	1	One/two nights a week
	2	From three nights a week to daily
Night waking duration	0	Few minutes
	1	Up to half an hour
	2	From one hour to more than two hours
Early waking frequency	0	From never to once/twice a month
	1	One/two nights a week
	2	From three nights a week to daily

Sleep diary

As described in section 1.3.3, a sleep diary is a preferred method: a) to quantify the severity of insomnia; b) to identify treatment targets to improve sleeplessness; c) to develop a detailed profile of the child's sleep habits; and d) to counter the disadvantages of retrospective measures through a regular monitoring of sleep. The psychometric properties of the sleep diaries have been well documented (Currie, Malhotra, & Clark, 2004; Haythornthwaite, Hegel, & Kerns, 1991; Wiggs & Stores, 1995). High internal consistency has been reported (Minde et al., 1994) as well as good agreement rates between parental diaries and videotapes of their child's sleep (Keener, Zeanah, & Anders, 1988).

In the current study, parents were given a sleep diary (see Appendix Eight) that was designed in a way which made it easy and immediate to complete. This simple format allowed also children, of a reasonable age, to contribute to its completion. The diary recorded the time that the child woke/was woken, the time that the child got up, time and duration of daytime naps, the time the child went to bed and went to sleep, and the nature and duration of any wakings at night. All the families who participated in

Phase One were asked to complete it during baseline (for a one week or three week period) whereas parents of TC or HFA with sleeplessness problems who took part in Phase Two were asked to keep completing the diary at least during the third week of the behavioural intervention (the reason why parents were not asked to fill in the diary for all the duration of intervention is that completing this for a long period of time could have perhaps demotivated the parents and make the completion of the diary less accurate) and after the intervention as at baseline for one week. After three months a one week sleep diary was mailed to the families and they were asked to complete it. The sleep diary was used only to interpret the actigraphy data of children without sleep problems who took part in Phase One. With regards to children with sleep problems, the diary was an indispensable tool not only because it aided interpretation of the actigraph data but also because it allowed tracking of the change in individual participant's sleep patterns by the use of graphical representation. Thus, the sleep diary was the main outcome measure of the behavioural intervention efficacy and the only method used to assess sleep at the follow up where it was completed for 1 week at 3 months post intervention.

The following sleep variables were graphed from the diary:

- Number of settling problems per week
- Duration of settling problems
- Number of night waking problems per week
- Duration of night waking problems
- Number of early morning waking

The Sleep Disturbance Questionnaire

The Sleep Disturbance Questionnaire (SDQ) (Espie, Brooks, & Lindsay, 1989) in Appendix Seven is widely used amongst adult populations to assess the frequency of attributions made by poor sleepers about the cause of their problem (Harvey, Gregory, & Bird, 2002; Zwi, Shawe-Taylor, & Murray, 2005). Evidence suggested that sleeplessness in adults might result from cognitive arousal (Espie, 2002). For instance, Espie et al. (1989) reported that cognitive items of the SDQ were the most highly rated and Harvey (2003) recently replicated these findings. Although there is no gold standard for subjective measurement of cognitive hyper arousal, Espie, Inglis, Harvey,

& Tessier (2000) obtained an internal consistency of .67 that indicates that SDQ had satisfactory reliability. SDQ includes 12 items. Each of them responds to a 5-point Likert scale (it is never true, it is rarely true, it is sometimes true, it is frequently true, it is always true). This questionnaire produces four factors (Factor 1 attribution concerning restlessness/agitation; Factor 2 attribution concerning mental overactivity; Factor 3 attribution concerning the consequences of insomnia; Factor 4 attribution concerning sleep-readiness) that accounted for 61% of the total variance. Beside its adequate psychometric property, this is the only questionnaire to have a child version for examining children's attributions for poor sleep. A recent pilot study by one of the applicants modified the Sleep Disturbance Questionnaire (MSDQ) to make it understandable for young children. The 12 items were rephrased (for instance "I can't get my sleep pattern into a proper routine" in the SDQ was modified with "I go to bed at different times every night and wake up at different times every morning" in the MSDQ) and the 5-point Likert scale was reduced to a 3-point scale (never or not often, sometimes, all the time or usually). In addition, the factor structure and acceptability of the MSDQ with children aged 5-16 years was assessed This study suggested that, as with adult populations, children's cognitions are relevant to sleeplessness and that this modified questionnaire provides a means of examining which areas are salient for them. The MSDQ was used in the current study and high total and subscales scores reflected greater sleep problems.

Evaluation of treatment programme questionnaire

This questionnaire (Appendix Twelve) was used to evaluate what the parents thought of the booklet in terms of relevance and helpfulness. Parents were asked to indicate, by selecting a response from a series of possible answers: a) who read the booklet; b) when they started following the advice described in it; c) their degree of satisfaction with their child's current sleep pattern; and d) how helpful they found the treatment. They were also given space to answer open-ended questions and indicate the best or most helpful and worst or least helpful aspect of the treatment (for example: "Please make any comments you wish about parts of the treatment that you thought were particularly helpful or you especially liked."). This questionnaire was a precious source of information on the parents' perception of the treatment programme and gave important feedback for future research.

Objective sleep measure

Given that questionnaires and diaries are subject to recall bias, as well as imprecision with respect to nocturnal arousals and sleep onset and offset times, children's sleep patterns were also assessed by objective measurement so as to complement questionnaire data.

Actigraphy

An objective measure of the child's sleep was obtained by means of actigraphy (see section 1.3.2 for more details). This involved the child wearing a *Basic mini-motionlogger actigraph* on the non-dominant wrist over 5 consecutive nights. Parents were instructed to put the monitors on their child's wrist when the child got into bed and to take it off when the child got up in the morning. Children and parents were also asked to press the event marker (the event marker allows to mark time with a button push that may indicate when a child went to bed or got up in the morning). A recording time epoch of 1 minute was used and nocturnal sleep/wake measures were estimated using the validated Sadeh actigraph scoring algorithm (Sadeh et al., 1989). The data was downloaded to a computer for analysis.

The sleep diary was used to assist in the analysis of objective sleep variables. For instance, wake up time, get up time, time to bed, time to sleep were all determined in accordance with the sleep diary (also the event marker was used to determine when the child went to bed and got up in the morning). When discrepancies were found between the sleep diary and actigraphy variables, parents were asked to give an explanation for such differences.

The following variables were derived for every night of recording:

- time to bed
- sleep onset time
- sleep latency (i.e. time in minutes taken to fall asleep between getting into bed to settle for sleep and time at sleep onset)
- time at final waking
- time out of bed
- total sleep time (minutes between sleep onset and sleep offset minus minutes awake)

- activity (percentage of night moving)
- minutes awake (total number of minutes spent awake between sleep onset and time of final waking)
- number of times awake after sleep onset
- sleep efficiency (percentage of time in bed spent sleeping)

See Figure 3.2 for a graphical representation of actigraphy data.

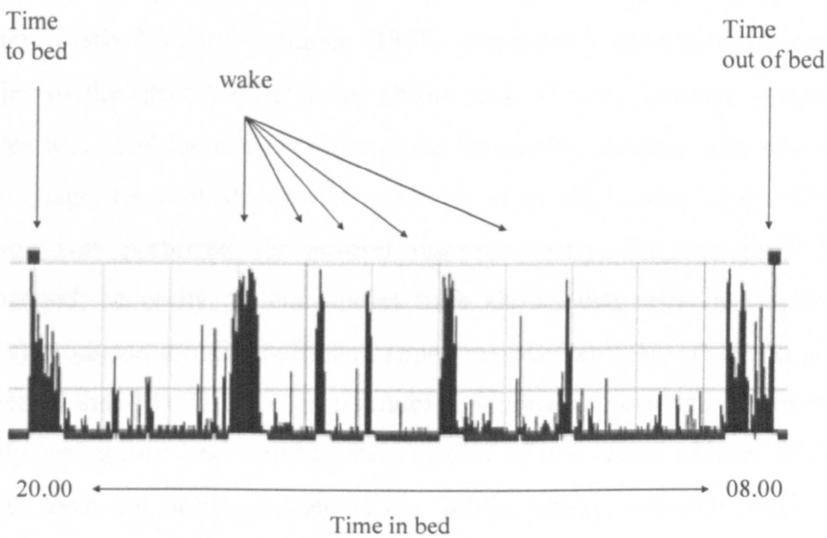

Figure 3.2 Illustration of actigraph recording. The two blue markers at the left and right side of the actigraph recording indicate the time the child went to bed and got up. The bar under the actigraph record indicates minutes scored as sleep by the Sadeh sleep-scoring algorithm. Open portions of the bar indicate minutes scored as awake. In this example the child went to bed at 8.00 pm and got up at 8.00 am and woke up more than five times during the night.

3.2.4 Creativity measures

Torrance Tests of Creative Thinking

The Torrance Test of Creativity (TTCT) (Torrance, 1974) is the most widely used test of creativity (Davis, 1997) and it has been translated into over 35 languages (Lissitz & Willhoft, 1985). With 25 years of extensive development and evaluation (Millar, 2002) the TTCT has large norm samples and longitudinal validations (Davis, 1997) and high predictive validity for a broad age range (Cropley, 2000). The figural form especially has equity benefit in terms of gender and race and for persons who have various language and cultural backgrounds (Cramond, 1993). This test was considered highly appropriate to the current research because it has been used in sleep studies with typically developing children (Randazzo et al., 1998) and in creative studies with autistic children (Craig & Baron-Cohen, 1999).

Although the use of the complete battery (Verbal and Figural) is recommended by Torrance (1974) and an update of the norms are now commercially available, the figural modes A and B and norms of the 1974 model were used in this research project. It was preferable to use only the figural mode rather than the complete battery, firstly because Torrance (1965) successfully used only the figural modes in studies of the growth of creative ability and secondly because completing abstract figures was considered more appropriate for autistic children who may have a history of language delay. With regards to the use of an old version of the TTCT, this older version was preferred for several reasons: firstly, the test itself has remained unchanged; secondly, recent studies have shown that reliability analysis of TTCT (1974) produced a high coefficient (Spearman-Brown=.93) (Prieto et al., 2006). This suggested that TTCT (1974) is a suitable instrument to evaluate creativity in children; thirdly, reliability and validity information of the latest edition of TTCT-Figural (1998) have not been provided (Kim, 2006); finally, research studies with autistic children have used the TTCT-Figural (1974) (Craig & Baron-Cohen, 1999).

The Figural Tests have two matched forms, A and B. They consist of three different types of activities: picture construction, picture completion and parallel lines or circles. In all these activities children were asked to finish incomplete figures so as to create interesting objects or pictures (Figure 3.3). Each of the three activities were scored on *originality* (when the responses were considered statistically rare), *elaboration* (when the responses were rich in unusual details), and picture completion and parallel lines or circles were scored also for *flexibility* (obtained by counting the number of different categories into which the responses fell) and *fluency* (obtained by counting the number of figures completed). Higher scores on originality, elaboration, flexibility and fluency reflected greater creative ability.

In picture construction (a) children were given a piece of paper in the form of a curved shape and required to think of a picture in which the shape was an integral part. An effort was made to elicit an original response by asking subjects to try to think of something that no one else will produce. Elaboration was encouraged by the instruction to add ideas that would make the pictures as complete and interesting as possible.

Picture completion (b) consisted of ten incomplete different figures which, on the basis of the Gestalt psychology, might elicit a tension to closure in the children. Thus, to produce an original response, the child had to control his tension and delay gratification of his impulse to complete the pictures in the simplest and easiest way possible.

Parallel lines or circles (c) tested the ability to make multiple associations to a single stimulus. Parallel lines as open figures should elicit the creative tendency to bring structure and completeness while the circles as a closed figure, requires the ability to disrupt or destroy an already complete form. Fluency is stimulated by the encouragement to produce as many pictures as possible and flexibility by the instruction to make as many different pictures and objects as possible.

An illustration of a figural test form A and B completed by children with and without autism is found in Appendix Fifteen.

Figure 3.3 An example of picture construction (a), picture completion (b), and parallel lines (c) from the figural mode A.

3.2.5 New developed creativity tests

Combining and flexibility task

This task investigated the creativity involved in imagining combinations using a variation of the Craig et al. (2001) task, created specifically for this study. The Craig et al. (2001) task, which involved the use of geometric shapes, aimed mainly to test the ability of children to fuse or combine representations in their mind and only marginally to test flexibility of thinking (the ability to produce many different ideas). In fact the Craig et al. (2001) task, consisting of a set of 5 pairs of shapes, devoted 4 trials to test combining ability (children were asked to match two shapes in order to make a possible object) and only the final trial to test for any fixedness of newly formed combinations (children were asked to form two different primary representations using one pair of shapes). Figure 3.4 illustrates Craig et al. (2001) task.

Figure 3.4 The stimuli used by Craig et al. (2001)

The variation that was devised (which was piloted on typically developing children) aimed to test more extensively the flexibility of thinking in children as this is thought to be an indispensable aspect of creativity as described in section 3.1.1. This new version of the Craig et al. (2001) test has two matched forms, A and B. Each form consists of two different tasks, the Combining task and the Flexibility task as shown in Figure 3.5.

- In the Combining task participants were presented with four trials. In each trial they were asked to form a real object by putting together two shapes (e.g. semi-circle + letter J = umbrella). This task requires the capacity to connect two representations in one's mind in order to create a novel one.
- In the Flexibility task participants were again presented with four trials and asked to form a real object by putting together two shapes. Once the children have completed the four trials this procedure was repeated (so that each trial is attempted twice). The order of presentation of the repeat trials was at random. By repeating all trials, *"fixedness"* of thinking was examined. The inability to generate two different novel representations or the persistence to give the same answer on both trials was indication of inflexible thinking.

The tasks were scored on a scale from 0 to 1 and the maximum score was 4 for both tasks. According to Craig et al.'s (2001) coding method, in the combining task a score of 1 was attributed each time the child was able to connect two shapes in a meaningful way whereas in the flexibility task a score of 1 was given when the child gave a different answer on the second attempt to link the shapes presented in the four trials. Higher scores indicated greater mental flexibility. The flexibility task was also scored for total number of responses and number of rare responses (those only occurring once) given in the four trials with the first and second attempt taken together. Rare responses were those showing creative strength, originality and also those that go beyond what is obvious and commonplace.

Appendix Sixteen contains the instructions used in the combining and flexibility tasks, and examples of the combining and flexibility tasks (Form A and B) completed by children.

Figure 3.5 The stimuli used in the new versions of the Craig et al. (2001) task. Form A and Form B contain a set of 4 pairs of shapes to assess flexibility of thinking.

Story telling task

This task, based on Craig and Baron-Cohen's (2000) free story-telling method, was developed to: a) assess if narratives produced by children with sleeplessness (with and without autism) contained less imaginary and real events than the story invented by children without sleeplessness; b) determine if children with autism spontaneously produce less imaginary-narrative than typically developing children; c) investigate if children with and without autism introduce imaginary elements into a narrative where no clues for imagination were given, or only when such cues were provided. Contrary to Craig and Baron-Cohen's (2000) free story-telling method in which children were verbally asked to tell two stories about a real and an unreal character, the newly designed story-telling method used visual prompts consisting of two matched sets of cards: form A and B containing four pairs of cards each. In each pair, one card illustrated a real/possible character or environment whereas the other was a variation of it which showed the same character or environment in an unreal/impossible version as showed in Figure 3.6 (see Appendix Seventeen for the complete sets of cards and instructions).

The story-telling task was divided into three phases: pre-test, test and post-test.

- In the pre-test or warming up stage the children were asked what they did the day before to help them to relax. Also the pre-test provided a baseline indication of verbal ability.

  Then, the series of 4 pairs of cards was shown to the children. The children were encouraged to choose one card from the real and unreal series of cards and then their preferences were noted.
- In the test phase the children were encouraged to invent two stories as lengthy as possible, based on the two cards they chose.
- The post-test was applied to verify the capacity of the children to discriminate between reality and imagination. Two cards, one reality-based and another imagination-based were presented and the child was asked to identify the card showing a picture which was not real. The same procedure was used for the rest of the cards and the answers noted.

The testing session was audio-recorded and transcribed by a native English speaker. According to Craig and Baron-Cohen (2000) the pre-test was scored on length of the story or number of words used and introduction of real facts (e.g "there were lions lying on the grass" or "a man wore a suit") whereas in the test phase not only real facts but also imaginary elements (e.g "a castle started to fly" or "the invisible road fell down") were scored. The post test was scored on a scale from 0 to 1 and the maximum score was 4. A score of 1 was attributed each time the child was able to recognize the unreal card. Inter-rater agreement between two different scorers was then calculated.

Appendix Eighteen contains examples of stories invented by typical children and children with autism.

Figure 3.6 Illustration of a real card and its correspondent unreal version

3.2.6 Child's neurobehavioural functioning assessment

Auditory digit span and sustained attention task

The digit span test included in the Wechsler Intelligence Scale for Children (Weschler, 2004) is one of the most commonly used measures of immediate verbal recall and attentional capacity in neuropsychological research (Ostrosky-solis & Lozano, 2006). This test comprised two modalities, 'digit forward' and 'digit backward'. A series of strings of digits were read to the children who are then asked to repeat them orally in the correct sequence (either forward or backward). The number of digits in each string increases from 2 to 9 forward and 2 to 8 backward. The test was discontinued if the child fails two consecutive trials. Total score corresponds to the maximum number of digits the subject is able to repeat correctly. These modalities of digit span require an adequate auditory attention and both depend on a short-term retention capacity though recently it has been argued that digit span tests implicate a more complex working memory mechanism (Baddeley, 2000).

The vigilance task was used to assess sustained attention ability (Gale & Lynn, 1972). Children were instructed to listen to a tape recording lasting for 16 minutes. The tape consisted of a continuous series of 32 random numbers. Random letters or 'wanted signals' were also presented at a rate of one per minute but randomly placed within each minute. The child had to respond to the wanted signal by writing the letter on a sheet of paper. Scores were obtained by counting a) the error of omission (when children did not respond to a wanted signal) and b) commission (when children erroneously responded to a number rather than to a wanted signal) committed in the first and second 8 minutes and in the total time. The reason why the 16 minutes vigilance task was divided in two 8 minute sections, was to see whether or not a decline in performance occurred in children with sleeplessness problems. As reported in section 2.3.1, few studies have investigated the link between lack of sleep and reduced memory and attention though high prevalence of sleep loss has been documented in school age children (Sadeh et al., 2000) and children with autism (Richdale & Prior, 1995). Thus, auditory digit span and vigilance tests were introduced in the current study to examine the association between sleep and cognitive performance.

Although there are many different types of memory and attention test available, auditory digit span and the sustained attention task were used because they could be administered to a moderately large group of children with minimal cost and in a short time.

Strengths and Difficulties Questionnaire

The Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997) is a well established 25 item behavioural screening questionnaire to be completed by parents of children aged 4-16 years (Appendix Six, section one). It provides quantifiable information on 5 subscales: 'prosocial behaviour' (positive attributes), 'peer problems', 'hyperactivity', 'conduct problems' (misbehaviour) and 'emotional disturbance'. Behaviours are rated on a 3-point scale (not true, somewhat true, certainly true) indicating the extent to which they apply to the child. Each subscale contains 5 items and the score can range from 0 to 10 provided that all the items have been completed. A total deviance score ranging from 0 to 40 can also be calculated based on the four subscales dealing with problems and excluding the pro-social scale. Higher scores on this questionnaire indicated greater difficulty except for the pro-social scale for which lower score indicated greater problems. Normal UK population values are provided and the scores can be banded into 'normal' 'borderline' and 'abnormal'. Band scores are shown in Table 3.4

This scale was considered particularly appropriate for the present study because a) it has been used in sleep studies with TC (Smedje, Broman, & Hetta, 2001) and HFA children (Allik et al., 2006); b) it was designed to discriminate between psychiatric and non psychiatric samples with a sensitivity of .87; and c) it focuses on strengths as well as difficulties.

Table 3.4 Normal, borderline and abnormal bands scores of the SDQ

	Normal	Borderline	Abnormal
Total deviance score	0-13	14-16	17-40
Emotional symptoms score	0-3	4	5-10
Conduct problems score	0-2	3	4-10
Hyperactivity score	0-5	6	7-10
Peer problems score	0-2	3	4-10
Pro-social behaviour score	6-10	5	0-4

3.2.7 Parental mental state assessment

General Health Questionnaire

The mental state of the parents was assessed using a standardised self-report questionnaire, namely the General Health Questionnaire (GHQ) (Goldberg & Williams, 1991) (Appendix Six section two). This is the most widely used measure of psychiatric disturbance in UK. This questionnaire does not detect long stage mental problems but it is a pure measure of present state. The original version consists of 60 items from which shorter versions have been developed (12, 20, 28, 30 items). Although the shorter versions are slightly less valid and reliable than the longer version (Hankins, 2008), whilst still being psychometrically sound, they are often preferred as they are quicker to administer. The 12 item version of the General Health Questionnaire was completed by parents to assess present state in relation to usual state.

Different methods have been used to score the GHQ as follows:

a) The standard method or binary method (0 =better than usual, 0=no more than usual, 1= worse than usual, 1= much worse than usual) which results in a score ranging from 0 to 12.

b) The C-GHQ where positive items (e.g. been able to enjoy your normal day-to-day activities) are scored as above whereas negative items (e.g. felt constantly under strain) are scored 0-1-1-1 thus assuming that the "no more than usual" answers indicate the presence of chronic problems rather than good health.

c) A 4-point Likert measure of severity where scores 0-1-2-3 are assigned to each response resulting in a score ranging from 0-to 36.

For each scoring method an empirical determinate threshold score indicates the likelihood of psychiatric illness. In a review of 17 studies on the GHQ-12, Goldberg and colleagues (1997) found that a cut-off score of 1/2 (a score of 1 or less indicating absence of mental illness and a score of 2 or more indicating the presence of disorder) yielded the best sensitivity (83.5%) and specificity (75.1%) rate for identifying persons with a psychiatric diagnosis.

Following the suggestion of Goldberg and Williams (1991), in the current study a threshold score of 11/12 was used to identify the presence of current emotional distress.

However given that there are considerable variations in sensitivity and specificity for a given threshold value (Cano et al., 2001; Goldberg et al., 1997), the 4-point Likert scoring method with high scores representing an increased likelihood of psychological distress was also used and thus the total GHQ (ranging from 0 to 36) was calculated for parsimony and simplicity.

CHAPTER FOUR

Phase one: Sleeplessness and creativity in typical developing children and in children with high functioning autism: Exploration of possible associations

4.1 INTRODUCTION

The high prevalence and persistence of sleeplessness problems in typically developing children and in children with high functioning autism, as described in chapter two, has been shown to play a role in the maintenance, or even the development, of neuro-behavioural problems. There are a numbers of ways in which sleeplessness could affect children's daytime functioning. Firstly, reduced or disrupted sleep such as loss of slow wave sleep has been seen to have serious effects on behaviour and mood (Horne, 1989). Also Rapid Eye Movement (REM) sleep is thought to be associated with learning process (Bloch et al., 1981) thus any reductions might impair the ability to learn and therefore interfere with the consolidation of knowledge. Secondly, the consequences of lack of sleep also appear to affect parental wellbeing which could have indirect effects on the children. For instance, in a large study of Swedish parents of 5-6 year old sleeplessness-suffering children, 34% reported increased sleeping difficulties after becoming a parent (Smedje, Broman & Hetta, 1998). Also Patzold, Richdale & Tonge (1998) using a sleep diary completed by parents, suggested that sleep problems of children with autism disturbed parents' sleep thereby increasing their stress. Thus, especially in children with autism, sleep problems add a considerable burden to the majority of parents who are already stressed, in a variety of ways, by having a child with a pervasive developmental disorder in the family.

Although it is known that sleeplessness problems are associated with children's daytime functioning in a variety of ways and that their parents have been seen to be more stressed, the most direct information on the effects of sleep duration on children's neurobehavioural functioning comes from studies that have used sleep deprivation or sleep restriction and extension paradigms (Allen, 2003; Carskadon & Dement, 1981a; Sadeh et al., 2003) whereas there is a need for more naturalistic study to examine the effect of inadequate sleep in children. In addition, sleep studies have rarely taken into account children's opinions of their own sleep problem symptoms or the children's cognitive factors associated with sleeplessness.

What is more important is the lack of research on the relationship between sleeplessness and higher cognitive function such as creativity. As described in section 2.3.2, creativity has been defined in different ways depending on the conceptual model used to describe it. With regard to the developmental model, children's creativity has been strongly connected to the idea of imagination. For instance, on the basis of the developmental theories, it is in imaginative play or self-expression that childhood creativity takes form. There has been considerable discussion concerning how creative ability develops. Feldman (1999) argued that the developmental process of creativity involves several dimensions such as cognitive and family aspects. With these considerations, any obstacle to the normal development process could seriously damage the children's ability to think divergently, to be original and flexible. Thus, it is supposed that those children who have sleeplessness problems, because of the impact that this might have on neurobehavioural functioning are seriously at risk of not following the normal trajectory of the creative developmental process.

The few studies that have investigated the relationship between lack of sleep and the consequences this has on the creative process have mainly been conducted with adults (Horne, 1988) and when typical children were included very elementary measures of sleep disturbance were used (Healey & Runco, 2006).

There are developmental disorders, such as autism, that have been found to be associated with reduced creativity in children (Craig & Baron-Cohen, 1999) although others have argued that high levels of creativity might occur in autistic adults (Fitzgerald, 2004; Jamison, 1994). For instance, Fitzgerald, 2004 made a detailed and accurate analysis of the personality of eminent creative people who are known to have or have had ASD such as the philosopher Ludwig Wittgenstein, Sir Keith Joseph and writer Lewis Carroll. However, the historico-clinical approach to autism has been criticised. Examining creative individuals, whether alive or not, for signs of autism will be always relative to the information available and will reflect the prejudice of the biographer. In addition, creators such as Temple Grandin or Stephen Wiltshire who have been diagnosed with autism are less common since impoverished imagination has been referred to as a key symptom in the standard diagnostic criteria for autism (APA, 1994) and has been documented in a number of studies with autistic children (Craig & Baron-Cohen, 1999; Craig et al., 2001; Scott & Baron-Cohen, 1996).

The contribution of sleep loss to the compromised creativity of children with autism remains unknown since the limited research conducted to examine links between sleep and creative function has looked at this relationship only in samples of typically developing children (Healey & Runco, 2006; Randazzo et al., 1998).

Therefore, there is a need to have a) more research to address the association between sleeplessness and creativity in typical children and children with autism, b) assess the effect of lack of sleep on children's neurobehavioural functioning and parental mental health in naturalistic conditions.

Hence, one primary goal and two secondary goals were addressed, as follows:

Primary goal

1. To assess the association between sleeplessness and creative thinking of children and adolescents with and without HFA.

Secondary goal

2. To assess the association between sleeplessness and daytime functioning of children and adolescents with and without HFA.

3. To examine the relationship between children's sleeplessness and parents' mental health and to explore the contribution of child's daytime behaviour to this relationship.

4.2 RESULTS

4.2.1 Statistical analysis

SPSS statistical package was used to analyse the data. Assessment of the normality of the data (using graphical normal probability plots as well as Lilliefor's statistical test) and assessment of homogeneity of variance (using the Levene test) revealed the data not to be normally distributed. This, coupled with the small sample size, indicated that non-parametric statistical tests would be most appropriated for all the sleep, creativity and neuro-behavioural functioning variables (except for age, scores on the British Ability Scale II (BASII), Autism Screening Questionnaire (ASQ) and Composite Sleep Disturbance Index (CSDI) which were analysed using parametric tests as these data did not violate the assumptions of parametric testing).

Median scores, range and mode were presented for each variable to give an indication of the distribution of scores. Where scores were measured on scales with large ranges it was not appropriate to present mode scores. For those variables analysed with parametric statistic tests, mean and standard deviation were presented. For all statistical tests a probability (or p) value of less than or equal to <.05 was taken to indicate statistical significance unless otherwise specified.

The analysis was carried out with three groups (typical children without sleeplessness problem (TC-S); typical children with sleep problems (TC+S) and high functioning autistic children with sleeplessness problems (HFA+S). The three groups were compared in terms of sleep, creativity and neuro-behavioural functioning using the Kruskal-Wallis test and when a significant difference was found, multiple-comparisons on three levels (TC-S vs TC+S; TC-S vs HFA+S; TC+S vs HFA+S) were then made using the Mann-Whitney test. To avoid type one error, the significance level of .05 was divided by the numbers of comparisons being made (Bonferroni correction). Thus, p=.016 was needed to reach statistical significance.

Spearman's correlation coefficient was used to investigate the relationship between measures of creativity and mental abilities whereas a partial-correlation was used to evaluate the links between children's sleep and parental mental state controlling for child behaviour. In both correlation types a .05 level of significance was adopted. The Wilcoxon's signed-rank test was used to assess differences between digit forwards and backwards scores and in the attention task to assess differences in terms of scores between the first and second 8 minutes of performance.

4.2.2 Participants

Out of 900 families invited to take part in the study, 119 agreed to participate (91 parents of typical children and 28 parents of children with autism made a total response rates of approximately 13%). A total of 61 children were considered eligible to take part in this study on the basis of the inclusion criteria described in section 3.1.1. Participants were then divided in groups according to the presence of autism and/or sleeplessness problems. The first was a group of 22 TC-S attending mainstream nursery (n=3) primary (n=12) and secondary (n=7) schools in Oxfordshire. The second was a group of 21 TC+S also attending mainstream nursery (n=4), primary (n=10) and secondary (n=7) schools in Oxfordshire. Both groups did not receive any special educational support during school time. The third group comprised 18 HFA+S attending mainstream nursery (n=2), primary (n=11) and secondary (n=5) schools in Oxfordshire and in other three counties at reasonable distance from Oxford. Seven of these children were supported in mainstream education by Autistic Resource Bases and 11 by learning support assistants. As can be seen in Table 4.1, the children did not differ significantly in terms of age nor general mental ability (GMA) as tested by the British Ability Scale II (BASII) (see Table 4.3) However the groups were significantly different in term of gender, schooling and scores of the Autism Screening Questionnaire (ASQ) (Berument et al., 1999).

The Composite Sleep Disturbance Index (CSDI) also differed significantly between the groups; Bonferroni post hoc tests showed that CSDI mean of TC-S was significantly lower that TC+S and HFA+S respectively at the $p < .000$ level.

These results suggested that the groups were equivalent in terms of age and GMA but differed in terms of reported sleeplessness and autism diagnosis and thus well represented the population of children with sleeplessness problems and/or autism. However it is important to highlight that the sleeplessness groups did not report high scores on the CSDI. In fact in a recent trial of behavioural treatment, a score of 4 representing for example a child with settling problems lasting more than 30 minutes at least three times a week was considered the minimum entry score to classify children with sleeplessness problems. However the different ages of the children in this may be salient, since parents might be more aware of wakings among younger children with learning disabilities (Montgomery, 2004).

Table 4.1 Descriptive details of the children in the three groups

	TC-S (n=22)	TC+S (n=21)	HFA+S (n=18)	Statistic tests p value
Age (years) Mean (sd)	9.33 (sd 3.13)	7.97 (sd 2.91)	9.14 (sd 2.75)	F= 1.30 p= .279
Gender (Number male and female)	Male=8 Female=14	Male=12 Female=9	Male=17 Female=1	χ^2= 14.1 df= 2 p=.001*
ASQ Mean (sd)	9.72 (sd3.72)	7.07 (sd 3.24)	23.72 (sd 6.04)	F=69.2 p=.000* HFA+S>TC-S, TC+S**
CSDI Mean (sd)	.58 (sd6.51)	4.38 (sd1.65)	4.48 (sd1.32)	F=64.6 p=.000* TC-S<TC+S, HFA+S**
Schooling (Number of children)	Mainstream=22 Educational Unit=0	Mainstream=21 Educational Unit=0	Mainstream=11 Educational Unit=7	χ^2=15.8 df= 2 p=.000*

***=significance at p<.05**
****=significance at p<.016**

For descriptive purposes, Table 4.2 shows: medication use, current health status of child and parental employment status in the three groups. The number of siblings within each family ranged from 0 to 3 (median=2; mode=3) for the TC-S group, from 0 to 2 (median=1; mode=1) for both the TC+S and HFA+S group.

Table 4.2 Medication use, current health status of child, and parental employment status in the three groups

	TC-S (n=22)	N	TC+S (n=21)	N	HFA+S (n=18)	N
Medication	Medication for		Moisture		Syrup for	
(including	asthma	2	cream for	2	constipation	1
sleep	Painkiller	1	eczema	1	Calcium	1
medication)			Painkiller		Homeopathic	
					medication	
					for allergy	1
Current	Asthma	2	Eczema	2	Constipation	1
health status	Cold	1	Cold	1		
Employment	**M**		**M**		**M**	
of	Home duties	11	Home duties	11	Home duties	6
Mother (M)	Service/sale	5	Social	3	Professional	3
and	Professional	2	service	2	Teaching	3
Father(F)	Education	2	Professional	2	Secretarial	3
	Health	1	Health	1	Health	1
	Self-employed	1	Secretarial		Student	1
					Unemployed	1
	F		**F**		**F**	
	Service/Sales	9	Labour	7	Professional	3
	Unemployed	2	Unemployed	3	Self-	1
	Professional	2	Sales	4	employed	1
	Labour	2	Education	2	IT	1
	Administration	2	Professional	2	Transport	2
	Health	1	Missing/Not		Social	1
	Transport	1	reported	3	service	2
	Missing/Not				Military	2
	reported	3			Unemployed	3
					Teaching	
					Banking	2
					Missing/Not	
					reported	

The British Ability Scale second edition (BASII)

The BASII was given to all 61 children as part of an initial descriptive evaluation. Following the rules outlined in the BASII handbook, n=8 children were administered the Early years battery (for ages 3:6 to 5:11) whereas n=53 were administered the School age battery (for ages 6:0 to 17:11). A General Mental Ability (GMA) summary score was extrapolated from the scales administered (verbal and spatial subscales) and pro-rated according to the BAII handbook (see section 3.2.2)

As indicated in Table 4.3, children in the three groups performed at average level (90-109) on GMA and clusters scores. One-way analysis of variance (ANOVA) was used to compare groups on these variable but no significant differences were found overall.

Table 4.3. Means, standard deviations and ANOVA of GMA, verbal and spatial cluster scores for TC-S TC+S HFA+S

	TC-S (n=22) Mean (sd)	**TC+S (n=21) Mean (sd)**	**HFA+S (n=18) Mean (sd)**	**(F value)** ***p*** **value**
GMA	94(12)	98(11)	94(13)	F=.639 p=.532
Verbal cluster Score	94(13)	98(10)	94(14)	F=.924 p=.403
Spatial cluster Score	97(11)	99(15)	97(15)	F=.102 p=.903

=significance at p<.05

To assess the differences between the groups in terms of cognitive profile, discrepancies between verbal and spatial scores and between those and the overall GMA score were analysed. Out of 22 TC-S, only 5 (22%) children demonstrated a significant discrepancy mainly in the S>V direction. On the contrary, the remaining two groups reported a greater number of discrepancies between verbal and spatial scores. In fact 10 (46%) TC+S and 8 (43%) HFA+S children showed a significant discrepancy between verbal and spatial scores. Percentages and numbers of discrepancies are reported in Table 4.4.

Table 4.4 Percentages and (numbers) of discrepancies (V-S, G-V, G-S) for TC-S, TC+S HFA+S

	TC-S (n=22)	**TC+S (n=21)**	**HFA+S (n=18)**
V-S	4% (1) V>S* 18% (4)V<S*	23% (5) V>S* 23% (5) V<S*	16% (3) V>S* 27% (5) V<S*
G-V	9% (2) G>V* 9% (2) G<V*	23% (5) G>V* 23% (5) G<V*	16%(3) G>V* 22%(4) G<V*
G-S	9% (2) G<S*	14%(3) G>S* 19% (4) G<S*	11%(2) G>S* 22% (4) G<S*

****V-S from 16 or 18 points difference for significance at p<.05***
****G-V from 9 or 10 points for significance at p<.05***
****G-S from 10 points difference for significance at p<.05***

4.2.3 Description of sleep problems in children with and without autism

In answer to the questions "In your opinion does your child have a sleep problem?" no children in the TC-S group were reported as having current sleeplessness problems whereas all children in the TC+S and HFA+S group were referred by parents to have sleep difficulties. When parents were asked: "Has he/she had a sleep problems in the past?" 1 (4%) TC-S, 3 (14%) TC+S and 15 (83%) HFA+S children were reported to have had a sleep problem in the past.

Although the three groups were not distinguishable in terms of age, younger children in the TC+S and HFA+S groups were more likely to have sleep problems than older children (66% of TC+S and 44% of HFA+S were aged 4-8 years, 23% of TC+S and 44% of HFA+S were aged 9-12 and 9% of TC+S and 11% of HFA+S were aged 13-16 years) (χ^2=2.1, df=2, p=.34).

The sleeplessness problems described by parents in the CSDI were those that have been suggested in the sleep literature as being particularly common in typically developing children over 5 years old and in children with autism. The percentages of children having these sleep problems or combinations of sleep problems on the basis of the CSDI can be seen in Table 4.5.

Table 4.5 Types of sleeplessness problems according to the CSDI, percentage and (number) of children in the TC-S, TC+S and HFA+S reported by parents to be showing them most nights

Type of sleeplessness problem	TC-S (n=22)	TC+S (n=21)	HFA+S (n=18)
Settling at bedtime	-	95%(20)	77%(14)
Settling to sleep	-	66%(14)	72% (13)
Night waking	-	19% (4)	22% (4)
Early waking	-	14%(3)	5%(1)
Settling and night waking	-	9% (2)	-
Settling and early waking	-	14% (3)	-
Night waking and early waking	-	-	5% (1)
Settling, night waking and early waking	-	-	-

Child Sleep Habits Questionnaire (CSHQ) completed by mothers

All mothers in all three groups completed and returned the questionnaire. The Kruskal-Wallis tests revealed that the group scores had significant difference on the CSHQ total score, bedtime resistance, sleep onset delay, sleep duration, sleep anxiety and night waking subscales. On the contrary, no significant differences were found on the parasomnia, sleep breathing disorder and daytime sleepiness subscales. Group scores can be seen in Table 4.6

Post hoc Mann-Whitney tests showed that there were no significant differences between TC+S and HFA+S in term of total and subscales scores. However TC-S scored significantly lower than TC+S on total score (U=108.5; p=.003), bed time resistance (U=116.5; p=.004) sleep onset delay (U=85.000; p=.000) sleep reduction (U=125.0; p=.008) and night waking subscale (U= 132.5; p=.009); TC-S also scored significantly lower than HFA+S on total score (U= 78.000; p=.001), sleep onset delay

(U=32.500; p=.000), sleep anxiety (U= 109.000; p=.012) and night waking subscale (U=111.5; p=.009).

Table 4.6 Medians, modes, ranges and Kruskal-Wallis test statistics of total and subscales scores as reported by parents in the CSHQ for TC-S, TC+S, and HFA+S

	TC-S (n=22) Median (mode) and ranges	**TC+S (n=21) Median (mode) and ranges**	**HFA+S (n=18) Median (mode) and ranges**	**(H value) *p* value**
Total sleep disturbance score	41 (38) 35-54	51 (51) 37-76	54(55) 37-76	H=15.9 p=.001* TC-S<TC+S, HFA+S**
Bedtime resistance	6 (6) 6-13	8 (6) 6-16	7 (6) 6-17	H= 12.2 p=.007* TC-S<TC+S**
Sleep onset delay	1(1) 1-3	2(2) 1-3	3(3) 1-3	H=29.7 p=.000* TC-S<TC+S, HFA+S**
Sleep reduction	4(3) 3-7	5(5) 3-9	6 (7) 3-9	H=14.24 p=.003* TC-S<TC+S**
Sleep anxiety	4(4) 4-9	5(4) 4-10	6(5) 4-11	H=7.60 p=.037 TC-S<HFA+S**
Number of night wakings	3(3) 3-5	4(4) 3-9	4(3) 3-8	H=9.50 p=.012* TC-S<TC+S, HFA+S**
Parasomnia	8(7) 7-13	10(9) 7-16	9(7) 7-15	H=8.26 p=.053
Sleep breathing disorder	3(3) 3-6	3(3) 3-7	3(3) 3-7	H=1.65 p=.921
Daytime sleepiness	12(12) 8-20	13 (13) 8-22	12(10) 8-20	H=.796 p=.673

=significance at p<.05
*****=significance at p<.016***

To examine the similarity between scores for the three groups and published normative scores (Owens, Spirito, & McGuinn, 2000), mean CSHQ subscale scores from the non-problem sleep group (TC-S) were compared with published scores for a community sample (see Table 4.7). Also mean scores from the problem sleep groups (TC+S and HFA+S) were compared with published scores for a clinic sample (see Table 4.8). Several interesting observations were evident when comparing these groups to normative data. All the subscale scores for the TC-S group were above the normative values of Owens' community sample except for bedtime resistance and number of night wakings which were only .6 and .10 lower that scores reported by the community sample. Thus the TC-S group had scores that were higher (more problematic) than the Owens' community sample. The differences ranged from .2 for sleep onset delay to 2.56 points for daytime sleepiness.

Table 4.7 Means and (standard deviations) of CSHQ subscale scores for Owens community sample and for TC-S

	Owens COMMUNITY SAMPLE (n= 1099) Mean (sd)	**TC-S (n=22) Mean (sd)**
Bedtime resistance	**7.06 (1.89)**	7.00 (1.92)
Sleep onset delay	**1.25 (.53)**	1.27 (.55)
Sleep reduction	**3.41 (.93)**	4.36 (1.59)
Sleep anxiety	**4.89 (1.45)**	5.23 (1.87)
Number of night wakings	**3.51 (.89)**	3.41 (.739)
Parasomnia	**8.11 (1.25)**	8.59 (1.94)
Sleep beathing disorder	**3.24 (.63)**	3.55 (.85)
Daytime sleepiness	**9.64 (2.80)**	12.2 (3.15)

Comparing Owens's clinical population, consisting of 154 paediatric sleep disorders patients with CSHQ subscale scores reported by TC+S and HFA+S, sleep onset delay, sleep reduction and daytime sleepiness scores in TC+S and HFA+S were higher than the mean scores of the clinic sample. The difference scores ranged from .39 for sleep onset delay to 1.01 points for daytime sleepiness for TC+S group and from .78 for sleep reduction to 1.51 points for daytime sleepiness for HFA+S.

Scores in all the remaining CSHQ subscales (bedtime resistance, sleep anxiety, number of night wakings, parasomnia and sleep breathing disorder) were lower than the clinic sample scores but higher than Owens' community sample scores (see Table 4.8).

Table 4.8 Means and (standard deviations) of CSHQ subscale scores for Owens clinic sample and for TC+S and HFA+S.

	Owens CLINIC SAMPLE (n= 154) Mean (sd)	**TC+S (n=21) Mean (sd)**	**HFA+S (n=18) Mean (sd)**
Bedtime resistance	**9.43 (3.49)**	9.14 (3.05)	8.44 (3.29)
Sleep onset delay	**1.80 (.88)**	2.19 (.75)	2.67 (.59)
Sleep reduction	**4.94 (1.98)**	5.71 (1.58)	5.72 (1.93)
Sleep anxiety	**7.09 (2.44)**	6.10 (2.21)	6.83 (2.47)
Number of night wakings	**5.69 (1.60)**	4.57 (1.83)	4.72 (1.74)
Parasomnia	**11.22 (2.53)**	10.1 (2.35)	9.67 (2.67)
Sleep beathing disorder	**4.71 (2.54)**	3.62 (1.11)	3.67 (1.13)
Daytime sleepiness	**11.99 (3.39)**	13.0 (3.86)	13.50 (3.77)

There were no significant differences between the three groups in the current study in terms of bedtime, sleep duration (weekday and weekend), and morning waking time. However a significant difference was found between the groups in terms of night waking duration. Mann-Whitney tests revealed that children in the TC+S and HFA+S groups woke at night for a time significantly longer than children in the TC-S group ($p<.016$). Details about bedtime, sleep duration, time of final waking in the morning, and night awaking duration in each group can be found in Table 4.9.

There appears to be a relationship between children's age and amount of time per night spent asleep in the TC-S and TC+S but not in the HFA+S group. In fact, age and sleep duration were significantly negatively correlated ($r= -.479$; $p=.024$) at the .05 level in the TC-S group. The amount of time spent asleep and age were also significantly negatively correlated in TC+S ($r= -.454$; $p=.039$) at the .05 level but not significantly correlated in HFA+S ($r=.215$; $p=.408$).

Table 4.9 Medians, modes, ranges and Kruskal-Wallis test statistics of bedtime, sleep duration, wake up time and night waking duration as reported by parents in the CSHQ for TC-S, TC+S and HFA+S.

	TC-S (n=22) Median (mode) and ranges	**TC+S (n=21) Median (mode) and ranges**	**HFA+S (n=18) Median (mode) and ranges**	**(H value) *p* value**
Bedtime (hours)	8.20 (20.00) 8-11	8 (8) 7-11	8.30 (8) 7-10	H=.342 $p=.843$
Sleep duration (hours)	9.45 (11) 7-12	9 (10) 7-12	9 (9) 4-10	H=5.15 $p=.072$
Morning waking time (hours)	6.40 (7) 6-8	7 (7) 6-8	7(7) 6-8	H=1.82 $p=.396$
Night waking duration (minutes)	.00 (0) 0-30	9 (5) 0-60	10 (0) 0-120	H=12.1 $p=.002$* TC-S<TC+S HFA+S**

****=significance at p<.05***
*****=significance at p<.016***

Children's self-report sleep questionnaires

With regard to the self-report version of the CSHQ, not all children were able to complete the questionnaire; out of 61 children, 19 in TC-S, 17 in TC+S and 14 in HFA+S group filled in the questionnaire. As expected, children in the TC+S and HFA+S group reported higher sleep disturbance scores than children in the TC-S group. Post hoc tests, in fact, revealed that TC-S scored significantly lower than TC+S (U=44.50; p=.000) and HFA+S (U= 52.50; p= .003) but no significant difference was found between TC+S and HFA+S (U= 114.00; p= .842). Table 4.10 shows medians, modes, ranges and H and p values of the total score of the CSHQ child version.

Table 4.10 Medians, modes, ranges and Kruskal-Wallis test statistics of total score as reported by TC-S, TC+S and HFA+S in the CSHQ child version

	TC-S (n=19) Median (mode) and ranges	**TC+S (n=17) Median (mode) and ranges**	**HFA+S (n=14) Median (mode) and ranges**	**(H value) *p* value**
Total sleep disturbance score	30 (28) 25-44	40 (40) 31-52	40.50 (38) 24-47	H=15.64 p=.000** TC-S<TC+S and HFA+S**

=significance at p<.05
*****=significance at p<.016***

There was no discrepancy between parents' and children's reports in the CSHQ. In fact the total score of the CSHQ self-report version for children was significantly positively correlated with the total CSHQ version for parents in TC+S (r=.569; p=.017) and HFA+S group (r=.655; p=.011). However the correlation between total disturbance score of the CSHQ version for parents and total CSHQ version for children did not reach level of significance in the TC-S group (r=.454; p=.051).

A total of 49 children(TC-S=17, TC+S=17 and HFA=15) completed the modified version of the Sleep Disturbance Questionnaire (MSDQ). Children in the TC-S scored significantly lower on total MSDQ score than TC+S (U=43.50; p=.002) and HFA+S (U=41.500; p=.003) but no significant difference was found between TC+S and HFA+S (U=89.000; p=.483) in term of total score. With regard to the factor scores significant differences were found only in term of "attribution concerning mental overactivity" (Factor 2) but not in the remaining three factors. In fact, children in the TC-S group reported lower scores than TC+S (U=49.500; p=.001) and HFA+S (U=32.000; p=.000) in Factor 2.

Table 4.11 shows medians, modes, ranges and H and p values of the total score of the MSDQ and its derivate Factors (Factor 1, attribution concerning restlessness/agitation; Factor 2, attribution concerning mental overactivity; Factor 3, attribution concerning the consequences of insomnia; and Factor 4, attribution concerning sleep-readiness).

Table 4.11 Medians, modes, ranges and Kruskal-Wallis test statistics of total and subscales (factors) as reported by TC-S, TC+S and HFA+S in the MSDQ

	TC-S (n=17) Median (mode) and ranges	**TC+S (n=17) Median (mode) and ranges**	**HFA+S (n=15) Median (mode) and ranges**	**(H value) *p* value**
Total score	18.50 (15) 13-28	25(25) 14-36	24(22) 18-34	H=12.4 *p*=.002* TC-S<TC+S and HFA+S**
Factor 1	9(7) 6-14	12 (12) 6-18	10.50(10) 7-18	H=6.73 *p*=.034*
Factor 2	4 (3) 3-9	8(8) 4-9	7(9) 5-9	H=15.3 *p*=.000* TC-S<TC+S and HFA+S**
Factor 3	3(3) 2-5	3 (3) 2-6	4(4) 2-5	H=2.20 *p*=.333
Factor 4	2(1) 1-3	3(3) 1-3	2(2) 1-3	H=5.54 *p*=.062

****=significance at p<.05***
*****=significance at p<.016***

The total score of the MSDQ was significantly positively correlated with the total score of the CSHQ self-report version for children in TC-S (r=.621; p=.010) and TC+S (r=.638; p=.011) but not in HFA+S group (r=.471; p=.104). There was concordance between parental and children reports of sleep problems. In fact, the total MSDQ score was significantly positively correlated with the total score of CSHQ version for parents in all three groups TC-S (r=.542; p=.030), TC+S (r=.619; p=.014) and HFA+S (r=.591; p=.026).

Actigraphy

Objective sleep data were obtained using actigraphy during the same week as a sleep diary was completed. Out of the total of 61 children, 21 TC-S wore the actigraphs for a median of 6 nights whereas 20 TC+S and 14 HFA+S children wore the actigraphs for a median of 7 nights. For 6 (9%) children the actigraphy data were not available. Reasons for this were as follows; the parents of two children (one in the TC-S and one in the TC+S group) did not attempt to put the monitor on their child's wrist for fear of disrupting their child's sleep and for 4 children in the HFA+S group the actigraph failed to record or retain the data. All of the children who wore the monitors kept them on throughout the recording period. The median scores and range on each sleep variable are presented to give an indication of the distribution of scores (see Table 4.12). Since scores were measured on scales with large ranges (e.g. time, percentages) most scores were different and multiple modes existed so it was not appropriate to present mode scores.

Table 4.12 Medians, (ranges) and Kruskal-Wallis test statistics of the actigraph variables for TC-S, TC+S and HFA+S

	TC-S (n=17) Median and (ranges)	**TC+S (n=17) Median and (ranges)**	**HFA+S (n=15) Median and (ranges)**	**(H value) *p* value**
Sleep Onset time (hours)	21.65 (20-24)	21.66 (20-23)	21.58 (20-24)	H=.672 *p*=.715
Wake up time (hours)	7.15 (6-8)	7.05 (6-9)	7.48 (6-8)	H=3.89 *p*=.143
Minutes awake during sleep	61 (10-187)	62 (19-205)	70 (7-198)	H =.308 *p*=.857
Total sleep time (hours)	8.83 (6-10)	7.91 (6-11)	7.86 (7-10)	H =1.06 *p*=.586
% night movement	12.96 (5-30)	14.39 (5-37)	13.31 (2-45)	H=.363 *p*=.834
Time to bed (hours)	21.10 (20-23)	21.01 (20-23)	21.33 (20-24)	H=1.46 *p*=.480
Time out of bed (hours)	7.31 (6-9)	7.38 (6-10)	7.66 (7-8)	H=3.98 *p*=.136
Sleep latency (minutes)	31.20 (9-56)	29.10 (10-63)	23.30 (10-85)	H=.163 *p*=.922
Sleep efficiency %	89.50 (15-97)	87.60 (68-96)	87.50 (69-99)	H=.175 *p*=.916

****=significance at p<.05***
*****=significance at p<.016***

When Kruskal-Wallis tests were used, no significant differences were found between the groups in any objective sleep variable. However in terms of total sleep time (minutes between sleep onset and sleep offset minus minutes awake) it appeared that TC+S and HF+S tended to sleep nearly 1 hour less than TC-S. In addition HFA+S tended to go to bed and to get up approximately .35 hours later than TC-S and TC+S. They also tended to wake up at night for nearly 10 minutes longer than TC-S and TC+S.

Spearman correlations were computed for bedtime, wake up time and duration of night waking derived from actigraphy and CSHQ measures. Significant positive correlations were obtained for bedtime in TC-S (r= .457; p=.043), TC+S (r=.557; p=.013) and HFA+S group (r=.721; p=.004) whereas significant positive correlations were found for wake up time (r=.578; p=.039) and duration of night waking (r=.637; p=.019) only in the HFA+S group.

4.2.4 Creativity in children with and without sleep problems

Torrance Tests of Creative Thinking (TTCT)

In order to determine the reliability of the creativity measures, the three activities which constitute the test (picture contraction, picture completion and parallel lines or circles) were all scored by two independent raters of whom one was blind to the identity of the participants and the aims of the study. The agreement between these two raters was calculated using intra-class correlation coefficient (ICC) with a two way mix model. The single measure ICC was .930 for the total creativity score, .900 for the originality score, .933 for fluency score, .919 for flexibility score and .802 for elaboration score. The inter-rater agreement on the creativity measures were sufficiently high to suggest that they were reliably measured and consequently they were included in the analysis.

All participants in all three groups completed the test. Significant group differences were found only on the total creativity score, originality score and elaboration score as no differences were seen on fluency and flexibility scores. Post hoc Mann-Whitney tests revealed that TC-S scored significantly higher on total creativity score (U= 67.000; p=.001) originality score (U=54.500; p=.000), and elaboration score (U=79.000; p=.001) than HFA+S. No significant differences were found between TC+S vs HFA+S and TC+S vs TC-S in any of these variables. Details of the creativity data are shown in Table 4.13.

Table 4.13 Medians, (modes), ranges and Kruskal-Wallis test statistics of the total TTCT – Figural score and originality, fluency, flexibility and elaboration scores for TC-S, TC+S and HFA+S

	TC-S (n=22) Median (mode) and ranges	TC+S (n=21) Median (mode) and ranges	HFA+S (n=18) Median (mode) and ranges	(H value) *p* value
Total creativity score	103(122) 20-58	72(60) 12-152	60(0) 0-134	H=12.2 *p*=.002* TC-S >HFA+S**
Originality score	43 (43) 11-61	26(10) 0-64	22(0) 0-41	H=12.9 *p*=.002* TC-S >HFA+S**
Fluency score	22 (22) 4-36	18(5) 5-33	19(20) 0-29	H=4.43 *p*=.109
Flexibility score	18(15) 4-23	13(13) 4-28	15(15) 0-22	H=4.91 *p*=.086
Elaboration score	16(1) 1-57	5(0) 0-31	2(0) 0-44	H=10.9 *p*=.004* TC-S >TC+S**

****=significance at p<.05***
*****=significance at p<.016***

As can be seen in Table 4.14, creativity scores in the three groups were compared to a normative sample (Torrance, 1974) except for the total creativity score for which norms are not provided in the TTCT 1974 version. The data used in preparing the norms were based on 1365 primary and secondary school children.

Consistent with the expectations, mean scores of all creative measures for HFA+S were lower than that for the normative sample. TC+S also scored below the average level of performance on all the creative measures except for originality which was only 1 higher than that for the normative sample. With regards to the TC-S group, mean scores of all creative measures were higher than that for the normative sample but the elaboration score was far lower than that for the normative data.

Table 4.14 Means and standard deviations of originality, fluency, flexibility and elaboration scores for normative sample and for TC-S, TC+S and HFA+S

	NORMATIVE SAMPLE (n=1365) Mean (sd)	TC-S (n=22) Mean (sd)	TC+S (n=21) Mean (sd)	HFA+S (n=18) Mean (sd)
Originality score	**28(10)**	39(11)	29(18)	22(12)
Fluency score	**21(6)**	22(6)	18(8)	16(8)
Flexibility score	**16(5)**	17(4)	15(7)	12(6)
Elaboration score	**68(26)**	20(17)	9(10)	6(10)

The relationship between creative thinking and cognitive ability in each group was evaluated with a series of Spearman's correlations between total creativity, originality, fluency, flexibility and elaboration scores as obtained by means of the TTCT and general mental ability (GMA), spatial and verbal ability as obtained by means of the BASII (see Table 4.15). The results showed that there were no significant correlations between any of these variables (*p*>.05) in the TC-S and TC+S groups. However, in the HFA+S group, total creativity score and originality score were significantly positively correlated with GMA at the .05 level. This result is consistent with previous studies reporting mild positive significant (Gough, 1976) correlations between measures of IQ and divergent thinking tests and suggest that mental ability and creativity are in some way connected in HFA+S children.

Table 4.15 Correlation between measures derived from TTCT figural form and BASII cognitive scales: Spearman's r coefficient and *p* value.

	GMA			**Verbal cluster score**			**Spatial cluster score**		
	TC-S (n=22)	**TC+S (n=21)**	**HFA+S (n=18)**	**TC-S (n=22)**	**TC+S (n=21)**	**HFA+S (n=18)**	**TC-S (n=22)**	**TC+S (n=21)**	**HFA+S (n=18)**
Total creativity score	.101 *p*=.664	.-215 *p*=.362	.492* *p*=.038	.035 *p*=.881	-.294 *p*=.208	.461 *p*=.054	.225 *p*=.327	-.154 *p*=.504	.237 *p*=.344
Originality score	.265 *p*=.246	-.210 *p*=.374	.486* *p*=.041	.144 *p*=.534	-.265 *p*=.259	.429 *p*=.076	.317 *p*=.161	.-162 *p*=.482	.256 *p*=.305
Fluency score	.199 *p*=.387	-.122 *p*=.608	.347 *p*=.159	.054 *p*=.817	-.160 *p*=.502	.400 *p*=.100	.322 *p*=.155	-.136 *p*=.555	.091 *p*=.719
Flexibility score	.135 *p*=.560	-233 *p*=.323	.450 *p*=.061	.089 *p*=.703	-.242 *p*=.304	.424 *p*=.080	.154 *p*=.506	-.200 *p*=.385	.218 *p*=.385
Elaboration score	.-095 *p*=.681	-.201 *p*=.395	.442 *p*=.066	-.200 *p*=.385	-.375 *p*=.103	.388 *p*=.112	.049 *p*=.835	-.062 *p*=.788	.243 *p*=.332

=significance at p<.05
****=significance at p<.016***

Combining and flexibility tasks

Of the 61 children, 12 TC-S, 10 TC+S and 10 HFA+S completed combining and flexibility tasks on form A whereas 10 TC-S, 11 TC+S and 8 HFA+S completed the matched form B (scoring details are given in section 3.2.5). All three groups reported a median total score of 4 in the combining task. When Kruskal-Wallis tests was used to compare groups on the numbers of correct responses no significant differences were found (H=.590; *p*=.745) overall. This result indicated that all the groups were able to combine two shapes together. This finding is also consistent with a previous creativity study where a similar task was used and where it was reported that typical children had combining ability as well as children with autism (Craig et al., 2001).

A median total score of 2, 3 and 3 for TC-S, TC+S and HFA+S respectively was reported in the flexibility task. Kruskal-Wallis tests revealed that there was no significant difference between the three groups in term of flexibility (H=.295; *p*= .863). However when children were asked to recombine the previously combined shapes, more of the TC+S (81%) and HFA+S (77%) groups made from 1 to 4 inflexible errors (giving the same answer in both trials or not responding at all) compared to TC-S (68%). This difference between the groups in terms of the number of inflexible errors still did not reach the level of significance (χ^2 =.718, *df*=2, *p*=.689). The flexibility task was also scored for total number of responses and number of rare responses (those occurring only once) given by children in the four trials (first and second attempt altogether). Table 4.16 shows that the total number of responses given by HFA+S children (n=88) was lower than the total number of responses given by TC-S (n=128) and TC+S (n=133) however the percentage of rare responses in the TC-S group (30%) TC+S (24%) and HFA+S (30%) appear to not differ.

Table 4.16 Total number of responses according to flexibility task form A and B as reported by TC-S, TC+S and HFA+S

	TC-S (n=22)	TC+S (n=21)	HFA+S (n=18)
Trial 1	40	37	25
Trial 2	26	33	28
Trial 3	33	31	21
Trial 4	29	32	14
Total	128	133	88

Story telling task

The inter-rater agreement on the story-telling task was sufficiently high to suggest that its scoring was reliable. In fact intra-class correlation coefficient (ICC) with two ways was .998 for number of words, .920 for number of real facts and .910 for number of imaginary elements.

Not all children completed the test. In the TC-S, n=1 child did not complete the test because the task was prematurely interrupted for an unexpected event whereas in the HFA+S group n=2 refused to invent a story despite being encouraged by the investigator. On the contrary, all children in the TC+S group participated in the story-telling task testing section. The Kruskal-Wallis test showed that the groups did not score significantly different in the number of words and introduction of real facts during the pre-test. Also no significant differences were found in terms of number of words, and the number of real and imaginary elements when a real card was used during the test phase. On the contrary, when an unreal card was presented the number of real facts introduced by children significantly differed between the group. Subsequent Mann-Whitney tests revealed that TC-S scored significantly higher on the number of real facts than TC+S (U=109; p=.005). However, no significant differences were found between the groups on number of words and number of imaginary elements when an unreal card was used. All children were able to discriminate between a real and unreal card, in fact all groups reported a median total score of 4 at post-test (scoring details are given in section 3.2.5). As expected no significant differences were found between the group on the numbers of correct responses (H=.580; p=.755). Details of the storing telling task are given in Table 4.17.

Table 4.17 Medians (ranges) and Kruskal-Wallis test statistics of the story-telling task variables for TC-S, TC+S and HFA+S.

	TC-S (n=21)	**TC+S (n=21)**	**HFA+S (n=16)**	**(H value p value)**
PRE-TEST Number of words	24 (1-96)	16 (0-101)	19 (0-88)	H=.125 p=.940
PRE-TEST Number of real facts	3 (1-17)	3 (0-10)	2 (0-6)	H=1.71 p=.418
TEST (real card) number of words	56 (0-147)	56 (0-522)	61 (0-270)	H=.735 p=.693
TEST(real card) number of real facts	5 (0-17)	3 (0-45)	4 (0-20)	H=1.36 p=.506
TEST (real card) number of imaginary elements	0 (0-6)	0 (0-7)	0 (0-10)	H=.234 p=.890
TEST (unreal card) number of words	65 (10-255)	38 (0-526)	79 (6-258)	H=7.49 p=.024* TC-S>TC+S**
TEST (test unreal card) number of real facts	7 (2-22)	4 (0-39)	6 (1-18)	H=3.80 p=.150
TEST (unreal card) number of imaginary elements	0 (0-2)	0 (0-3)	0 (0-7)	H=.152 p=.918

****=significance at p<.05***
*****=significance at p<.016***

4.2.5 Cognitive functioning in children with and without sleep problems

Auditory digit span and sustained attention task

All three groups completed the digit forwards and backwards task. The medians, modes and ranges for each of the three groups and analysis of results are presented in Table 4.18. When auditory digit span scores forwards and backwards were analysed significant differences between the groups were found in terms of digit forwards but not in the digit backwards scores. Multiple comparisons showed that TC-S scored significantly higher than TC+S on the digit forwards test at the .014 level of significance. However no significant differences were found between TC-S vs HFA+S and TC+S vs HFA+S.

To compare the scores of digit forwards and backwards in each group the Wilcoxon test was used. Digit forwards scores were significantly higher than digit backwards scores for the TC-S (Z=-3.9; p=.000), for the TC+S (Z=-3.4; p=.001) and for the HF+S group (Z=-3.2; p=.001).

Table 4.18 Medians,(modes) ranges and Kruskal-Wallis test statistics of digit forwards and digit backwards for TC-S, TC+S and HFA+S.

	TC-S (n=22) Median (mode) and ranges	**TC+S (n=21) Median (mode) and ranges**	**HFA+S (n=18) Median (mode) and ranges**	**(H value) *p* value**
Digit forwards	7(8) 2-12	6(6) 2-9	7 (8) 3-10	H=6.632 p=.036* TC-S>TC+S**
Digit backwards	3(3) 0-8	4(4) 0-8	4(3) 0-8	H=1.34 p=.509

****=significance at p<.05***
*****=significance at p<.016***

With respect to the sustained attention task, out of 61 children, 4 (18%) TC-S, 5 (23%) TC+S, and 10 (55%) HFA+S completed the task only for the first few minutes (the task lasted for 16 minutes) refusing to carry on to the end of it. Thus, their data were not included in the analysis because too little of the test was completed to produce reliable estimates of the children's attention span.

As described in section 3.2.6 for those who completed the test (18 TC-S, 16 TC-S and 8 HFA+S) each individual response sheet was scored for omission and commission errors. When non-parametric tests were performed non significant group effects were found. Median, mode, ranges and Kruskal-Wallis test are showed in Table 4.19.

Table 4.19 Medians, (modes) ranges and Kruskal-Wallis test statistics of omission and commission errors as reported by children in the TC-S, TC+S and HFA+S groups in the total time of the sustained attention task.

	TC-S (n=18) Median (mode) and ranges	**TC+S (n=16) Median (mode) and ranges**	**HFA+S (n=8) Median (mode) and ranges**	**(H value) *p* value**
Omission errors (0-16 minutes)	3 (1) 0-16	6 (0) 0-23	1.5 (0) 0-13	H=3.42 p=.180
Commission errors (0-16 minutes)	1.5 (0) 0-7	2 (0) 0-21	1 (1) 0-14	H=.364 p=.833

**=significance at p<.05*
***=significance at p<.016*

To investigate whether or not a decline in performance occurred, the sustained attention task was divided into two time periods: 0 to 8 minutes and 9 to 16 minutes. When the Wilcoxon test was used, surprisingly TC-S showed a significant increment in performance (less omission errors) during the second 8-minutes period, whereas no significant difference in terms of omission errors were found between the first and second 8 minute period in TC+S and in HFA+S group. Details of the sustained attention task for each group are given in Table 4.20.

Table 4.20 Medians, (modes) ranges and Wilcoxon's signed-rank test of omission and commission errors as reported by children in the TC-S, TC+S and HFA+S groups in the first and second 8 minutes of the sustained attention task.

	TC-S (n=18) Median (mode) and ranges	**TC+S (n=16) Median (mode) and ranges**	**HFA+S (n=8) Median (mode) and ranges**	**(Z value) *p* value**
Omission errors (0-8 minutes)	2 (0) 0-10	3 (1) 0-13	1.50 (0) 0-9	TC-S Z=-2.00, *p*=.045*
				TC+S Z=-.239 *p*=.811
Omission errors (9-16 minutes)	1 (0) 0-6	2 (2) 0-14	1 (0) 0-5	
				HFA+S Z=-.542 *p*=.588
Commission errors (0-8)	1 (0) 0-3	1 (0) 0-3	0.50 (0) 0-6	TC-S Z=-.478, *p*=.633
				TC+S Z=-.-071 *p*=.943
Commission errors (9-16 minutes)	0 (0) 0-7	1 (0) 0-21	0 (0) 0-6	
				HFA+S Z=-.333 *p*=.739

=significance at p<.05
****=significance at p<.016***

The number of wanted signals (a maximum of 32 letters or wanted signals could be identified) recognised by children appeared to improve significantly with age in the TC-S (U=14.00; p=.025) and TC+S (U= 9.00; p=.022) but not in the HFA+S group (U=4.500; p=.365). In fact 15 TC-S and 15 TC+S aged 4-10 recognised respectively a median of 27 (mode=27, range=6-32) and 24 (mode=24; ranges=9-31) wanted signals whereas the remaining 7 TC-S and 6 TC+S aged 11-16 reported slightly better scores with a median of 31 wanted signals (mode=31, ranges 26-32) for the TC-S group and a median of 30 wanted signals (mode=29, ranges 22-32) for the TC+S group. HFA+S children also tended to improve with age in the sustained attention task. Thus, 11 HFA+S children aged 4-10 reported a median of 28 wanted signals (mode=25, ranges 25-31) whereas 7 children aged 11-16 recognised a median of 31 wanted signals (mode=32 ranges 18-32). To verify if mental abilities were associated with vigilance capacity, a series of Spearman's correlations were run between GMA, verbal and spatial cluster scores and all the sustained attention task variables (wanted signals, omission and commission errors in the total time, first and second 8 minutes) but no significant relationships were found in the groups ($p > .05$).

4.2.6 Behavioural functioning in children with and without sleep problems

Strengths and Difficulties Questionnaire (SDQ)

Children with autism showed more problematic behaviour than children in the TC-S and TC+S group. Multiple comparisons in fact demonstrated that HFA+S scored significantly higher on total deviance score (U=56.000; p=.000), peer problems score (U=41.500; p=.000) and significantly lower on pro-social behaviour score (U=59.000; p=.000) compared to TC-S. Also HFA+S scored significantly higher on peer problems score (U=66.000; p=.000) and significantly lower on pro-social behaviour score (U=60.500; p=.000) than TC+S. No significant differences were found between TC-S and TC+S in the total and subtest scores. The medians, ranges and Kruskal-Wallis test statistics of total and subscales score can be seen in Table 4.21

Table 4.21 Medians, ranges and Kruskal-Wallis test statistics of total and subscales scores as rated by parents in the SDQ for TC-S, TC+S and HFA+S.

	TC-S (n=22) Median and (ranges)	**TC+S (n=21) Median and (ranges)**	**HFA+S (n=18) Median and (ranges)**	**(H value) *p* value**
Total deviance score	13 (3-22)	14 (6-28)	19 (11-29)	H=12.7 *p*=.002* HFA+S>TC-S**
Emotional symptoms score	2 (0-7)	4 (0-10)	5(1-9)	H=4.30 *p*=.116
Conduct problems score	2 (0-4)	3 (0-8)	3 (0-9)	H=4.96 *p*=.084
Hyperactivity score	4(3-8)	4 (3-7)	5(3-9)	H=2.74 *p*=.253
Peer problems score	3 (0-4)	3 (0-6)	5 (2.-9)	H=18.8 *p*=.000* HFA+S>TC-S, TC+S**
Pro-social behaviour score	8(5-10)	8 (3-10)	5(1-7)	H=16.2 *p*=.000* HFA+S<TC-S, TC+S**

=significance at p<.05
****=significance at p<.016***

However it appeared that also children's sleeplessness problems perhaps were related to daytime behavioural problems. According to Goodman (1997) scores could be banded in normal, borderline and abnormal (these categories are described in section 3.2.6, Table 3.4). In the TC-S group, the mean scores for all SDQ variables (except for peer problems score which was categorised as borderline because it was 0.3 points above normal value) were in the normal range. Whereas in the TC+S and HFA+S groups, mean scores for all the SDQ variables were banded borderline or abnormal (except for hyperactivity and pro-social behaviour scores which were categorised as normal in the TC+S group). The number of children in the normal, borderline and abnormal band can be seen in Table 4.22.

Table 4.22 Type of behavioural problems according to the SDQ percentage and (number) of children in the TC-S, TC+S and HFA+S group reported by mothers as being in the normal, borderline and abnormal bands.

	Normal			Borderline			Abnormal		
	TC-S	TC+S	HFA+S	TC-S	TC+S	HFA+S	TC-S	TC+S	HFA+S
Total deviance score	81% (18)	47% (10)	16% (3)	-	14% (3)	11% (2)	18% (4)	38% (8)	72% (13)
Emotional symptoms score	66% (14)	42% (9)	38% (7)	4.5% (1)	19% (4)	11% (2)	31% (7)	38% (8)	50% (9)
Conduct problems score	72% (16)	47% (10)	33% (6)	18% (4)	19% (4)	38% (7)	9% (2)	33% (7)	27% (5)
Hyperactivity score	72% (16)	81% (17)	50% (9)	18% (4)	14% (3)	27% (5)	9% (2)	4.8% (1)	22% (4)
Peer problems score	45% (10)	38% (8)	5.6% (1)	18% (4)	19% (4)	16% (3)	36% (8)	42% (9)	77% (14)
Pro-social behaviour score	81% (18)	81% (17)	38% (7)	18% (4)	9% (2)	22% (4)	-	9% (2)	38% (7)

The proportion of TC+S and HFA+S whose total deviant score, peer problems and pro-social behaviour scores were rated as borderline or abnormal was higher than the proportion of TC-S in these bands. However it is important to highlight that when total and subscale means were compared to UK normative data (Meltzer, Gatward, Goodman, & Ford, 2000) obtained from 10298 parents (see Table 4.23) all three groups reported higher mean score than that for the normative sample (TC-S scored just above the average level of performance on the conduct problem scale which was only 0.1 higher than that for the normative sample).

Table 4.23 Means and standard deviations of total and subtest scores of the SDQ for normative sample and for TC-S, TC+S and HFA+S.

	NORMATIVE SAMPLE (n=10298) Mean (sd)	TC-S (n=22) Mean (sd)	TC+S (n=21) Mean (sd)	HFA+S (n=18) Mean (sd)
Total deviance score	**8.4 (5.8)**	11.6 (5.0)	14.6 (6.3)	18.6 (4.7)
Emotional symptoms score	**1.9 (2.0)**	2.8 (2.4)	4.2 (2.7)	4.3(2.2)
Conduct problems score	**1.6 (1.7)**	1.7 (1.3)	2.9(2.1)	3.4 (2.7)
Hyperactivity score	**3.5 (2.6)**	4.6 (3-8)	4.4 (1.2)	5.2(1.7)
Peer problems score	**1.5 (1.7)**	2.3 (1.6)	3.0 (1.8)	5.5 (1.9)
Pro-social behaviour score	**8.6 (1.6)**	7.2 (1.7)	7.3(1.9)	4.7(1.7)

4.2.7 Associations between parental mental state and children's sleeplessness problems

General Health Questionnaire (GHQ-12)

All mothers of all 61 children completed the questionnaire. Five (22%) TC-S, 12 (57%) TC+S and 7 (38%) HFA+S parents scored at or above the cut-off point of 12 (χ^2=4.83, *df*=2, *p*=.089). However it is important to highlight that studies have found considerable variations in term of sensitivity and specificity for different threshold values (Golberg et. al., 1997).

Mothers of TC-S who completed the GHQ-12 questionnaire reported a score ranging from 2 to 16 (median=7) whereas parents of TC+S and HFA+S reported a score ranging from 3 to 34 (median=14) and from 5 to 18 (median=10) respectively. When the groups were compared using Kruskal-Wallis test a significant difference was found between the groups (H=12.7; *p*=.002). Subsequent multiple-comparisons revealed that parents of TC+S scored significantly higher (U=86.000; *p* =.001) than TC-S. Although HFA+S reported a median total score 3 points higher than that of TC-

S, no significant differences were found between the groups at the .016 level (U=106.00; p=.033). Also when mothers of HFA+S children were compared with mothers of TC+S no significant differences were revealed (U=126.000; p=.121).

To determine if parental mental state was associated with children's sleeplessness a zero order correlation between total score of the GHQ-12 and CSDI was run. The relationship between these two variables was also examined with total score of SDQ as control variables. The zero order and first order correlation coefficients and p values can been seen in Table 4.24

Table 4.24 Correlation between measure of mothers' stress derived from GHQ-12 and children's measure of sleep derived from CSDI: Spearman's r coefficient and p value of zero and first order coefficients.

Zero order correlation	**TC-S sleep**	**TC-S behaviour**	**First order partial correlation controlling for children's behaviour**	**TC-S sleep**
Total GHQ-12	.65 p=.792	-.268 p=.267	**Total GHQ-12**	.056 p=.824
	TC+S sleep	**TC+S behaviour**		**TC+S sleep**
Total GHQ-12	.367 p=.102	.547 p=.010**	**Total GHQ-12**	-.257 p=.824
	HFA+S sleep	**HFA+S behaviour**		**HFA+S sleep**
Total GHQ-12	.471 p=.077	.601 p=.018*	**Total GHQ-12**	.286 p=.321

****=significance at p<.05***

*****=significance at p<.016***

There was little association between children's severity of sleeplessness and mothers' stress either before or after controlling for children disruptive behaviour in the three groups. It seems likely that additional variables may be suppressing any relationship.

In the TC+S group, total GHQ-12 score was significantly related to total SDQ (r=.547; p=.010). As expected, more challenging children's daytime behaviour (reflected in higher total deviant score) was associated with higher level of maternal mental distress. Also in the HFA+S group, total score of SDQ was positively related with parental mental distress ($r=.601; p=.018$).

4.3 DISCUSSION

The results of this study indicated that sleep problems are common in childhood and suggested that a mild form of sleep disturbance may frequently affect typical children and to a greater extent children with autism. The most frequent sleeplessness problems reported by mothers of TC+S and HFA+S were difficulties going to bed, settling to sleep and night wakings. Children with sleeplessness (with and without autism) complained of intrusive and uncontrollable thoughts during the pre-sleep period significantly more than children without sleeplessness problems. This indicated that cognitive factors are associated with sleeplessness in childhood.

Creativity abilities of TC+S and HFA+S were compromised compared with normal values and also general cognitive functioning was reduced in children with sleeplessness. While children with autism showed significantly more daytime behaviour problems than typically developing children, the proportion of TC+S and HFA+S whose behaviour was classified as borderline or abnormal tended to be higher than TC-S. Thus, it might be that sleeplessness difficulties affected negatively daytime behaviour in typical developing children and aggravate behaviour problems seen in children with autism. Finally, mothers of TC+S suffered the negative effects of having a child with reduced sleep but surprisingly a similar result was not found in the HFA+S group.

Prevalence rates and type of sleeplessness problems

The sleep problems prevalence rates, in the current study was comparable to those obtained in previous studies with typical school age children (Owens, Spirito, McGuinn et al., 2000) and children with autism (Patzold et al., 1998; Wiggs & Stores, 2004). As expected, sleep problems tended to be more prevalent in young typical children (occurring in 66% of TC+S aged from 4 to 8) but this result also suggested that high rates of sleeping difficulties are still reported in children over the age of 4. High sleep problem rates were found in older age autistic children (occurring in 44% of HFA+S aged from 4 to 8 and 44% aged from 9 to 12). According to previous literature, many sleep problems in infants and children, if not promptly treated, may develop into chronic sleep disturbance (Kataria et al., 1987). Furthermore certain intrinsic and extrinsic risk factors seen to be associated with autism (e.g challenging behaviour, family stress) may impede HFA children to overcome early childhood sleep problems. The fact that sleeplessness problems reported by parents of HFA children had not recently appeared but difficulties were present in the past was confirmed by the high percentage of HFA children (83%) with a sleep problem in the past which was still present at the time that the study was carried out.

Sleeplessness problems most frequently reported by parents were difficulties going to bed and settling to sleep (settling problems at bedtime and settling to sleep problems were reported respectively by 95% and 66% of parents of TC+S and 77% and 72% of parents of HFA+S). Beside HFA+S and TC+S were also shown to wake up at night for a time significantly longer and more frequently than TC-S.

Similarly, on the CSHQ, total sleep disturbance score, sleep onset delay and number of night waking subscales scores were significantly higher in the TC+S and HF+S groups than TC-S sample. These results are consistent with recent studies that showed: a) that sleep onset delay and night waking problems persist and are common in typical childhood (Steinet al., 2001); and b) that difficulties in initiating sleep and long lasting night waking are also frequent problems in HFA children (Richdale and Prior 1995). Other problematic aspects of sleeplessness reported by parents in the CSHQ (bedtime resistance, sleep reduction and sleep anxiety) also differed between the groups. In particular children in the TC+S group were regarded by their parents as being not easy to get to bed and sleeping for a significantly shorter time than those children in the non-sleeplessness problem group (it is curious that such a difference was not found between TC-S and HFA+S and the reason for this inconsistency remain unclear). What might account for this result was that children in the TC+S group were approximately one year younger than children in the TC-S group. On one hand the increased dependence of younger children on their caregivers at bedtime, and on the other hand the fact that bedtime resistance and sleep reduction subscales target children's sleep autonomy might have inflated these subscale scores. With regards to the HFA+S group, it is possible that diagnoses such as autism rather than age might account for significantly higher sleep anxiety scores reported by parents of HFA+S children compared to parents of TC-S (although no difference between HFA+S and TC+S were found). This result is in accordance with previous research where anxiety-related behavioural features were prominent in parental reports of the sleep of children with autism (Wiggs and Sores, 2004).

Of interest, however, is the finding that the CSHQ subscale scores for TC-S were slightly elevated (except for bedtime resistance and number of night wakings) compared to scores published for a normal community sample from USA (Owens, Spirito, & McGuinn, 2000). In addition daytime sleeplessness subscale scores reported by parents of TC-S more closely resembled the scores of children referred to a sleep clinic. When considering this result it is important to be mindful of the fact that insidious social changes may lead to modifications in the accepted 'norms' of child sleep, within the Western world. For example, a study suggests that early school start times are associated with children having reduced amounts of sleep and, in turn, impaired academic performance (Epstein, Chillag, & Lavie, 1998). As another example, computer game playing, internet use, television viewing, possession of mobile phones have all been linked with reduced sleep and reduced opportunities for sleep (Tazawa & Okada, 2001; Van Den Bulk, 2004) in adolescents. Although parents of TC+S and HF+S reported problematic sleep behaviour on CSHQ, only sleep onset delay, sleep reduction and daytime sleepiness scores were above the Owens' clinic sample values.

This result reflects a discrepancy between CSDI used as selective criteria to identify children with sleeplessness problems (Wiggs & Stores, 1998a) and scores on the CSHQ. This is not surprising since a similar incongruity between CSHQ and generic parental sleep reports has been described (Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008). It might be that the specificity of the CSHQ questions and the opportunity to respond to numerous questions regarding several aspect of sleep provided more accuracy than CSDI elicited. However the CSDI has been successfully used in a number of studies (Montgomery et al., 2004; Richman & Graham, 1971) have allowed researchers to group childhood sleeplessness in broad categories according to the frequency and duration of the sleep problems and have permitted to make comparisons with other works.

Parental reports of child's sleeplessness appeared not to be influenced by socio-demographic factors. As shown in Table 4.2, mothers or fathers in the three groups were all employed and so able to provide education and support for their children. Adverse family factors including parenting standards and socioeconomic circumstances such as homelessness (Sheldon, Dekker & Levy, 1991) are also likely to be important for the insurgence of sleep problems in childhood although they have been rarely studied.

Children's report of sleep problems

In this research, children in the three groups acknowledged the sleep problems reported by parents since parental and child's reports were positively correlated. This result suggests that parents did neither neglect nor overestimate the children's sleep difficulties. Interestingly TC+S and HFA+S complained of intrusive and uncontrollable ruminative thinking during pre-sleep period significantly more than TC-S. Consistently, Espie, Brooks, & Lindsay (1989) reported that the cognitive items of the Sleep Disturbance Questionnaire ("My mind keeps turning things over", "my thinking takes a long time to unwind" and "I am unable to empty my mind") were the most highly rated by adults with insomnia. Also, Lichstein & Rosenthal (1980) showed that people with insomnia frequently experience excessive cognitive activity whilst trying to get to sleep. The authors asked a large sample of adults with insomnia to make a categorical judgement as to whether cognitive disturbance, somatic disturbance, both or neither were primary in the cause of their sleep disturbance. Cognitive disturbance was ten times more likely than somatic disturbance to be cited as the main cause of the insomnia.

The fact that, in the current study, children with sleeplessness also experienced mental over-activity during the pre-sleep period provided evidence that not only in adulthood (Nicassio et al., 1985) but also in childhood cognitive factors may be responsible for the maintenance of sleeplessness. Individuals who suffer from insomnia tend to be very preoccupied about their sleep and about the daytime consequences of not getting enough sleep. These excessive worries and ruminations trigger automatic arousal and emotional distress and plunge the individual into an anxious state (Harvey, 2002). Such arousal has been treated effectively with adults using cognitive therapy (Morin et al., 2006). Thus, it may be worth considering applying, with the appropriate age-related variations, cognitive behavioural theories to children's sleeplessness with a view to the development of specific cognitive therapies which may be helpful for children whose sleeplessness is not helped by behaviour therapy alone.

Objective sleep measure outcomes

Actigraphy appeared to be an acceptable method of quantifying the sleep of children with and without autism. One week of sleep monitoring using sleep diaries and actigraphy did not detect any significant group differences in any of the sleep variables. The discrepancy between parental report and objective sleep recording might be due to the fact that parental reports are subjective measures of sleep and consequently open to contamination from a variety of sources such memory failure and parental tiredness (Sadeh et al., 1991). However significant positive correlations between CSHQ sleep scales and actigraphy measures for bedtime were obtained in the three groups. Also significant correlations coefficients were acquired for wake up time and night waking duration in the HFA+S group. This result provides evidence of significant concordance between subjective and objective sleep measures although the group differences reported by parents in the CSHQ were not found by actigraphy measures. This is not surprising since parental knowledge of sleep events and the objective measurement of these events might differ (Sadeh, 2004). Also, parental oversensitivity to sleep difficulties could have made them overestimate their children's sleep problems. This may be true especially for parents who have a child with autism; a study has suggested that objective sleep measurements of children with autism were similar to that of normal children although they were referred by their parents as having sleep problems (Hering et al., 1999). However there are a number of factors which indicate that this is unlikely to be the case. Firstly, there did appear to be an association between parent and child reports. Children in the sleeplessness groups (who were of an age where they could complete the questionnaire) corroborated parental reports and described themselves as having trouble getting to sleep. The researcher also stressed how important it was that children expressed their own ideas in the questionnaires. Secondly, actigraphs could have overestimated sleep time if the children were awake but still in bed. Sadeh (1994) suggested that the discrepancy between actigraphic data and parental reports are mainly due children's inability to soothe themselves into sleep without parental assistance. Thus, those children with sleep problems can be distinguished only by the fact that they make their parents aware of the fact that they are awake. This interpretation suggests that treatment designed to alter the children's behaviour or cognition upon finding themselves awake might be of benefit. Finally, the fact that the most common parental complains (bedtime resistance, settling to sleep) were not confirmed by the actigraphy results is most likely due to the fact that many of children's settling difficulties took the form of refusal to go to bed which occurred before the child wore the monitor.

Associations between creativity and sleeplessness in children with and without autism

The results found in this study showed a difference in terms of creativity between autistic and typical children. In fact, the HFA+S scored significantly lower on total creativity score and originality than TC-S. Of particular interest, however, is the finding that such differences in terms of total creativity and originality scores were not found between HFA+S and TC+S and between TC-S and TC+S. Perhaps a reasonable explanation for such findings might be based on the combination of a) the impoverished creativity seen in the autistic condition, and b) on the creative impairment seen in the sleep restricted condition. Thus the poor creative ability found in children with autism might be due not only to the autism specific deficit but also to a mild form of sleeplessness.

Different theories can be used to explain the creative deficit seen in children with autism. The *executive function theory* explains the imagination deficit in autism in terms of difficulties in planning and disengaging from routine action. For instance, there are studies (Leevers and Harris 1998) that have shown that children with autism find it difficult to draw a novel picture, for which they have never developed a procedure. The weak point of this theory is that it also predicts other deficits, such as fluency and flexibility on which HFA+S in this study did not seem to be more impaired than TC+S and TC-S. Also this theory does not justify why no differences in term of creativity scores were found between HFA+S and TC+S.

Others suggest that creative impairment in autism is an expression of the *theory of mind* deficit which is a prerequisite in the understanding of pretend play and mental states in others. This theory also referred to as *imagination deficit theory* implies that the deficits in communication and social understanding which have been associated with a theory of mind abnormality, may turn out also to be connected to the problems in imagination and creativity (Craig & Baron-Cohen, 1999). Although this theory explains the lack of originality and also a global creativity impairment which were seen in this study in HFA+S, again this theory *per se* does not explain why differences in terms of creativity were not found between HFA+S and TC+S.

The results of the current study raise the possibility that a mild form of sleeplessness is sufficient to induce impairments which mirror the underlying autistic-creative deficit. A control group of HFA without sleeplessness, which is difficult to compile since the majority of them suffer from sleeplessness problems, would help to explain this relationship further.

Although there are no studies that have looked at the association between creativity and sleeplessness in autistic children, the literature on sleep in typical children supports the idea that sleeplessness can impair creativity as other studies have shown that verbal creativity and abstract thinking were affected by a single night of restricted sleep (Randazzo et al., 1998). However, in contrast to this result, another study (Healey & Runco 2006) has indicated that highly creative children experience more sleep difficulties than non-creative children. This study suggested that the "activity of the mind" associated with creativity contributes to the development of sleeplessness. On the contrary, the results of the current study show that TC+S and HFA+S who suffer mental over-activity were less creative than TC-S. Perhaps it is not only the quantity of mental activity but also the nature of the thinking during pre-sleep which reduces creativity and causes problems of falling asleep in children. Considerable research attention has been directed toward delineating the content of pre-sleep cognitive activity in adults but there is a lack of research with children. Studies indicated that family, work (Watts, Coyle, & East, 1994) general worries, attempts to solve problems (Harvey, 2000) thoughts of getting out of bed (Wicklow & Espie, 2000) were contents of pre-sleep cognitive activity in adults. Thus, to find out the nature of the thoughts that keep awake children at night and prevent them from sleeping might have important implication for their creativity, since high level cognitive functions appear to be particularly sensitive to the effects of disturbed sleep.

The fact that sleeplessness problems might negatively affect creative ability in children is also confirmed by the fact that originality, fluency, flexibility and elaboration of TC+S and HFA+S seemed to be compromised compared with normal values. In fact TC+S and HF+S scored below the average level of performance in all the creative features measured (except for originality score which was 1 point above the average value in the TC+S group) whereas TC-S creativity scores were at or above the average level of performance. However poor elaboration scores were found to be far below the normative level in all groups. Several reasons might account for the poor elaboration ability in the three groups. Firstly, the time limit might have prevented children from adding details to their pictures; secondly, the instructions might have not sufficiently encouraged children to embellish their drawings. On the contrary, children were allowed enough time to complete their pictures. For instance, a flexible time limit (over the 10 minutes suggested in the Torrance Directions Manual, 1974) was adopted, especially with younger children. Also specific instructions for administering test activities (Torrance, 1974) were used. These instructions included expression such as " after you think of an idea keep adding to it and build it up" or "put as many ideas as you can in each picture" which encouraged children to add new elements to their pictures.

Poor creative ability found in children with sleeplessness cannot be attributed to discrepancy in the mental ability since all three groups reported general mental ability rated as normal. In addition general, verbal and spatial ability scores were not correlated with the creative measures in the TC-S and TC+S group. On the contrary total creativity score and originality were positively correlated with general mental ability for HFA+S group suggesting that cognitive skill might have a role in the creativity of children with autism.

The combining and flexibility tasks did not show significant differences between the groups. With regards to the combining task, all children seemed to be capable of joining two shapes in order to make a new object. This task required the ability to a) mentally represent a new object, and b) to execute it in reality. This result is consistent with a previous study in which no significant difference was found in terms of combining ability between typical children and children with autism (Craig et al., 2001). However, when children were asked to recombine two previously combined shapes more TC+S and HFA+S than TC-S tended to commit an inflexible error giving the same answer or by not responding at all. This result in accordance with previous studies where lack of mental flexibility has been proven to be a consequence of sleeplessness in typical children (Randazzo et al., 1998) and also a feature of autism (Craig & Baron-Cohen, 1999) which perhaps could be made worse by inadequate amounts of sleep.

However, it is important to highlight that the lack of differences between the three groups in the combining and flexibility task may be due to the fact that the test was unable to measure responses above its ceiling and was compromised by a lack of variability. The tasks in fact contained a limited number of trials which might have been insufficiently challenging. This was a result of designing the tests to ensure that children with autism had every chance to pass. Thus, the limited nature of the tasks might a) have prevented measurement of the true mean or average score of the groups, and b) have created an artificial clustering since the majority of scores were at the maximum possible for the tests in the three groups.

The flexibility task was also scored for total number of responses and number of responses occurring only once. Although the percentage of rare answers given by TC+S and HFA+S resembled those provided by TC-S group, children with autism reported a reduced number of total responses compared with the remaining two groups. This result confirms the reduced fluency ability seen in children with autism in the TTCT (Craig & Baron-Cohen, 1999).

This study also investigated whether narratives produced by children with sleeplessness (with and without autism) contained less imaginary and real events than the stories invented by children without sleeplessness. As predicted, no group differences were found on the length of narrative in the test and pre-test conditions. This result confirmed the absence of significant group discrepancy in terms of verbal ability tested by means of BASII. However a significant impairment in the production of real facts by TC+S was found. When an impossible card was used, TC+S were less likely to introduce real elements into a narrative than TC-S (although no differences between TC-S and HFA+S were observed). This result is consistent with the impaired performance on the verbal version of the TTCT seen in sleep deprived typical children (Randazzo et al., 1998). Certain aspects of verbal processing, including verbal generation have been shown to activate the frontal lobe, thought to regulate creative ability, in addition to other language brain areas. Thus, it is possible that lack of sleep reduced the function of this area and caused a verbal creativity deficit. What remains unclear is a) why no differences between TC-S and HFA+S were observed, b) why no difference was found between TC-S and TC+S in terms of imaginary elements, and c) why impairment in number of real facts was not found in TC+S when a real card was used. This might be due to the fact that the themes offered in the unreal or impossible cards elicited more the invention of hypothetical events than the real cards.

The story-telling task set out to retest the earlier findings that children with autism have a deficit in the production of an imaginative story (Craig & Baron-Cohen, 2000) was not proved, with neither real nor unreal cards. This discrepancy between Craig & Baron-Cohen's study and the present one may be due to the children with autism in this research having a higher verbal ability (autistic children's verbal mental age was lower than their chronological age in the Craig and Baron-Cohen (2000) study).

The lack of group differences in the story telling task might have been due to the small sample sizes. A further possibility is that no autism-specific deficit was seen here because the cards were inappropriate to this sample's developmental level. In fact, some of the cards depicted adults which may have not promoted the children's process of identification in the character. Also, it may be that the cards were not sufficiently ambiguous to activate any imaginative process. The same argument applies to the simplified tests used by Leevers and Harries (1998) and Craig et al. (2001) who did not find significant differences in terms of representation of unreal entities between typical children and children with autism.

Of interest, however, is the finding that all three groups reported a very poor imagination in the real and unreal card condition. According to Gardner (1982), as children begin school, they enter into a stage of development where they learn conformity and consequently creativity appears to decline. However others have argued that decreased creativity may appear at different ages and for various aspects of divergent thinking (Runco, 1999). What might account for the TC-S lack of imagination is perhaps the fact that they reported daytime sleeplessness scores in the CSHQ that resembled scores of children referred to a sleep clinic. Sleep deprivation might cause emotional distress, feeling of confusion, irritability and anxiety (Hall et al., 2000) which are psychological states not conducive to creativity and imagination. An experimental work with psychiatric patients and normal subjects showed that anxiety and rigid defence mechanisms did not facilitate creative functioning (Gudmund & Ingegerd, 1983).

Cognitive profile of typical developing and autistic children with and without sleeplessness problems

Using the BASII, the patterns of cognitive abilities were examined in the three groups. With regards to means scores, it was found that TC-S, TC+S and HFA+S obtained a GMA score respectively of 94, 98 and 94 and the three groups' overall performance was classified "average" according to the classification ratings of the BASII. However all groups fell towards the lower end of average range (90-109). In terms of cluster scores, all groups obtained verbal and spatial scores also categorised as average. Thus clusters and GMA scale scores were homogeneous in the three groups, contrary to previous research where the cognitive profile of children with autism has been seen as differing from typical children (Joseph, Tager-Flusberg, & Lord, 2002). However, the BASII delineated differential patterns of cognitive abilities in the three groups. The cluster scores difference provides a measure of strengths and weaknesses in children's mental skills and might be indicative of a specific learning difficulty. For instance higher verbal and spatial ability and lower non-verbal ability scores are often due to working memory difficulty that are typically found in children with dyslexia (Ott, 2007). Thus, discrepancy factor which refers to a gap between cognitive abilities is a key aspect in the diagnosis and assessment of children's mental functioning so as to ensure that they receive adequate and specific educational support on the basis of their cognitive profile. In this study, discrepancies (V-S, G-V, G-S) were relatively infrequent in the TC-S (except for V<S which occurred in about 18% of cases) compared to TC+S and HFA+S which occurred in a higher number of cases and also seemed to be slightly in favour of spatial ability in the case of children with autism. This is in line with previous studies where spatial ability has been found superior to verbal ability in the autistic group (Lincoln et. al., 1988).

It is important to be cautious when interpreting differences between cluster scores because fairly large differences between clusters are not unusual in the normal population (Bishop & Butterworth, 1979). It is also important to highlight that in this study the majority of children with autism (n=10) did not show significant verbal-spatial discrepancy. On the contrary a Wechsler (2004) IQ profile with verbal IQ depressed relative to performance IQ and peak subtest score on Block design has traditionally been associated with autism (Happe´, 1994) and has even been suggested as a possible diagnostic aid (Lincoln, Courchesne, Kilman, Elmasian, & Allen, 1988). The lower rate of autistic children with uneven intellectual abilities found in this study compared to previous research might be due to age-dependent factors. In fact there are studies that show that the relative weakness in verbal ability exhibited on standardised cognitive measures by younger and more able children with autism is to some extent due to age and may reflect the effect of the developmental delays in speech and language that lessen over time in some children (Joseph et al., 2002).

Studies have found differences between verbal and spatial abilities in children with neurological disease (Brookshire et al., 1995) developmental disorders (Mervis, Morris, Bertrand, & Robinson, 1999) and reading comprehension difficulties (Nation, Clarke, & Snowling, 2002) however whether discrepancies in children with sleeplessness occur, remain constant, become more apparent over time or resolve following sleep treatment is not known. No studies to date have addressed these issues in children with lack of sleep. Also influences on the severity of the sleep problems on verbal and spatial discrepancies have not been examined. Thus, it is not known whether children with more severe sleep problem demonstrate larger discrepancies in their verbal-nonverbal abilities. The fact that in this study, nearly half of the TC+S (n=10) showed a discrepancy which was equally rated in the V>S and V<S directions might be indicative of a general cognitive functioning reduction. Of course, this finding begs the question of why a pattern of inferior performance in one cognitive domain is present in some TC+S but not in others? It might be speculated that unevenly developed skills which are the outcomes of fundamental differences in neurobiological substrates (Bodary & Miller, 2000) and which reflect impairments in different areas of functioning (e.g. verbal, spatial, social, and behavioural) are made worse by lack of sleep.

Neurobehavioural functioning in children with and without sleeplessness problems

A number of studies have suggested that small reductions of sleep alter neuro-behavioural functioning in children and that memory and attention seem to be affected by lack of sleep (Allen, 2003; Dahl, 1996; Randazzo et al., 1998; Sadeh et al., 2003). The current research, in which TC-S performed better on digit forwards tests than TC+S, appeared to be in accordance with these previous findings. However such differences in terms of digit forward scores was not found between TC-S and HFA+S. A reasonable explanation for such peculiar result might be based on the fact that digit span task has been known to be a peak ability in those autistic children who are not mentally retarded (Joseph et al., 2002) which could have override the lack of sleep. However more recently a discrepancy of performance among working memory-related tasks in autism spectrum disorders has been established with a study (Bennetto, Pennington, & Rogers, 1996) finding an impaired memory function in autism. In the current study the digit backwards scores were significantly lower than forwards digit span scores not only in TC+S and HFA+S group but also in TC-S, so a reduction in memory ability cannot be explained by lack of sleep in this sample. However it might be possible that since TC-S group reported daytime sleeplessness scores on the CSHQ similar to scores reported by children referred to a sleep clinic, they also showed reduced memory. This explanation also applies to the lack of dissimilarity in term of digit backwards scores seen in TC-S, TC+S and HFA+S.

Although no significant difference was found between the groups in the sustained attention task it is important to note that 55% of HFA+S took part in the task only for few minutes and refused to carry on. Such drop-out could be indicative of a reduced attention span. It might also be that the monotonous and repetitive nature of the task caused distress to the HFA+S. Studies investigating the attention of children with autism have found that their attention span is maintained for activities that interest the child (Attwood, 1998) enabling the children to sustain attention for a considerable length of time. Attention to tasks and activities that are outside their range of interest, however, is likely to be much shorter, momentary or non-existent (Wing, 1998). This behavioural trait displayed by children with autism has been described in the literature as selective attention (Sohlberg & Mateer, 1989).
Interestingly and in contrast to another study where reduced attention ability was observed over time (Gale & Lynn, 1972) an increment in performance following the first 8 minutes period was found though it was significant only in the TC+S group. The fact that there was a tendency in all three groups to perform better in the second 8-minutes period might be due to the fact that children needed to familiarise themselves with the test and so used the first minutes to learn how to do it. Although this result is intriguing and unexpected since decrease in performance is a typical feature of the vigilance task, it must be acknowledged that "attention" is a complex concept, and the present task might not be a real measure of children's attention skills in real life. The failure to correlate performance with GMA and the findings of improved wanted signals scores with age confirmed a) early findings indicating the possible existence of attention as an independent factor from intelligence (Ware, 1961) and b) developmental study showing improved attention ability in older children (Gale & Lynn, 1972).

This study suggests, as others have found (Allen, 2003), that mild forms of sleeplessness are associated with daytime behaviour difficulties. Children with autism reported significantly more daytime behaviour problems than the two groups of typically developing children. This result is not surprising since disruptive behaviour is one of the features of autism, however it was also noted that TC+S tended to have higher total deviance score, emotional symptoms and conduct problems scores compared to TC-S. Children with sleep onset delays and problems sleeping through the night have been found to be at more risk of behavioural problems (Wiater et al., 2005). Similarly, other studies have found that sleep problems are associated with aggressive behaviour (Achenbach, 1991; Richdale, Francis, Gavidia-Payne, & Cotton, 2000) emotional difficulties (Gregory & O' Connor, 2002) poor communication (Quine, 1991) in typical children and in children with an intellectual disability though some research studies have provided contradictory findings to those that have been reported (Clements et al., 1986; Wiggs & Stores, 1996a). Despite emotional symptoms and conduct problems tending to be higher in TC+S and HFA+S, surprisingly such a trend was not found in terms of their hyperactivity scores; severely sleep deprived children have been seen to manifest their lack of sleep in day-time over-activity (Guilleminault, Korobkin, & Winkle, 1981) and the fact that such a result was not found in the current study might be explained by the fact that children reported mild form of sleeplessness which did not impact severely on daytime behaviour. A study that investigated the relationship between severity of sleep problems and behaviour difficulties, in fact, found that children who were rated as severe had higher scores on the behaviour problem index (Cunningham et al., 1986) while those children with mild ratings reported reduced behaviour problem score (Quine, 1992). Similarly, another study found that children with sleep problems showed significantly more types of challenging behaviour (irritability, hyperactivity, stereotypical behaviour) and challenging behaviour of a greater severity than children without sleep problems (Wiggs & Stores, 1996a). As was expected, HFA+S reported higher peer problem scores and lower pro-social behaviour scores than TC+S and TC-S. Children with autism have severe communication problems and difficulties in making friendships. They might appear odd to the other children and might be isolated (Wing & Gould, 1979).

When the total and subscale scores of the SDQ were compared to UK normative data, although all three groups reported rates above (below for the pro-social scale) the average normative score, TC+S and HF+S reported rates far higher (lower for the pro-social scale) than that of normative sample. In addition, the pattern of results with the majority of TC-S in the normal band and TC+S and HFA+S in the borderline or abnormal category is clearly indicative how a mild form of sleeplessness affects negatively daytime behaviour in typical children and exacerbate behaviour difficulties seen in children with autism.

The effects of children's sleeplessness on the mother's mental health

As mentioned in the introduction, when children's sleep is disturbed it is frequently the case that parents are affected too. The result of this study suggests that child sleeplessness not only impaired the children's daytime behaviour but also their parent's mental state. The results indicated that parents of TC+S suffer the negative effects of having a child with sleep problems since they reported higher score on the GHQ-12 than TC-S. Similarly, studies have reported that scores on maternal depression were significantly higher in mothers of typical children who had a sleep problem as compared to those who had a child without sleep problem (Hiscock & Wake, 2001a; Scott & Richards, 1990). This finding has also been reported in a study where maternal stress was associated with settling problems, night waking problems and with co-sleeping problems of children attending mainstream schools (Quine, 2001). However, nearly half of mothers of TC+S (n=9) scored below the cut off point used to identify the presence of current emotional distress and also maternal mental state did not appear to be associated with children's sleep. What might account for this result is the limited psychometric property of the GHQ-12 as a screening questionnaire for psychiatric morbidity (Hankins, 2008) however several research works have establish that this is a valid and reliable instrument to detect symptoms of psychological distress (Cano et al., 2001; Goldberg et al., 1997).

Another possible explanation might rely on mothers' personality or psychological traits. Perhaps those mothers who did not report scores above the critical threshold were more able to cope with their children's sleep problems and maybe had more safeguards against stress. Despite the current research work not providing evidence for the association between parents' personality and stress response, other studies have found that mothers with personal traits such as neuroticism or introversion experience a negative mood state in response to their child's sleep problems (Gelman, Jory & Macris, 1998).

A similar explanation might apply to parents of children with autism who scored below the critical value of 12 and who reported neither significant difference in terms of mental health when compared to mothers of TC-S nor association between their mental state and children's sleep. Parents of HFA+S who might have learn to adapt to the demands of having a child with autism are more likely to tolerate any problems and to be less affected by sleep difficulties. There is evidence that less anxiety is experienced by those who believe to have control over a problem (Houston, 1972) and also that those who use maladaptive parental coping strategies are more at risk of depression (Hall et. al.,1991).

An investigation into the coping strategies used by parents would have helped to provide a better understanding of this issue. It is also possible that the children's characteristics, school support and the family's socio-economic status might account for this result. For example, studies found that the stress experienced by parents of children with multiple disabilities seems to be influenced, by child's behavioural problems (Baker, Blacher, Crnic, & Edelbrock, 2002) night-time disturbance (Wiggs, 2007) and by the family's social and economic circumstances (Quine & Pahl, 1985). With regards to the children's characteristics the fact that the children's mental ability was within the average level, and that they were affected by a mild form of sleeplessness might explain the reduced stress level found in the majority of HFA+S mothers. There is, moreover, ample evidence that the presence of behavioural problems in children with intellectual disability represent a significant stressor to parents behind stress arising from other child's factors. Previous studies have suggested that children with sleep problems are more likely to display more behaviour problems (Aronen et al., 2000). Although a similar pattern was seen in this study, the majority of HFA+S children (n= 15) reported conduct problem scores that were categorised as normal or borderline which might explain the low level of parents' stress.

It is also possible that the mother's reduced stress level in the HFA+S group was influenced by school support received by their children. Autistic children were in fact attending mainstream school, where support was provided by individual help, or special educational units for children with PDD. Similarly, Bristol (1987) found that the positive adaptation of mothers of autistic children was predicted by adequacy of social support.

Also parents appeared to have a good level of education as the majority of them occupied positions (e.g. professionals, teacher etc.) which required at least a secondary education. Thus, an adequate cultural and economical level which helped parents to have a better understanding of the issues related to the autistic condition and to have the economic resources necessary to educate their child, could perhaps explain the reduced GHQ-12 scores found in mothers of HFA+S children.

4.3.1 Limitations

The limitations of this research need to be taken into account when considering the results and the ensuing implications of this study. The response rate implies that the results may not be reflective of the population at large. Out of 900 families invited to take part in the study, only 91 (10%) parents of typical children and 28 (3%) parents of children with autism agreed to participate.

It was not possible to enquire about the non-responders since the individual names of children who were contacted through mainstream schools were not made available to the investigators for confidentiality reasons. What is surprising is the very low response rates of parents who had an autistic child despite Oxfordshire Autism Support Service, Oxfordshire Autistic Society for Information and Support (OASIS) and National Autistic Society showing willingness to have their members informed about the study. With the high prevalence, intensity, and negative impact of sleep problems in children with autism, one would expect more parents to be involved in the research. This low rate of response may be due to the fact that parents of children with autism do not have time to be involved in research due to other more prevalent demands. In addition an intended fourth group of children with high functioning autism and no sleeplessness (HFA-S) could not be compiled. It is possible that the screening questionnaire might have appeared most interesting and relevant only to families where children had a sleep problem. However this is unlikely to be the case since the information sheet accompanying the questionnaire stressed the importance of the participation of children both with and without sleep problems. On the contrary it is probable that the difficulty of recruiting autistic children who did not display sleep difficulties is due to the fact that most children with autism have sleeplessness problems. This result confirms Inanuma's (1984) findings, that autistic children may be particularly likely to develop sleep problems and the reason for this needs to be explored.

However, it is important to recognise that a control group in a pervasive development disorder population is likely to be different from a control group in a normal population. For example, the highly individualised problems and abilities of children in the ASD might have lead one to question whether HFA-S and HFA+S groups should be contrasted and compared or whether the difference in terms of measured abilities in these two groups are due to sleep problems or other confounding variables. However the fact remains that the difficulty to recruit HFA-S children has limited the interpretation of these results.

A further limitation was the fact that CSHQ scores for the TC-S group were elevated in comparison to scores for the normal community example and that daytime sleeplessness subscale scores resembled those reported by the clinic sample. These factors might have diminished the difference between the three groups. In general, it appears that children have reduced opportunities for sleep in the modern society. For instance, computer game playing, internet and mobile phones use have all been associated with reduced sleep in childhood and adolescence (Tazawa & Okada, 2001; Van Den Bulk, 2004).
Thus, the current study provides preliminary information on the association between sleep and creativity that warrants further research with more representative sub-groups. Additionally, research in relation to parental recognition of sleep problems in children is warranted. It is important to know why some parents will view their child's sleep disturbance as a problem, yet others will not.

The results of this study, as well as other studies (Craig et al., 2001; Leevers & Harris, 1998) in the field of autism, do not provide a clear answer to whether or not children with autism are impaired on imaginative or reality based creativity. Nevertheless, future research needs to use numerous and more complex tasks to assess creativity which will help to establish whether autism is associated with lack of imaginative or reality-based creativity, or both. This study shows, however, that a verbal task can be used with children with autism to measure their verbal creativity ability.

Another issue to be considered was the broad age range. The upper age range of the sample could have been limited to 10 years old in order to minimise the possible effects of pubertal changes on sleep behaviour. Also it could have been useful to assess whether there are patterns of sleep difficulties that are specific to a given age and how these sleep problems are associated to creative ability. Thus, this study needs to be replicated with a larger sample to allow separate estimates by age. In addition this study did not address possible differences in terms of sleep timing on school versus non-school nights. In future studies it would be important to investigate the differences in sleep between week days and the weekend, especially in older children who tend to stay up later at night and sleep longer in the morning at the weekend.

This study had some methodological weaknesses which could be overcome in future research. First, as described, the three groups achieved nearly the maximum score possible in the combining and flexibility task; this meant that differences between the groups could not be detected, if they existed. It would be desirable to avoid such a ceiling effect in the future. Second, the large dropout rate (55%) reported by children with autism in the sustained attention task indicated that this was not an appropriate task to measure attention span in these children. Another study has also shown that autistic children's difficulties in sustaining attention on imposed tasks may be attributable to the motivational contingencies of task rather than to a primary impairment in the ability to sustain attention (Garretson, Fein, & Waterhouse, 1990). Thus, the task pleasure may have been increased in this study. For instance, children's motivation could have been improved through the use of rewards for completing the 16 minute task. Also the presentation of the wanted signals at a rate of one per minute and the duration of the task could have been reduced. Children might show a decrease in attention or refuse to continue a task when it has a long time to completion or requires the child to be alert to receive continuous stimuli (Akshoomoff, 2002). Thus, the child starts to fail, or miss items, anxiety increases and performance declines.

Another issue that needs to be addressed is the fact that the sleep diary was only used to interpret actigraphy data. Perhaps statistical analysis of sleep diary data would have unravelled the reason for the lack of consistency between objective recording and parental reports. However recent studies (Sadeh, 2008; Werner, Molinari, Guyer, & Jenni, 2008) have shown that parents' reports are not always a good way to collect information about children's sleep patterns. For instance, older children are less likely to require attention when they wake up at night, and therefore their parents are less likely to be accurate in documenting their night-wakings.

Many environmental factors may exert influences on sleep and activity. Among these are noise (Horn, Pakhurst, Reyner, Hume, & Diamond, 1994) temperature (Muzet, Libert, & Borbely, 1988) and sleep setting (Ophir-Cohen, Epstein, Tzschinsky, Tirosh, & Lavie, 1993). Daily logs which provided essential information for editing the actigraphic data could have also reported child subjective sleep related experience so as to enhance the understanding of the children's cognitive factors associated with sleep problems. However, given the young age of some participants, it was not always possible to involve them in completing the diary.

The assessment of children's objective sleep might have been improved by following children over a prolonged time period because some of the factors that might cause errors in actigraphic assessment are then held constant (Chambers, 1994). For example if a child was habitually restless and the actigraph therefore underestimates sleep, this error will remain constant and actigraphy will accurately assess changes in estimated sleep efficiency but not the absolute number of minutes slept. However, in the current study, children wore the monitor for an average of 6 or 7 nights which is commonly considered in relevant literature to be a sufficient time period to assess sleep in children.

Also the actigraphs failed to record or retain the sleep data in 4 children with HFA. Because some devices have shown sensitivity to humidity or temperature and also failed for unknown reasons (Sadeh et al., 1995) it might be that the actigraphs used in this research stopped working for similar reasons.

It could be also speculated that children with autism perhaps have impaired the functionality of the actigraphs. Given that HFA children have a systemising tendency, meaning the drive to analyse the variables in a mechanical system (Baron-Cohen, Richler, Bisarya, Gurunathan, & Wheelwright, 2003), and that the Basic Mini-motionlogger has a battery that is easy to remove, it might be that children with autism who felt the need to understand how the actigraph worked, in some way compromised its functionality. However the fact that children with autism might have damaged the actigraph's functionality remains only a hypothesis since there is no specific evidence of this. Future studies implementing more resistant and modern devices are needed.

4.3.2 Positive aspects

Despite these drawbacks, this study has a number of positive aspects. Firstly, the use of both objective and subjective tools for assessing the children's sleep has enabled the results of this study to provide a comprehensive and thorough account of the sleep problems faced by children with and without autism. Standardised, widely used measures of sleep were used in the current study. The results also have important implications for any studies evaluating interventions by suggesting a need to monitor objective measures of child's sleep as well as parental report to establish whether intervention actually results in improved child sleep or merely in reduced signalling behaviour that make parents aware of the fact that children are awake. In addition, the findings of this study confirm that actigraphs are, in general, an appropriate tool to assess sleep in children with autism despite the monitor failing in some cases.

Secondly, this study has examined the association between sleep disturbance and pre-sleep arousal in children. In contrast to a growing knowledge base in adults, only very few studies have investigated the cognitive factors associated with insomnia in typical children (Gregory et al., 2008) and no study had looked at this association in atypical children. The current study is the only research to have addressed this issue in children with autism and to have showed that TC+S and HFA+S frequently experience excessive cognitive activity whilst trying to get to sleep. As mentioned in the discussion of this chapter, should this result be replicated and extended, it might be worth considering whether cognitive behavioural therapy successfully used with adults can be adapted for use with children.

Thirdly, this study is one of the few to have used children's reports of sleep. The majority of sleep studies (Beltramini & Hertzig, 1983; Honomichl et al., 2002; Laberge et al., 2001; Schreck et al., 2004; Smedje et al., 1998) are in fact based on parental reports whereas only a limited number of works have examined self-reported sleep in children (Gaina et al., 2004; Gregory et al., 2008; Ward et al., 2008). In the current work, children were a precious source of information since they allowed the unveiling of the cognitive component associated with lack of sleep in this population. Also, this study reveals how children with HFA can complete a questionnaire about their sleep patterns. This indicates that they were able to reason about the causes that keep them awake at night. In addition, children's questionnaires were important to confirm parental reports. Future research aiming to develop statistically sound questionnaires to assess the attributions made by children with sleeplessness about the cause of their problem are needed.

Fourthly, this research study constitutes one of the few attempts to look at objective sleep and cognitive and behavioural functioning in children. The results of this study have in fact significant clinical and educational implications. They highlight the need for parents and professionals to be aware of the consequences of sleeplessness. This is perhaps the first study to have analysed cognitive discrepancy (e.g. verbal vs spatial) in children with sleeplessness problems and to have found partial results showing uneven mental ability in children with sleeplessness difficulties. Poor quality and quantity of sleep can lead to decrements in daytime functioning that may negatively impact on a child's wellbeing and quality of life.

Fifthly, a further positive aspect of this research study is that it was diagnostically specific. The current study was in fact specifically addressed to children with autism in contrast with previous investigations which included a diagnostic heterogenic sample (Robinson & Richdale, 2004) limiting interpretation regarding specific disorders and placing restrictions on the conclusions being made. However this limitation is common in the literature about individuals with intellectual disability (Didden & Sigafoos, 2001) and a heterogeneous group is often required due to low participation rates and the low frequency of many developmental disorders.

Finally, most important is the fact that this study has shown how lack of sleep might reduce high cognitive functions such as creativity in children. Only two studies have investigated the association between creativity and sleeplessness in typical children but these works either used limited measure of sleep (Healey & Runco, 2006) or applied an acute sleep restriction paradigm (Randazzo et al., 1998). On the contrary the current study has evaluated the relationship between sleep and creativity by means of well established measures in a naturalistic setting.

This study also represents a first attempt to understand the relationship between creativity and sleep in children with autism. Despite previous works having investigated sleep (Hoshino et al., 1984a; Schreck et al., 2004) and lack of creativity in children with autism (Craig et al., 2001; Scott & Baron-Cohen, 1996), no study has looked at the relationships between these two factors in this population. In this study, HFA+S demonstrated creative deficit. This is an important preliminary result that requires future investigation with the inclusion of an HFA-S group. This study has also contributed to extend the limited research about creativity in autism and have confirmed those studies supporting the creative deficit theory of autism (Craig & Baron-Cohen, 1999). In addition new tests such as combining and flexibility task and the story telling task were designed to test for creativity and to see whether or not narratives produced by children with sleeplessness (with and without autism) contained fewer imaginary and real events than the stories invented by children without sleeplessness. These tests, with the opportune modifications could be used in future research to clarify the current findings.

These results raise the question of whether children with a lack of sleep who present low creative ability could benefit from better management of their sleep schedule. This will be explored in the next chapter.

4.4 SUMMARY

The results suggested that typical children and children with autism might frequently be affected by a mild form of sleep disturbance. Difficulties in going to bed, settling to sleep and night wakings were sleeplessness problems most frequently reported by mothers. Of interest, however, is the finding that the CSHQ subscale scores for TC-S were elevated compared to scores published for a normal community sample (Owens, Spirito, & McGuinn, 2000). This suggests that sleep deprived children are becoming the "norm" in the Western society.

In the current study, children with sleeplessness (with and without autism) experienced mental over-activity during the pre-sleep period. This provided evidence that not only in adulthood (Nicassio et al., 1985) but also in childhood cognitive factors may be responsible for the maintenance of sleeplessness.

Despite no differences being found in terms of objective sleep measures, impaired creativity scores were found in the sleeplessness groups. This was confirmed by the reduced creativity of TC+S and HFA+S when compared with normal values. Of interest, however, is the finding that all three groups reported a very poor imagination in the story-telling task.

Given the reduced creativity found in children with autism only when compared with the TC-S group, it might be speculated that a mild form of sleeplessness is sufficient to induce impairments which mirror the underlying autistic-creative deficit. A control group of HFA without sleeplessness, which was difficult to compile since the majority of them suffered from sleeplessness problems, would help to explain this relationship further.

In this study, uneven cognitive abilities were relatively infrequent in the TC-S compared to TC+S and HFA+S which occurred in a higher number of cases. Also the memory of TC+S and attention of HFA+S were reduced compared to these cognitive abilities in TC-S. This might be indicative of a general cognitive functioning reduction in sleepless children. Children in the HFA+S group reported significantly more behavioural problems than the remaining two groups. However when the total and subscale scores of the SDQ were compared to UK normative data, the pattern of results with the SDQ total and subscale mean scores in the normal band for TC-S and borderline or abnormal band for the TC+S and HFA+S group perhaps indicates that a mild form of sleeplessness negatively affects daytime behaviour in typical children and exacerbates behaviour difficulties seen in children with autism.

The results suggested that parents of TC+S suffer the negative effects of having a child with sleep problems whereas parents of HFA+S who might have learned to adapt to the demands of having a child with autism were less affected by their children's sleep difficulties. Whether the association between sleep and creativity can be manipulated by means of treating sleeplessness problems in children is addressed in the next chapter.

CHAPTER FIVE

Phase two: Booklet-delivered behavioural treatment for sleeplessness problems in typically developing children and children with high functioning autism: does it affect creativity?

5.1 INTRODUCTION

Previous studies have suggested that insufficient sleep negatively affects memory, attention (Allen, 2003; Dahl, 996) and creativity (Randazzo et al., 1998) in typically developing children and also exacerbates autistic symptoms (Schreck et al., 2004) as well as causing maternal depression (Gregory et al., 2005).

Many of these adverse affects can be reversed by removal of the sleep disturbance as a means of perhaps improving overall function of the child. Reductions in children's neurobehavioural problems (Seymour, Bayfield, Brock, & During, 1983) and improvements in parental mental health (Szyndler & Bell, 1992) have been associated with successful behavioural therapy for sleeplessness problems. As described in section 2.4, behavioural intervention is the treatment of choice for most sleeplessness problems in TC (Owens et al., 1999) and children with autism (Schreck, 2001) however whether or not reduced sleeplessness results in improved creative ability in children with and without autism is unknown.

Although there are a number of behavioural treatments for sleeplessness problems which have been empirically validated with TC (Mindell et al., 2006) and used with children with HFA (Weiskop et al., 2005) these studies have typically involved conventional therapist-driven face-to-face delivered therapy and only a limited number of studies have investigated the efficacy of intervention delivered by a written advice booklet only.

As described in detailed in section 2.4.5, a self-administered booklet has been found to be as effective as face-to-face intervention for the treatment of sleep problems in TC infants (Weymouth et al., 1987) and pre-school children with learning disabilities

(Montgomery et al., 2004) with positive outcomes maintained at six months follow-up and a high percentage of parents declaring to be satisfied with the written booklet intervention (Montgomery et al., 2004). These results suggest that parents can successfully utilise self-help information about sleep-related behavioural interventions without the need for professional help. Such treatment has important cost implications. Given the increasing demands on healthcare providers in most countries (Gidman, 2006), services might be inadequate to meet these needs and methods of delivering the most cost-effective interventions to the greatest number of patients is worthy of investigation. Therefore, booklet-based interventions might be a solution to this increasing healthcare request, because they can be administered to a large number of people with minimal costs (Montgomery et al., 2004).

However the methods of delivery of behavioural advice are an area which needs further investigation. Given the usefulness of booklet-based behavioural intervention found with infants, this study aimed to investigate whether or not this method of delivery can be also effective in the treatment of sleep problems in older children. Furthermore, as children with autism are frequently reported by parents as having experienced sleep difficulties (Richdale & Prior, 1995) they are therefore a group for whom identifying a method of delivery which can reach a large number of parents and be easy to use would be beneficial.

However, several factors may affect the effectiveness of a booklet-based behavioural intervention for sleep problems. For example, in Phase One of this study, it was found that sleep-related cognitive factors were associated with the presence of sleep disturbances in TC and HFA children which may require more individualized attention than written information can provide. Also, children with autism have a number of special characteristics which could make it difficult to implement behavioural treatment via self-help intervention without extra support.

The second phase of the study therefore had the following aims:

Primary goal

1. Assess the efficacy of a booklet-delivered behavioural treatment for sleeplessness in school age typical and autistic children.

Secondary goal

2. Assess the effects of successful booklet-delivered behavioural treatment for sleeplessness problems on creative thinking.
3. Describe parents' evaluation of the booklet-based behavioural treatment for sleeplessness problems (in children with and without autism).

5.2 METHODS

5.2.1 Design of Phase Two

An initial intention to use a randomised controlled trial to assess the efficacy of the booklet-based behavioural treatment was substituted with a multiple baseline design. This was considered a more appropriate design given the reduced number of participants in the TC group and HFA group recruited to Phase Two.

A multiple baseline design with a varying time schedule was used. Participant families were randomly assigned to one week or three week baseline assessment. After this initial assessment which was implemented during Visit one and two, all mothers received the same booklet on behavioural treatment and were asked to use it with their child for a six week intervention period (from Visit two to Visit three). Three months after completion of the intervention a 1 week follow up was conducted with all the parents. A more detailed description of the methods used in Phase Two is reported in Chapter Three.

5.2.2 Participants

A total of 29 children with sleeplessness problems (16 TC and 13 HFA) aged 4-11 were recruited from the pool of subjects whose mothers took part in Phase One (see section 3.1.3). Parents of these children were sent a letter of invitation (Appendix Nine) and an information sheet (Appendix Ten) to take part in a booklet-delivered behavioural intervention for childhood sleeplessness.

However not all the families contacted took part to the behavioural intervention trial. Mothers of 11 TC did not proceed past visit two for various reasons (n=3 did not respond to contact from researcher; n=1 had moved without any forwarding address;

n=2 declined because the family was too busy and n=5 refused to participate without giving a clear explanation).

With regards to mothers of children with autism, n=3 did not proceed past visit two because of reluctance to engage in treatment for fear of worsening the child's behaviour; n=2 began the intervention trial after five weeks from the baseline period (despite many telephone calls to each mother when they were encouraged to begin the treatment) so they were not included in this analysis because too much time elapsed between visit two and visit three and consequent data were not useful; n=2 parents refused because they were too busy and n=2 were not interested in booklet-based behaviour treatment.

As a result, mothers of 5 TC and 4 HFA with sleeplessness problems completed the study. Descriptive details of participants are in Table 5.1

Table 5.1 Children's name, group, age, gender, type and severity of current sleeplessness problems

Subject	Group	Age	Gender	Type and severity of current sleeplessness problem
Esme	TC	4:4	F	SEVERE settling to bed/sleep
Lucy	TC	6:4	F	MILD settling to bed/sleep
Teddy	TC	7.8	M	SEVERE settling to bed/sleep MILD night and early morning waking
Jack	TC	4:2	M	SEVERE night waking and co-sleeping MILD settling to bed/sleep
Ben	TC	6:2	M	SEVERE night waking and co-sleeping MILD settling to bed/sleep
Dominic	HFA	4:10	M	SEVERE settling to bed/sleep, night waking and co-sleep
Andrew	HFA	10:3	M	SEVERE settling to bed/sleep
Joseph	HFA	7.10	M	SEVEREsettling to bed/sleep
Ross	HFA	7:11	M	SEVERE settling to bed/sleep MILD night waking

5.2.3 Procedure

At Visit one and two, previously described measures were administered and these were presented and discussed in Chapters Four and Five. Only measures which are pertinent to the intervention are described below.

Visit one

Mothers were interviewed to gather descriptive information and also asked to complete a questionnaire which included the *Composite Sleep Disturbance Index*, (CSDI) and *Autism Screening Questionnaire* when more than 10 days elapsed between the first contact and visit one. Parents were also given a *Sleep Diary* to complete for the successive one or three weeks depending on which baseline length they were assigned to. Children were given a sleep questionnaire which consisted of a modified version of the *Sleep Disturbance Questionnaire* (MSDQ). They were also given an actigraph to wear for a minimum of 5 consecutive nights during the first week or third week of the three week baseline assessment. Also during visit one the *British Ability Scale II* (BASII) was used to assess children's general mental ability (GMA) and verbal and spatial skills

Visit two

One, or three, weeks later the sleep diary, sleep questionnaires and actigraphs were collected. Children were also administered the figural form of the *Torrance Test of Creative Thinking* (TTCT) and *Story telling task.*

Booklet-delivered behavioural intervention

Those parents who decided to participate were sent a booklet containing information about behavioural techniques for children with sleeplessness problems and asked to use the strategies described for a six-week period as explained in more detail in section 3.1.4. They were also asked to keep a sleep diary for the duration of the intervention phase. For those families who felt that this would not be possible or for those that failed to keep diaries adequately during weeks 1 and 2 families were asked to monitor at least for the third week of treatment (to allow comparison across families).

Visit three (Post-treatment)

After six weeks, a post intervention sleep assessment was carried out (MSDQ, 1 week sleep diary, actigraph). Mothers were also given a *Questionnaire to evaluate the treatment.* Children's creative abilities were reassessed as for visit two (TTCT, story telling task). One week later the sleep diary, sleep questionnaires and actigraphs were collected.

Follow-up

Three months later a postal follow-up assessment consisting of a 1 week sleep diary was sent to mothers.

5.3 RESULTS

5.3.1 Statistical analysis

Data were analysed for individual children and the CSDI was used to assess the severity of the children's sleep problem (see section 3.2.3). The main outcome measures used to evaluate the behavioural intervention efficacy were derived from the *sleep diary.* Thus, data obtained from parental diaries were graphed in order to investigate the effects of treatment and to provide a more personal and qualitative view of each child's sleep over the baseline, intervention, post intervention and follow-up period. Each graph was tailored to the particular sleep problem shown by that particular child. Visual analysis of the graph data was used to analyse the following sleep variables:

1. number of problems settling to bed and to sleep;
2. duration of settling to bed and sleep;
3. number of night wakings;
4. duration of night wakings.
5. number of early morning wakings

For every sleep variable graphed, the following comparisons were made: baseline versus intervention, baseline versus post-intervention and baseline versus three months follow-up (Thackeray & Richdale, 2002; Weiskop et al., 2005). To determine reliability of the visual analysis, two independent raters (one of whom was not associated with the study) examined each graph according to a set of modified criteria (Weiskop et al., 2005) on the following scale:

1. Substantial improvement with data showing a significant increase in terms of frequency or duration of the variable (e.g. settling problem, night wakings and early wakings went from every night or more than three times a week to one/zero time per week; duration of settling problem went from more than one hour to less than 30 minutes; duration of night waking went from more than 30 minutes to less than 15 minutes).
2. Moderate improvement with data showing a clear increase in terms of frequency or duration in the variable but not sufficient to be considered a substantial change (e.g. number of settling problem, night waking and early waking went from two times per week to one/zero time per week; duration of settling problem went from 30 minutes to less than 15 minutes; duration of nigh waking went from 15 minutes to less than 5 minutes).
3. No change when data appeared unmodified from baseline to intervention
4. Moderate deterioration with data showing a clear decrease in terms of frequency or duration in the variable. Thus, the number of settling problem, night waking and early waking went from one/zero times per week to two times per week; duration of settling problem went from less than 15 minutes to 30 minutes; duration of night waking went from less than 5 minutes to 15 minutes.
5. Substantial deterioration with data showing a significant decrease in term of frequency and duration in the variable. Thus, settling problems of night waking and early waking problems went from one/zero times to every night or more than three times a week; duration of settling problem went from less than 30 minutes to more than one hour; duration of night waking went from less than 15 minutes to more than 30 minutes.

Graphs were not labelled, but the raters were given an indication of the desired direction of change to determine whether an increase was related to an improvement or deterioration. Percentage agreement between the independent raters was calculated and if they disagreed on a rating they conferred and came to an agreement. A total of 31 graphs were rated and 72 comparisons between phases were made. Before conferring the two raters agreed on 65 (90.2%) of the comparisons.

Bedtime, sleep latency, sleep duration, night waking and wake up time obtained for each child from parental diary and actigraph were compared at baseline and intervention phase to assess differences between subjective and objective measures of change in sleep behaviour. Subjective ratings of change in sleep were also examined from children's responses to the MSDQ.

To assess change in creative ability, originality, fluency, flexibility and elaboration scores obtained from TTCT were compared at baseline and at post-intervention and to a normative sample (Torrance, 1974).
Story telling task was used to assess if narrative produced by children improved at post- intervention. The story telling task consisted of three phases (pre-test, test and post test). The stories invented at pre-test and test phase were scored on number of words, introduction of real facts and imaginary elements whereas at post test scores were assigned for ability to distinguish between real and unreal pictures. Details of the story telling task are presented in section 3.2.5

5.3.2 Typical children group

Case 1: Esme
Esme was a 4:4 year old typically developing child who lived with her parents and 9 year old brother with autism. She spent all the mornings at home with her mother who was a housewife, and attended a nursery school for few hours in the afternoon. Her father was a secondary school teacher.

Based on the descriptive category labels for General Mental Ability (GMA) scores of the BASII, Esme's overall performance was classified in the average range (108), spatial ability score above was classified as average (112) and a verbal ability score in the average (102) range. According to the BASII, for this child, there was no significant discrepancy between the verbal and spatial ability scores.

Esme was described by her mother as exhibiting difficulties in settling to sleep by herself. This problem had persisted for more than 2 years and was rated as severe because of its nightly occurrence and because it lasted for more than one hour. Of specific concern was the fact that each night the child called out for attention and 'whinged', disturbing all the family members unless the mother stayed in the child's bedroom cuddling and hugging her while she fell asleep. Sometimes this led the mother having to wait for the child to fall asleep late in the evening. As a result of this bedtime behaviour, the mother was becoming increasingly frustrated with her child's sleep and concerned that her child's behaviour at bedtime could worsen. The child's mother had never sought professional help thinking erroneously that her child's sleep behaviour would improve with age.

At the time of the study Esme was not on any medication.

Assessment of changes in sleep behaviour: Esme

One week baseline sleep diary recording indicated that every night the child had problems with settling to sleep and took a mean of 30 minutes to fall asleep. After approximately eight days of intervention and at post-intervention there was a MODERATE IMPROVEMENT in the child's settling to sleep difficulties (see Figure 5.1 and Figure 5.2). Esme did not demand maternal presence to fall asleep at the beginning of the night but only disturbed her parents when she needed to go to the toilet. However data shows a mild upward trend in settling problems at 1 week post-intervention and at 3 months follow-up. According to her mother, Esme relapsed into bad sleep behaviour because the child was beginning to attend full time nursery school and she was feeling anxious about being without her mother for more than a few hours.

As shown in Table 5.2 there were no changes for mean values of bedtime, sleep latency, sleep duration, minutes of night wakings and wake up time from baseline to post-intervention according to both sleep diary and actigraph data. Diary records were consistent with actigraphy scoring except for sleep latency. The activity monitor data suggested that Esme might have fallen asleep 19.8 minutes earlier at baseline and 25 minutes earlier at post-intervention than that indicated by her mother in the sleep diary. In addition total sleep time as recorded by actigraphy (minutes between sleep onset and sleep offset minus minutes awake) increased by 1 hour and sleep efficiency improved by 8.3% .

When Esme was asked to complete the child version of the sleep questionnaire (MSDQ) she was not able to understand the questions and consequently data are not available for this child.

These results should be interpreted with caution because the sleep diary was not completed for the whole 6 week intervention phase but only for the second week. Also actigraphy data are based on only 1 night of recording at post-intervention as Esme was not keen to wear the monitor at night. On the contrary the values of the actigraphy parameters at the baseline period are based on 7 nights of sleep recording.

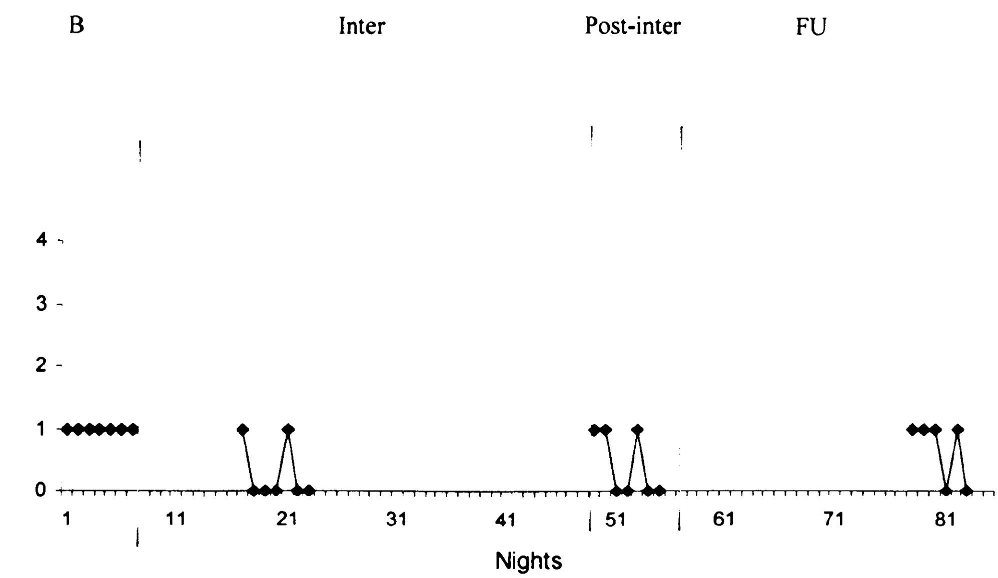

Figure 5.1 Number of problems settling to sleep during baseline (B), intervention (Inter), 1 week post-intervention (post-inter) and follow-up (FU) based on diary recording, for Esme.

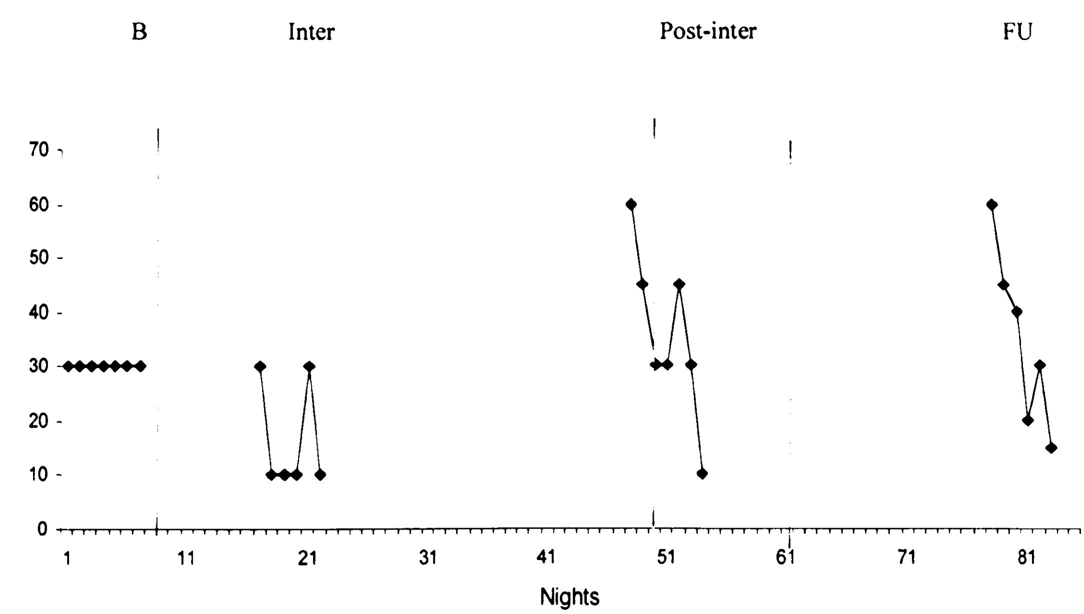

Figure 5.2 The amount of time (minutes) taken to fall asleep during baseline (B), intervention (inter), 1 week post intervention (post-inter) and follow-up (FU) according to diary recording, for Esme.

Table 5.2 Mean (sd) of sleep variables assessed by diary and actigraphy for Esme

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.13	20.40	30.00	10.20	9.73	9.95	15.00	25.00	6.95	6.47
	(.19)	(6.47)	(0.00)	(5.50)	(0.82)	(50.36)	(5.00)	(2.00)	(0.73)	(47.90)
Post-intervention	19.85	20.27	35.00	10.00	10.30	10.00	13.00	16.00	7.14	7.00
	(.28)	(0.00)	(15.9)	(0.00)	(0.69)	(0.00)	(3.00)	(0.00)	(0.53)	(0.00)

Assessment of change in creativity: Esme

Esme showed a slight improvement in all the creativity measures of the TTCT following intervention. Creativity scores increased by 68 points for total creativity, 15 points for originality, 25 points for flexibility, 12 points for fluency and 16 points for elaboration. However when these scores were compared to a normative sample (based on 1365 primary and secondary children) her scores in all creative measures (except for flexibility) were below the average level of performance (see Table 5.3).

Esme appeared to have improved in the story-telling task following intervention (see section 3.2.5 for details of the story telling task and scoring criteria). With respect to the pre-test, there was a reduction of 2 words from baseline to post intervention and the number of real facts (3) introduced did not change at the two time points. At post intervention, Esme used 39 more words, introduced 2 real facts and 1 imaginary element ("*the invisible road fell down*") in the reality based story. Improvement was also noted when she was asked to invent a story based on an imaginary card. At baseline she refused to tell a story and stated: "*I can't think of anything*" though she was stimulated and encouraged. On the contrary, at post-intervention she made up a story with 52 words introduced 4 real facts but did not add any imaginary elements to her account. She was able to discriminate between real and unreal cards.

Table 5.3 Creativity scores based on TTCT at baseline and post-intervention compared to normative means (standard deviation) for Esme

	NORMATIVE SAMPLE (n=1365) Mean (sd)	Baseline score	Post-intervention score
Originality score	**28 (10)**	8	23
Fluency score	**21 (6)**	4	16
Flexibility score	**16 (5)**	0	25
Elaboration score	**68 (26)**	0	16

Case consideration-Esme

After implementing booklet-delivered behavioural intervention for eight days, Esme's mother reported that her child's settling problems slightly decreased. Esme learned to stay by herself in her bedroom and demanded less maternal comfort to fall asleep. Her mother stated, *"Generally Esme is better at going to sleep, however we get the odd nights when she will not settle easily"*. This result suggests that young children can learn within only few days how to behave at bedtime. The fact that this moderate improvement was not maintained at post-intervention and at 3 months follow-up might be due to diverse reasons. Esme's mother reported that she had erroneously started to respond again to her child's request for attention at bedtime and did not proceed consistently with the booklet's advice. Esme was, in fact, being integrated full time at the nursery school and was going through a tough time. Thus, her mother felt that it was better not to expose her daughter to additional frustration by neglecting her requests of cuddles and hugs at night. Parents find it stressful to ignore their distressed children at bedtime and are often unwilling to use this method with them (Hall and Nathan, 1992).

Also Esme's mother was feeling exhausted as she was parenting an older son with autism and sleeplessness problems. Perhaps being exhausted interfered with the implementation of the behavioural intervention in the long run. Thus, Esme's mother returned to use negative sleep-related interaction with her child to stop protest crying.

But, if parents respond to their child's demands of attention after a certain amount of time, he/she will only learn to cry longer the next time and will not develop frustration tolerance. In fact, the amount of time that Esme took to fall asleep at post-intervention and 3 months follow-up exceeded baseline level.

Esme was not able to complete the sleep questionnaire (child form) despite maternal attempts to explain the questions to her. This is not due to poor mental skills as her cognitive performance was classified in the average range according to the BASII. Perhaps statements listed in the questionnaire such as "I can't empty my head of thoughts at bedtime" or "my body feels tense" or "I feel upset if I don't sleep" or questions "Is it hard for you to go to bed?" required an understanding of abstract concepts (e.g. tense, upset) which a child of her chronological age might find difficult to grasp.

Maternal reports and objective sleep measures were congruent except for sleep latency which Esme's mother tended to overestimate. However, the discrepancy between maternal and objective recording in terms of sleep latency was due to the fact that Esme's mother attached the actigraph to her wrist after she was already in bed for a while; Esme was reluctant to wear the monitor and needed to be convinced to put it on when bedtime occurred.

The fact that Esme accepted to put on the monitor for only 1 night at post-intervention might reflect Esme's effort to have her mother's attention. In fact, each night, the child made her mother go into her bedroom more than once to attempt to attach the monitor to her wrist.

Following the intervention phase, Esme improved in all the creativity measures of TTCT, though they remained below the average level of performance (except flexibility). Improvements were also noted in the story-telling task.

Case 2: Lucy

This participant was a 6:4 year old girl who lived with her parents (mother was a housewife and father was a secondary teacher) and two younger brothers. During the day Lucy attended primary school. According to the BASII, the child had an average level of mental ability (109). When looking at subscale scores, however, verbal ability was classified as above average (115) and spatial ability in the average range (99). As indicated by BASII a difference of 16 points between verbal and spatial scores was significant at .05 level, it is therefore possible to conclude that, for Lucy, spatial ability was a relative weakness compared to verbal ability although average when compared to the general population.

Lucy had difficulties with settling to sleep. The mother indicated that this problem was life-long and was rated as mild because it occurred one or two nights a week for up to thirty minutes. The mother's main concern was the fact that the child complained of being incapable of relaxing and stopping thinking at bedtime. Although she did not disturb her parents when unable to sleep, she kept a light turned on until late to read in her own bedroom. This behaviour disrupted her sibling's sleep with whom she shared the room. The mother also reported that her daughter appeared tired during the day and struggled to wake up in the morning to go to school. Parents had attempted to help her giving a reward each time she went to sleep early; however this method appeared not to be successful.

Lucy was not on medication.

Assessment of changes in sleep behaviour-Lucy

Visual analysis of the graphs indicated that there was a MODERATE DETERIORATION in settling to sleep when the baseline was compared to intervention and post-intervention and three months follow up. According to Lucy's mother, difficulties settling to sleep worsened ten days after baseline. Anxiety due to returning to school after the summer holiday was suggested as the cause of the increased sleep problems. This is consistent with the data which shows an upwards trend in the phases following baseline. Numbers of problems per night and amount of time taken to fall asleep are shown in Figure 5.3 and Figure 5.4.

Sleep diary data are in line with actigraphy recording (Lucy wore the monitor for 7 nights both at baseline and post-intervention) except for sleep duration and night wakings (Table 5.4). It appears that Lucy's mother overestimated her child's sleep duration by nearly one hour at post intervention. Also she was not aware of her daughter's night wakings as she did not record any nocturnal wakings for the duration of the study and affirmed that her child never woke in the night. According to the objective measure, from baseline to post-intervention, Lucy's night wakings increased by 62.43 minutes whereas total sleep time and sleep efficiency decreased respectively by 1.27 hours and 9.48%.

Lucy completed the MSDQ at baseline and post-intervention. There was an increase in the total score by 7 points, in Factor one (attributions concerning restlessness agitation) by 6 points and Factor two (attributions concerning mental over-activity) by 1 point whereas there was no change in the scores of the remaining two factors.

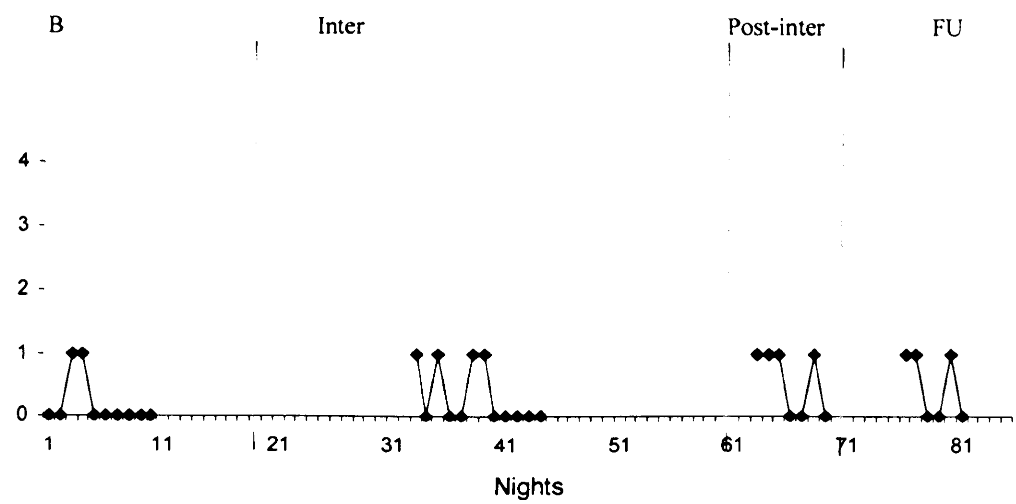

Figure 5.3 Number of problems settling to sleep during baseline (B), intervention (inter), 1 week post-intervention (post-inter) and follow-up (FU) based on diary recording, for Lucy.

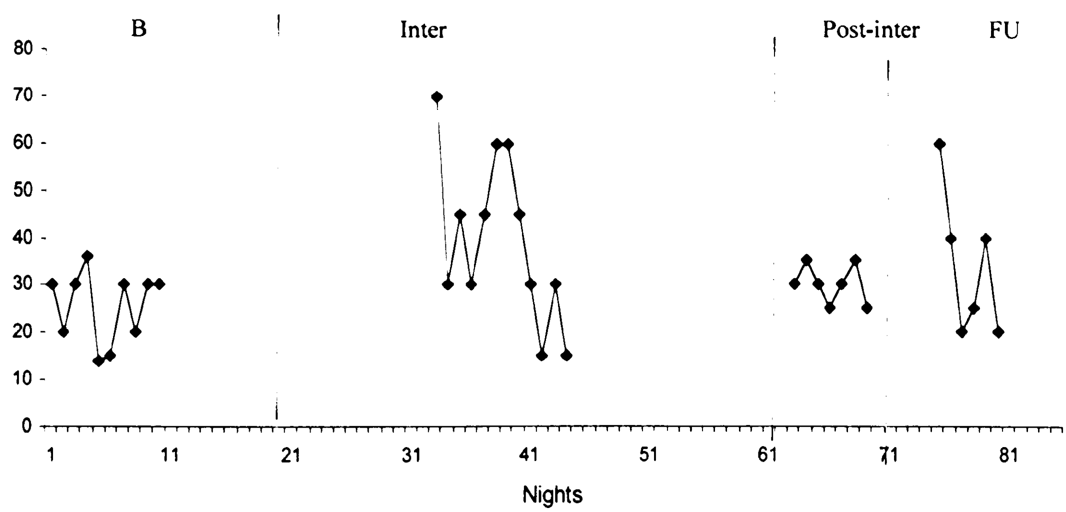

Figure 5.4 The amount of time (minutes) taken to fall asleep during baseline (B), intervention (inter), 1 week post intervention (post-inter) and follow-up (FU) based on diary recording, for Lucy.

Table 5.4 Mean (sd) of sleep variables assessed by diary and actigraphy for Lucy

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.00	19.52	25.50	18.86	10.69	10.61	0	25.14	6.69	6.47
	(0.00)	(8.98)	(7.56)	(12.1)	(0.48)	(24.4)	(0.00)	(18.0)	(0.48)	(26.2)
Post-intervention	19.65	20.04	31.42	27.29	11.26	10.32	0	87.57	7.10	6.54
	(0.43)	(10.9)	(4.75)	(14.8)	(0.51)	(36.0)	(0.00)	(13.8)	(0.13)	(26.7)

Assessment of change in creativity: Lucy

Lucy improved in all the creative measures from baseline to post-intervention period. With respect to the TTCT, the total creativity score increased by 29 points, originality increased by 18 points, fluency by 2 points, and flexibility by 4 points and elaboration by 5 points. All scores were above the average level of performance at post-intervention except elaboration which was far below normative values (see Table 5.5). Improvement was also noted in the story telling task. At baseline, during the pre-test Lucy appeared timid and refused to speak as opposed to the post-intervention phase where she was more talkative. She used 28 words and introduced 3 different real facts. When she was asked to invent a story based on a real card, she was also much more fluent at post intervention where she used 97 more words and introduced 6 more real facts than at baseline. Improvement was also noted with the unreal condition. Lucy used 35 more words and 1 more real fact at post-intervention than at baseline. However, there were no changes in terms of introduction of imaginary elements in both conditions (real and unreal). Lucy was, in fact, not able to introduce any imaginary elements at all with both the real and imaginary card.

Table 5.5 Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Lucy

	NORMATIVE SAMPLE (n=1365) Mean (sd)	Baseline score	Post-intervention score
Originality score	**28 (10)**	20	38
Fluency score	**21 (6)**	22	24
Flexibility score	**16 (5)**	13	17
Elaboration score	**68 (26)**	5	10

Case consideration: Lucy

Introduction of the booklet-delivered behavioural intervention did not result in the elimination of bedtime disturbance for Lucy. Prior to intervention the child exhibited two bedtime disturbances per week which increased by two/three more nights per week in the successive phases. At the same time the amount of time taken to fall asleep increased according to sleep diary data and total sleep time and sleep efficiency deteriorated according to the objective sleep measure. This result might depend on several factors. According to maternal report, anxiety due to the end of the summer holiday and the beginning of the school term had made Lucy's sleep problems worse. However it is possible that the child's sleep difficulties did not improve because her mother did not implement the 6 week behavioural intervention. In fact her commitment was not evident from the way the sleep diary was completed as data were collected only for 10 days during baseline. When telephone consultations were scheduled for the 6 week period intervention, during the first phone call she expressed her doubts about the usefulness of the booklet for the kind of sleep problems with which her child was affected. Thus, it is possible that low levels of motivation might have made Lucy's mother decide not to implement the treatment. However, it is also possible that her mother applied the behavioural treatment but this was not effective for Lucy who might require an alternative form of intervention. There is extensive sleep literature reporting that behavioural interventions produce clinically significant reductions in settling and night waking problems in infants and young children (Mindell et al., 2006) and that many parents have successfully used intervention methods based on information provided by media (e.g book, parenting magazine, TV) (Johnson, 1991) however the effectiveness of such methods delivered is these various formats has not been proved with older typical children. There is, in fact, a lack of knowledge about what factors are associated with school age children's sleeplessness problems and how to treat them. As shown in the MSDQ, Lucy complained of mental over-activity and restless and agitation at bedtime. Thus, it may be possible that the behavioural techniques described in the booklet did not address underlying child cognitions which might be maintaining the sleeplessness in older children.

It is unlikely that the upwards trend noted in Lucy's sleep problems was due to the behavioural intervention. Having only few data points for the baseline leaves doubts to the actual level of the sleep problem prior to the treatment. It is possible that if more baseline data had been collected, the sleep variables during treatment would be shown to be merely a continuation of a trend which was undetected at baseline. Also there are no studies that have concluded behavioural intervention is detrimental in the treatment of sleeplessness in children.

Maternal report of absence of night waking was not corroborated by actigraphy results. Correspondence between these subjective and objective measures is generally good (Sadeh, 1990) although, unsurprisingly, not always without some differences (Acebo et al., 2005). This discrepancy between actigraphy recording and sleep diary data might be due to the fact that Lucy woke at night but went back to sleep on her own without requiring parental intervention or letting their parents know that she was awake.

With regards to the creativity scores of the TTCT, they all improved and were above the average level of performance (except for elaboration) at post-intervention. Similar improvement was found in the story-telling task following the treatment phase. However the child did not introduce imaginary elements, neither at baseline nor at post-intervention.

Case 3: Teddy

This child was 7:8 year old boy who lived with his parents and an older brother. His mother was an editor and his father was an engineer. The boy's mental abilities were at a high level. Teddy, in fact, obtained a GMA score of 133 which is "very high" according to the classification system of the BASII. Spatial ability score (139) was also classified as very high, whereas verbal ability score (117) was in the above average range. The BASII indicate that a difference between verbal and spatial ability greater than 20 points is significant at the .05 level so it may be concluded that Teddy spatial abilities were a strength compared to his verbal skill.

According to CSDI completed by his mother, Teddy had severe problems with settling to sleep, in conjunction with a mild form of night and early morning waking. The child's sleep problems started one year ago and had progressively increased in severity. Of specific concern for his mother was the child's difficulty in falling asleep. He complained of ruminative thoughts at night, finding it hard to relax and feeling physically tense. Parents' current attempts to overcome their child's sleep difficulties included having a long walk before dinner time, placing a music speaker in his room to help settle him to sleep and having a bath before bed. The child was also provided with a small reading-light and taught to remain in his own bed if he woke in the night or in the early morning. Although the child did not disturb any members of the household at night, his mother was very concerned about him because he appeared drowsy and listless during the day. His mother also reported that he often dozed off at school and performed poorly in class.

At the time the study was conducted, Teddy was not receiving any medication.

Assessment of changes in sleep behaviour: Teddy

Figure 5.5 and Figure 5.6 respectively depict the number of bedtime problems and the amount of time taken to fall asleep at three time points (baseline, intervention and post-intervention). Three week baseline sleep diary recording indicated that almost every night Teddy had settling to sleep problems and took a mean of 40.83 minutes to fall asleep (Table 5.6). Figure 5.7, Figure 5.8 and Figure 5.9 also showed that Teddy had a mild form of night waking problems (ranging from 0 to 2 problems per week and lasting for an average of 4.25 minutes) and woke up before 5.00 am two times in a week. There was a SUBSTANTIAL IMPROVEMENT in the child's setting to sleep difficulties, and a MODERATE IMPROVEMENT in night and early waking problems at intervention and post-intervention phase. Three month follow-up diary was not returned so data are not available.

Actigraphy failed to work at baseline, consequently objective sleep measures are only available for 7 nights post-intervention. Sleep diary recording was corroborated by actigraphy data except for night wakings which were underestimated by Teddy's mother. On the basis of the actigraphy recording, Teddy's average total sleep time was 8.1 hours and sleep efficiency 89.19 % (sd3.2). According to his mother, both sleep measures were reduced because the week in which actigraphy recording occurred, Teddy had friends sleeping over and he stayed up for hours.

The child himself reported feeling more relaxed and less agitated at bedtime. This was confirmed by the MSDQ total score which decreased by 2 points. Also scores of Factor one (attributions concerning restlessness/agitation), Factor two (attributions concerning mental overactivity) and Factor four (attributions concerning lack of sleep readiness) were reduced by 2,1,1 points respectively, following intervention as opposed to Factor three (attribution concerning the consequences of insomnia) which increased by 1 point.

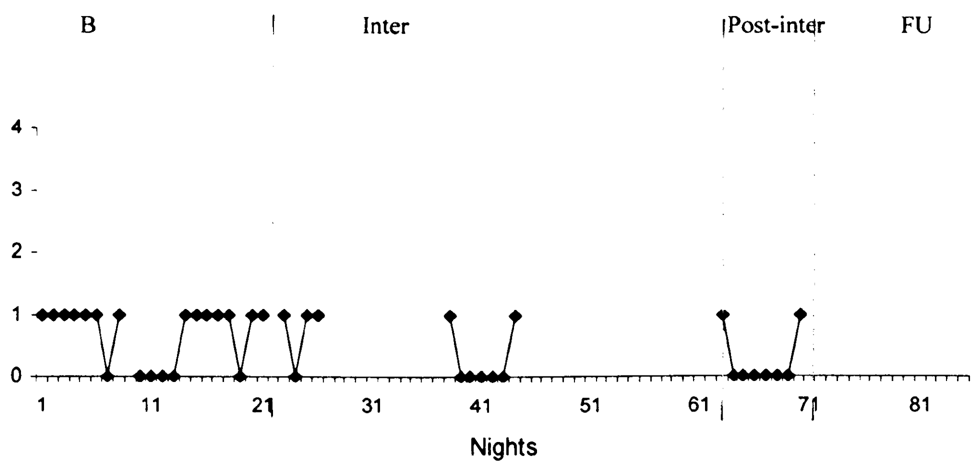

Figure 5.5 Number of problems settling to sleep during baseline (B), intervention (inter), 1 week post-intervention (post-inter) and follow-up (FU) based on diary recording, for Teddy.

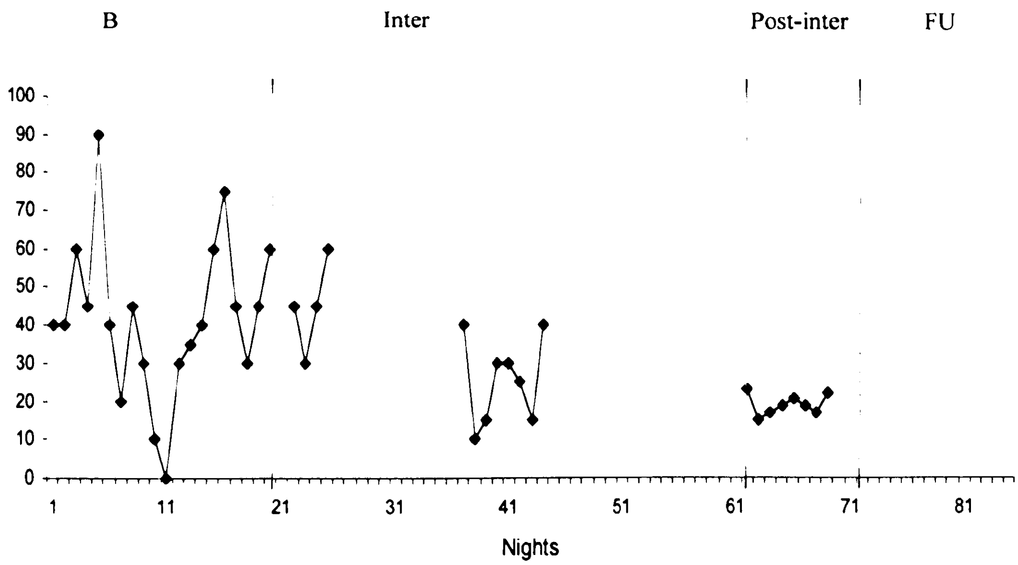

Figure 5.6 The amount of time (minutes) taken to fall asleep during baseline (B), intervention (inter), 1 week post intervention (post-inter) and follow-up (FU) based on diary recording, for Teddy.

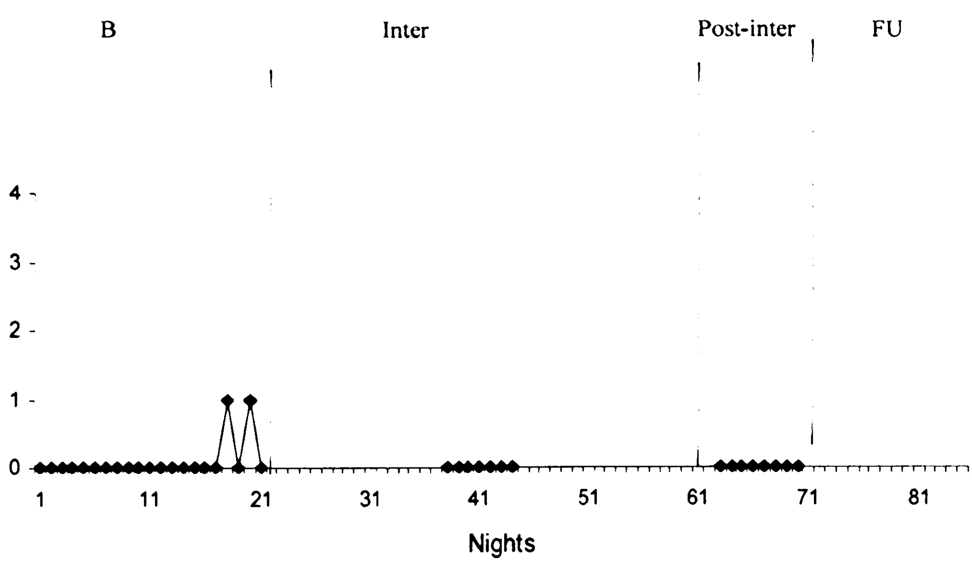

Figure 5.7 Number of night wakings during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Teddy.

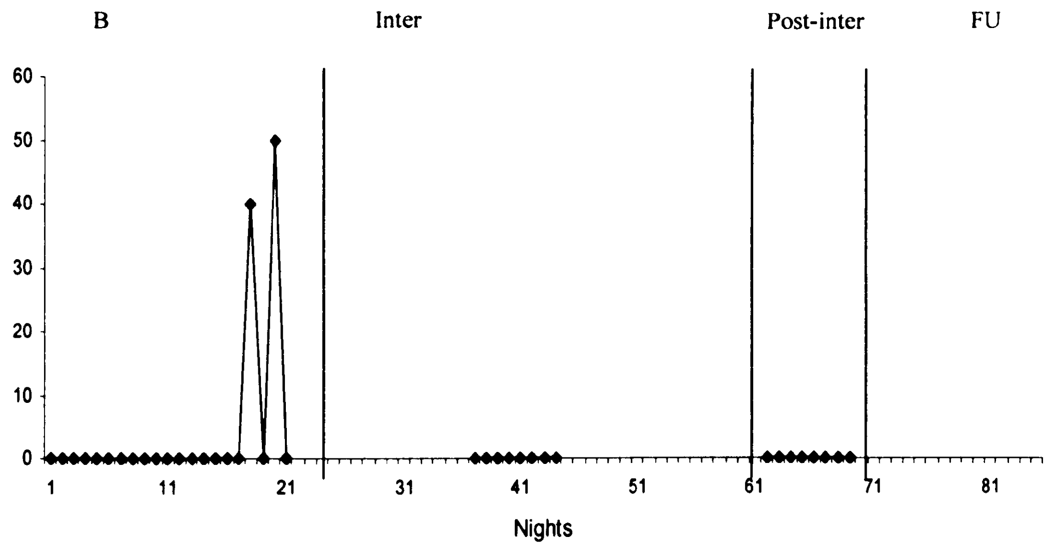

Figure 5.8 Duration (minutes) of night wakings per night during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Teddy.

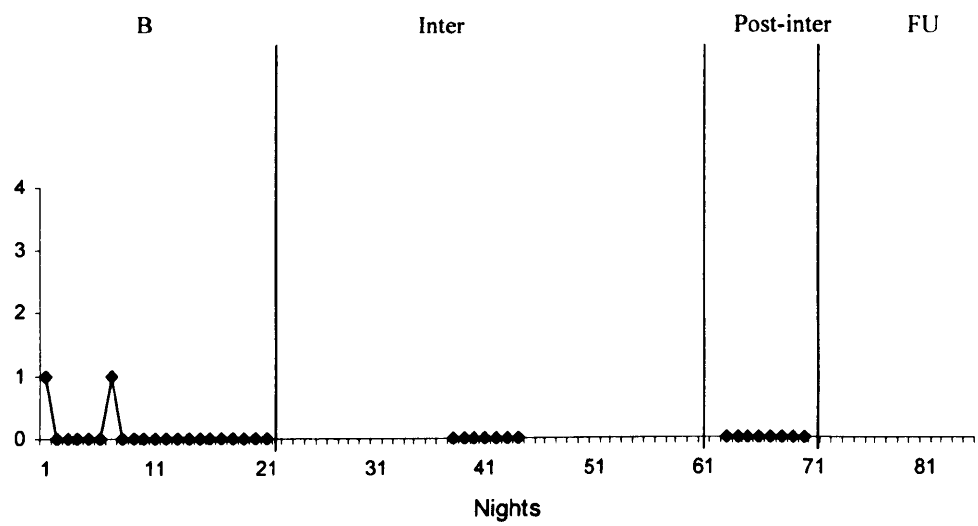

Figure 5.9 Number of early waking mornings per night during baseline (B), intervention (inter) and post-intervention (post-inter) based on diary recording, for Teddy.

Table 5.6 Mean (sd) of sleep variables assessed by diary and actigraphy for Teddy

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.80 (0.50)	-	40.83 (21.43)	-	9.39 (0.72)	-	4.25 (13.10)	-	6.37 (0.66)	-
Post-intervention	21.18 (0.72)	20.55 (16.65)	16.25 (4.55)	22.71 (10.06)	9.11 (0.97)	9.15 (34.35)	0 (0.00)	62.86 (18.40)	6.52 (0.34)	6.26 (24.62)

Assessment of change in creativity: Teddy

Table 5.7 shows that all the creative measures of the TTCT (except for fluency) improved from baseline to post intervention. Total creative score increased by 30 points, originality by 6 points, and flexibility by 5 points and elaboration by 19 points. However Teddy's performance was at the average level only in fluency whereas the remaining three creative scores stayed below normative values. With regards to the story-telling task, at baseline, Teddy used 55 words and introduced 5 real facts during the pre-test. He used 84 words, 3 real facts and 1 imaginary element for the reality based story and 31 words, 3 real facts and 1 imaginary element for the imagination-based story. Change scores are not available as Teddy refused to invent a story at post-intervention. In fact, post-intervention testing occurred on the occasion of a special event; Teddy's older brother had just came home from holiday and Teddy was very distracted and in a rush to go and play with him.

In an attempt to administer the tests in a better condition, an attempt to reschedule the visit for the next week was made but this was not possible because the family was unavailable.

Table 5.7: Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Teddy

	NORMATIVE SAMPLE (n=1365) Mean (sd)	Baseline score	Post-intervention score
Originality score	**28 (10)**	17	23
Fluency score	**21 (6)**	16	16
Flexibility score	**16 (5)**	11	16
Elaboration score	**68 (26)**	4	23

Case consideration: Teddy

Six week behavioural intervention appeared to be successful with a school age typical child whose settling to sleep problems, night and early waking problems were more related to cognitive factors rather than behavioural.

At baseline, Teddy's mother reported that her child was unable to fall asleep at night even though he did not disturb any members of the household. The fact that cognitive hyperarousal might have been the reason why Teddy could not easily fall asleep at bedtime was further confirmed by the MSDQ child version. Teddy himself reported feeling tense, worried and his *"head full of thoughts"* when lying in bed. Parents' past attempt to rectify Teddy's sleep difficulties establishing a sleep routine was not very effective as the diary data revealed Teddy's bedtime and get up time were incompatible with the child's natural sleep timing prior to the intervention. In fact, Teddy mother's comments such as *"he wakes up earlier than I'd like at about 6.15am"* or *"I want him to go to bed at around 8.00pm"* showed that maternal expectations of time and duration of sleep maybe were clashing with Teddy's sleep needs who reported, during visit one, that he was asked to go to bed when he was not feeling sleepy. Two weeks after baseline, during a phone call, it was revealed that his mother had established more age-appropriate sleep and wake times for Teddy moving the bedtime routine later by 15 minutes each night and gradually delaying her child's breakfast time the following morning. According to diary recording, this method made Teddy fall asleep an average of 24.5 minutes earlier and wake up in the morning an average of 15 minutes later. At post-intervention his mother wrote in the diary the following note: *"Teddy's sleep is pretty much back to normal! He sometimes wakes early but he goes to bed well, falls asleep easily, and only ever wakes up enough to disturb someone on the odd occasion he has a bad dream (and that's not often). He is much happier at school, and much less sleepy (though I don't know which is cause and which is effect). I think because he is happier at school, he is perkier, which means he's properly sleeping, which mean that night times are better".*

All the creative measures of the TTCT slightly improved from baseline to post-intervention however remained below the average level of performance (except for fluency). Change scores are not available for the story telling task as Teddy refused to invent a story at post-intervention.

Case 4: Jack

This participant was 4:2 year old boy who lived with his parents and his brother Ben who was 2 years older. His mother was a social worker and his father was an office manager.

The boy's overall performance as measured in the BASII was classified in the average range (100). The scores on the verbal and spatial scales were almost the same, 99 and 101 respectively, and were classified in the average range. The difference of 2 points between the verbal and spatial score did not reach the level required for the 0.05 level of significance.

Jack was reported by his mother as having frequent night waking, co-sleeping and occasional problems settling to bed. These problems which started at age two years were respectively rated as severe and mild. The child was considered a restless sleeper who would often wake up multiple times per night, appear in his mother's bedroom and demand to sleep in bed with her. He was difficult to re-settle after such wakings so the problem was often resolved with the child being allowed to sleep in the parents' bed for the entire night As a result of these recurring night wakings and co-sleeping his mother reported that her own sleep was being disrupted.

One or two times a week he also displayed settling difficulties consisting of refusing to go to bed, moaning and whimpering which led the mother to physically take him into his bed and wait for him to fall asleep. The parents had never looked for professional help.

At the time of study Jack was treated with an emollient for eczema.

Assessment of changes in sleep behaviour: Jack

Visual analysis of graphed baseline data revealed that Jack had frequent night waking and practiced co-sleeping almost every night (from Figure 5.10 to Figure 5.12). At intervention and post-intervention, the rate at which the child fell asleep alone in his own bed and slept through the night was judged to have SUBSTANTIALLY IMPROVED. At post intervention co-sleeping was practiced by Jack during one night only after the child had a bad dream as reported by his mother *"I reassured Jack that it was nothing but a bad dream and that he should get back to sleep but he was scared to death of monsters, and sweating, shaking and wheezing so I allowed him to stay".* According to parental report, reduced co-sleeping was accompanied by improved night waking problems. At post intervention the child learned to stay in his own bed and disturbed their parents only on rare occasions.

NO CHANGE was noted for Jack in the number of settling to sleep problem and amount of time taken to fall asleep, perhaps because of a mild baseline level of problem (Figure 5.13 and Figure 5.14). It was not possible to see if improvements were maintained at three months follow-up as the 1 week diary was not returned.

Jack did not complete the MSDQ as he was not able to understand the questions.

Diary recording of bedtime, wake up time and sleep duration was corroborated by data based on 7 nights of actigraphy recordings but discrepancies were found in terms of sleep latency and night wakings (Table 5.8). Jack's mother tended to underestimate, to a large extent, sleep latency and night wakings. This might be due to the fact that she collected sleep data for four days at the post-intervention period and thus visual analysis is based on these days only. In addition, according to actigraphy recordings, sleep efficiency and total sleep time were poor, both at baseline and post intervention. In fact sleep efficiency and total sleep time for Jack were 68% and 7.3 hours at baseline and 73% and 7.4 hours at post intervention.

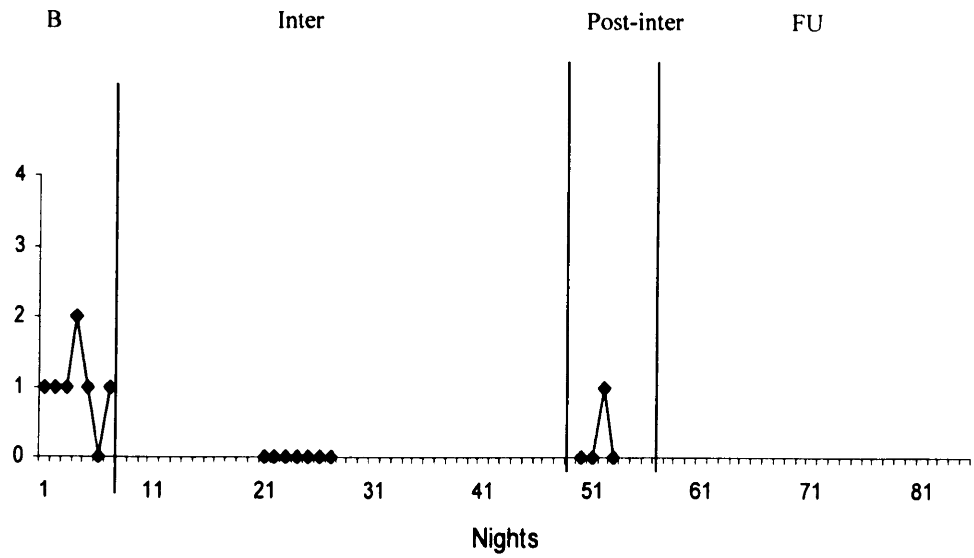

Figure 5.10 Number of night wakings during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Jack.

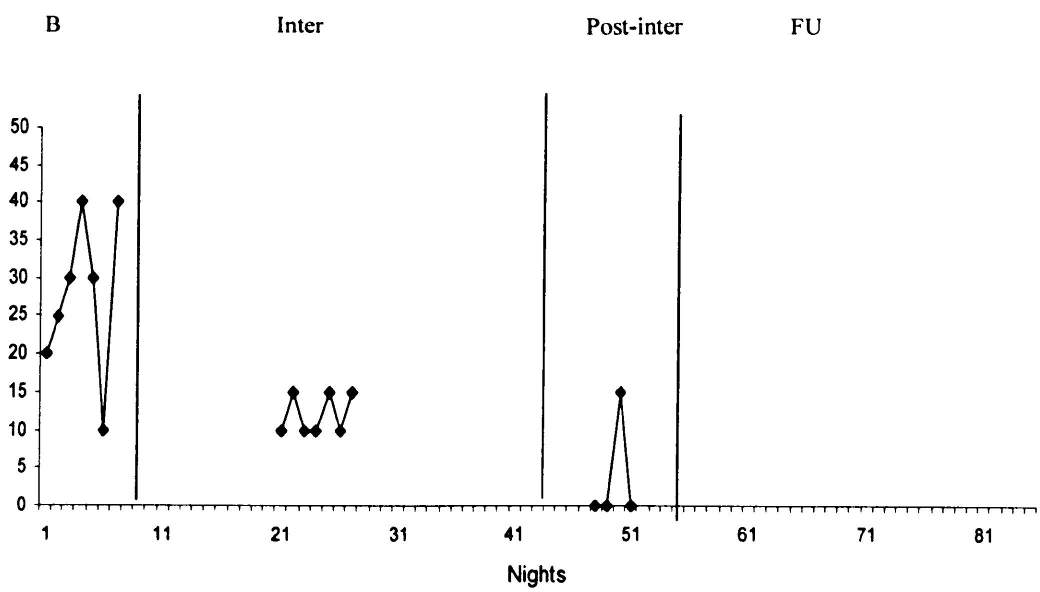

Figure 5.11 Duration (minutes) of night wakings per night during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Jack.

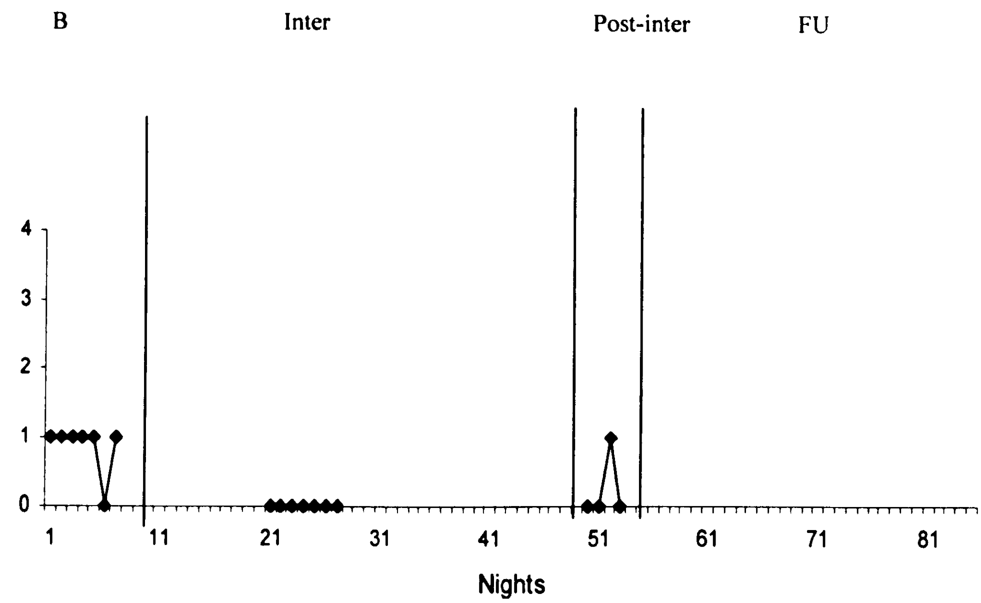

Figure 5.12 Number of times per night that Jack fell asleep in his own bed during baseline (B) intervention (inter) and post-intervention (post-inter) based on diary recording.

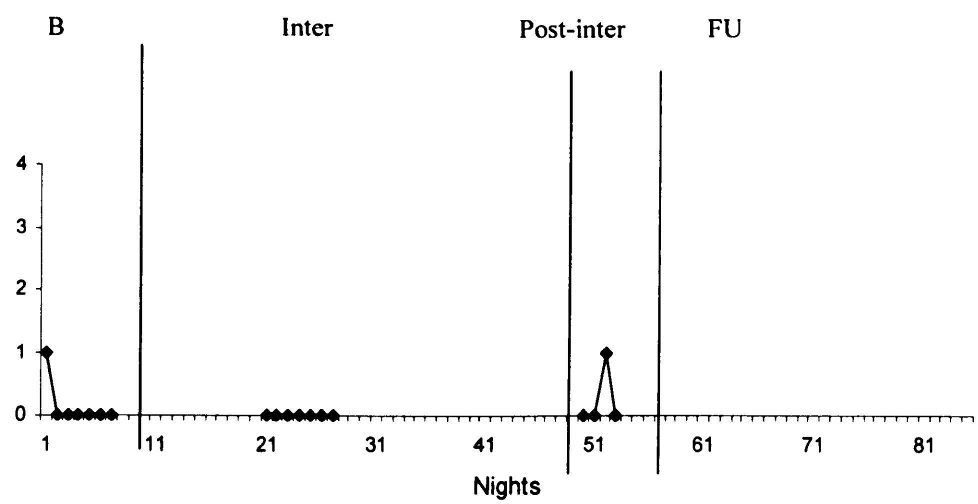

Figure 5.13 Number of problems settling to sleep during baseline (B), intervention (inter), 1 week post-intervention (post-inter) base on diary recording, for Jack.

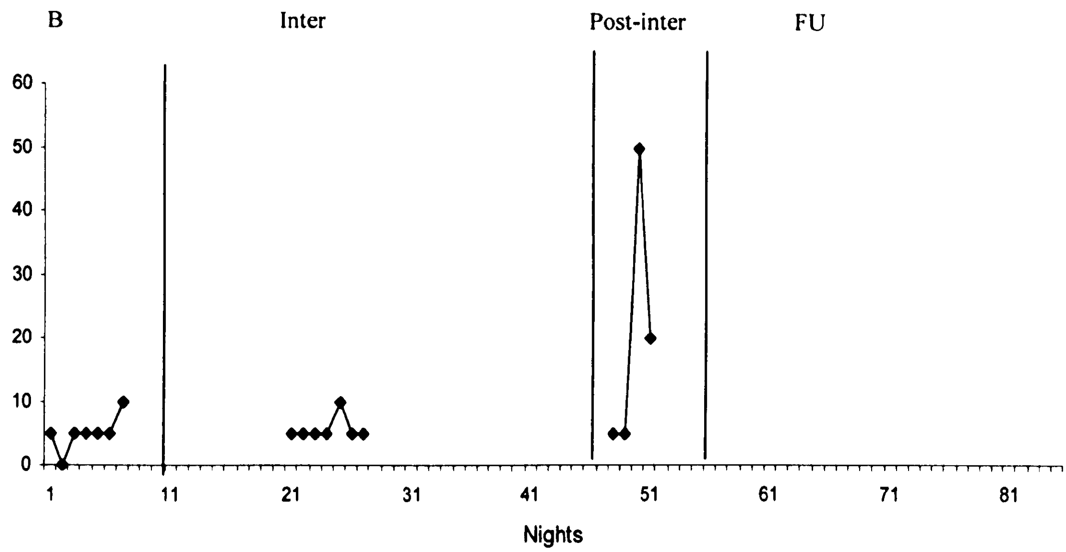

Figure 5.14 The amount of time (minutes) taken to fall asleep during baseline (B) and intervention (inter), 1 week post intervention (post-inter), based on diary recording for Jack.

Table 5.8 Mean (sd) of sleep variables assessed by diary and actigraphy for Jack

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.15 (1.09)	20.22 (75.72)	5.28 (2.36)	18.86 (5.87)	11.21 (1.51)	10.85 (60.95)	27.85 (10.74)	210.71 (51.15)	7.49 (0.70)	7.15 (68.82)
Post-intervention	20.68 (1.60)	20.31 (94.37)	18.75 (18.87)	44.60 (33.46)	10.46 (1.54)	13.92 (48.46)	3.75 (7.50)	159.00 (26.43)	7.25 (0.15)	7.46 (52.65)

<u>Assessment of change in creativity: Jack</u>

Jack showed to have improved in all the creative measures of the TTCT (Table 5.9). Total creativity scores increased by 58 points. There was also an evident enhancement in the originality measure for Jack whose score increased by 33 points. Fluency and flexibility increased by 5 and 9 points respectively at post-intervention. A similar change in elaboration scores was reported with an increment of 11 points reported by

Jack. All creative measures (except for elaboration) were at the average level of performance at post-intervention.

Jack seemed to have improved in the story-telling task following treatment. During the pre-test phase, he used 14 more words and introduced 1 more real fact. When he was asked to invent a story based on a real card, Jack used 9 more words and introduced 1 more real fact but he did not introduce any imaginary elements. A better score was also yielded in the imaginary base story following intervention. Jack used 5 more words, introduced 1 more real fact. Also in this case Jack did not add imaginary elements in his story.

Table 5.9 Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Jack

	NORMATIVE SAMPLE (n=1365) Mean (sd)	Baseline score	Post-intervention score
Originality score	**28 (10)**	14	47
Fluency score	**21 (6)**	19	24
Flexibility score	**16 (5)**	8	17
Elaboration score	**68 (26)**	0	11

Case consideration: Jack

Results demonstrated the effectiveness of behavioural treatment for treating settling, co-sleeping and night waking difficulties in early age typical children.

According to maternal report, Jack's sleep problems improved at post-intervention. During the last home visit, she stated that settling, co-sleeping and night waking problems were almost eliminated and that when, on the rare occasions night wakings occurred, Jack returned to bed with little difficulty.

However, according to objective sleep measure Jack was sleep deprived since he was sleeping around 7 hours at baseline and at post-intervention. This result is not surprising since sleep disturbances are one of the most frequently reported behaviour problems affecting children from the general population with estimated rates of about

30% (Owens, Spirito, McGuinn et al., 2000). Maternal reports of night wakings were also not corroborated by actigraphy measure. On the basis of diary recording, durations of night waking were far shorter than the objective amount of time the child spent awake at night. This might be the product of a change in the maternal perception of the problem rather than a real change in the child's sleep. It is possible that the parents, who no longer shared their bed with the child, were simply not as aware of the child's nocturnal wakings. It is, in fact, very likely that if a young child wakes up during the night and the parents are present, he or she may signal discomfort and call out for attention. This result is also congruent with a study with adolescents with autism where objective sleep recording were incongruent with subjective sleep measures and failed to demonstrate changes in objective sleep which were related to treatment (Oyane & Bjorvatn, 2005).

At post intervention, only on one occasion did Jack go to sleep with his parents because he had a bad dream. Night-time fears follow a developmental path linked to cognitive development and are viewed as a normal development phase of the young child (King, Ollendick, & Tonge, 1997). In addition, the development of self recognition, during the first years of life, leads to an awareness of being without the comforting maternal figure during the night and makes the child actively seek physical contact. Thus, Jack's mother might have found it difficult to neglect her child's need for comfort after waking.

These results also confirms that it is a common false perception that night waking and bed sharing occur mainly in babies and toddlers and that their prevalence declines with age. A longitudinal study found that co-sleeping and night wakings after infancy reached a peak prevalence of 38% at 4 years old (Oskar & Heidi, 2005). Thus, different functional processes at different developmental stages might lead to nocturnal wakings and bed sharing. For instance, specific developmental changes in separation-attachment processes, cognitive capabilities to develop self-recognition and night-time fear may contribute to the persistence of night waking and co-sleeping during early childhood.

Following intervention all the creative measures of the TTCT increased. Improvement was also noted in the number of words and real facts introduced in the story telling task, however Jack did not add any imaginary elements in his accounts.

Case 5: Ben

This participant was 6:2 year old and was the older brother of Jack.

He obtained a GMA score of 105 which was in the average range of performance according to the BASII classification system. The child's verbal and spatial scores were classified above (114) and at average (94) levels of performance respectively. The observed difference of 20 points between these two scores was significant at the .05 level.

Like his younger brother, he also displayed severe night waking and co-sleeping problems as well as a mild form of settling to sleep problems which appeared when he was two years old. When the time to go to bed arrived, the child was reluctant and tried to delay bedtime. He needed to be told to put on his pyjamas and get ready for sleep. The mother stated that he watched cartoons on DVD in the lounge till late, ignoring her requests. His behaviour was a bad example for his younger brother who also refused to go to bed making the parents feel exhausted. Night wakings occurred in both children who shared the same bedroom and consequently disturbed each other. When the younger brother went to sleep in his parents' bed, Jack did not like to be left alone and also demanded to share the bed with them. His mother, however, reported that nocturnal wakings and co-sleeping were slightly more manageable in Ben. Like with the younger son, the parents had never considered asking for professional help with this child's sleep problem.

Ben also had eczema and was treated with a cream moisturiser.

<u>Assessment of changes in sleep behaviour: Ben</u>

Baseline data indicated that Ben had frequent night waking and practiced co-sleeping almost every night (Figure 5.15 to Figure 5.17). Ben's mother reported that he was disturbed in the middle of the night by his younger brother with whom he shared the room. A SUBSTANTIAL IMPROVEMENT in the number of nights Ben fell asleep alone in his own bed and slept through the night was observed at intervention and post-intervention phase. Ben learned to stay in his own bed and co-sleeping was no longer a problem. He was taught not to follow his younger brother in his parents' bedroom and to ignore Jack if he tried to chat with him at night.

A MODERATE IMPROVEMENT was noted in the number of settling to sleep problems and the amount of time taken to fall asleep for Ben, who went to bed quicker without the need to be asked several times to get ready for sleep (Figure 5.18 and Figure 5.19). As for Jack it was not possible to see if improvements were kept at three months follow-up as the 1 week diaries were not returned.

Ben understood and completed the MSDQ. Total MSDQ and Factor two scores (attribution concerning mental overactivity) reduced by 1 and 2 points respectively whereas the remaining 3 factors stayed the same.

Data based on 7 nights of actigraphy recordings confirmed diary data for bedtime, wake up time and sleep duration, but differences were found in sleep latency and night wakings between diary and actigraphy data (Table 5.10). As with Jack, Ben's mother tended to underrate sleep latency and night wakings. Also, according to actigraphy recordings, sleep efficiency and total sleep time were poor, both at baseline and post-intervention. The sleep efficiency and total sleep time for Ben were 57% and 8.0 hours at baseline and 67% and 6.6 hours at post-intervention. However, this reduction in the amount of sleep found in Ben at post-intervention might be due to the fact that the child wore the monitor during an atypical week as his mother wrote on the diary: *"last week was a bit unusual being end of the term and being invited to barbeques, cricket matches etc. so an unusual series of late nights, plus Ben was invited to a sleepover".*

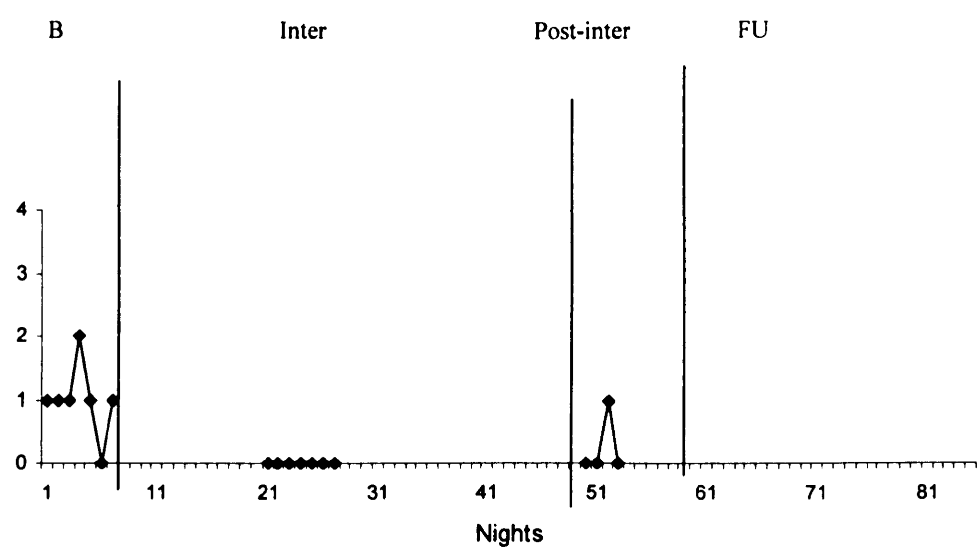

Figure 5.10 Number of night wakings during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Jack.

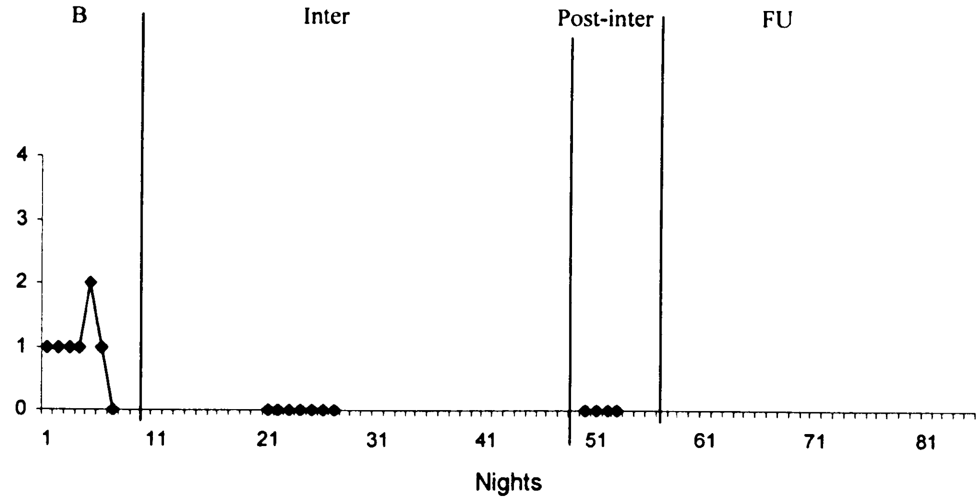

Figure 5.15 Number of night wakings during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Ben.

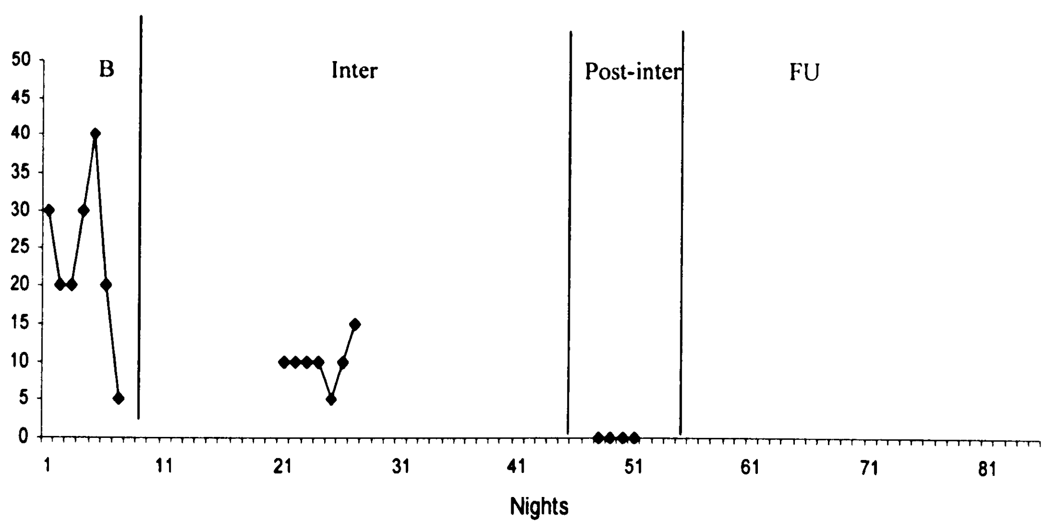

Figure 5.16 Duration of night wakings per night during baseline (B), intervention (inter) and 1 week post-intervention (post-inter), based on diary recording, for Ben.

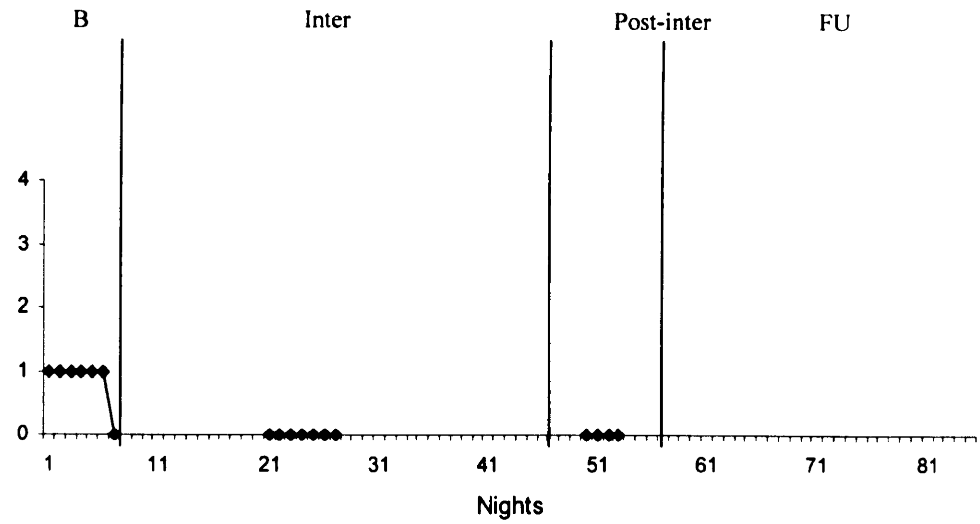

Figure 5.17 Number of times Ben fell asleep in his own bed during baseline (B), Intervention (inter) and post-intervention (post-inter).

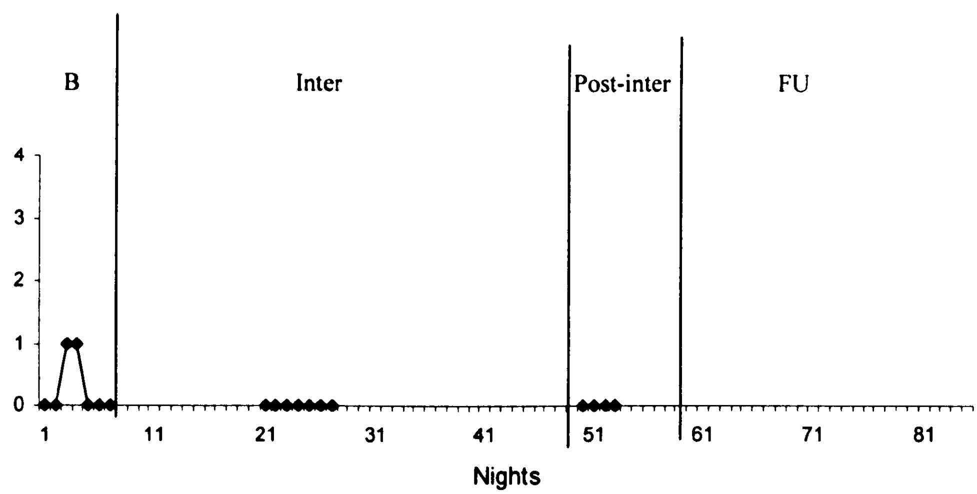

Figure: 5.18 Number of problems settling to sleep during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Ben.

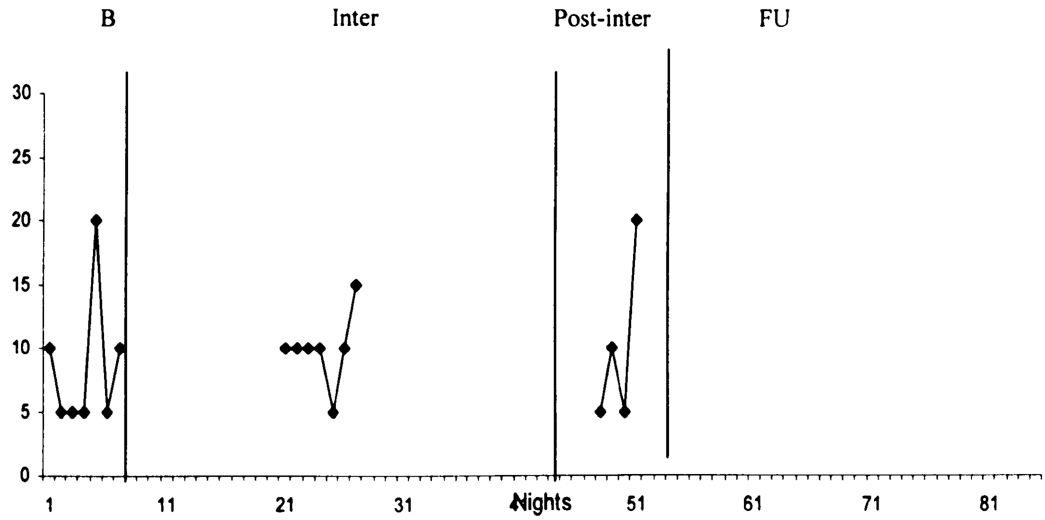

Figure: 5.19 The amount of time (minutes) taken to fall asleep during baseline (B), intervention (inter) and 1 week post-inter (post-inter) based on diary recording, for Ben.

Table 5.10 Mean (sd) of sleep variables assessed by diary and actigraphy for Ben

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.25 (1.16)	20.08 (76.36)	7.50 (6.12)	42.57 (31.51)	10.67 (1.50)	10.28 (40.31)	26.66 (8.16)	135.57 (35.85)	7.02 (0.53)	7.08 (67.72)
Post-intervention	20.68 (1.60)	20.44 (87.70)	10.00 (7.07)	33.00 (15.67)	10.46 (1.45)	9.86 (69.30)	0 (0.00)	194.80 (59.43)	7.25 (0.15)	7.08 (20.90)

Assessment of change in creativity: Ben

Ben showed to have improved in all the creative measures of the TTCT (Table 5.11). Total creativity scores increased by 24 points but there was not an evident enhancement in the originality measure for Ben whose score increased only by 3 points. Fluency and Flexibility increased respectively by 5 and 4 points at post-intervention. Despite Ben showing a greater improvement in the elaboration score reporting an increment of 12 points at post-intervention, the elaboration score remained below the average level. On the contrary, all the creative measures (originality, fluency and flexibility) were at the normal level of performance at post-intervention.

Ben appeared to have improved in the story telling task following intervention. During the pre-test phase, Ben used 29 more words and introduced 2 more real facts. When he was presented with a real card to invent a story, Ben used 7 more words and introduced 1 more real fact. In contrast with Jack, Ben introduced an imaginary element as a part of his story (*"the flying birds turned into bones"*) at post-intervention. A better score was also yielded by Ben in the imaginary base story following intervention. In fact he used 14 more words and introduced 2 more real facts and 2 imaginary elements *("...his hands disappeared and then reappeared")* in his account.

Table 5.11 Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Ben

	NORMATIVE SAMPLE (n=1365) Mean (sd)	**Baseline score**	**Post-intervention score**
Originality score	**28 (10)**	35	38
Fluency score	**21 (6)**	16	21
Flexibility score	**16 (5)**	14	18
Elaboration score	**68 (26)**	17	29

<u>Case consideration: Ben</u>

Results show the efficacy of behavioural intervention for treating settling, co-sleeping and night waking difficulties in typical children over the age of 6.

At post-intervention phase there was a significant improvement in Ben's sleep difficulties according to maternal report. Not only had Ben learned to sleep trough the night but also to stop imitating his younger brother's bad sleep habits such as going to sleep in their parents bedroom in the middle of the night. Ben himself, who completed the sleep questionnaire version for children, reported sleeping better. As for Jack, these results were not supported by improvements in actigraphy measures of sleep following intervention. Ben was seriously sleep deprived at baseline and post intervention phase. In fact he slept an average of 8 hours at baseline and 6 hours at post-intervention. As previously mentioned, this reduction in the amount of sleep found in Ben at post treatment might be due to the fact that the child wore the monitor during an atypical week.

Maternal reports of night wakings were also not supported by actigraphy measurements. Ben's mother tended to underestimate the duration of night waking compared to the objective amount of time the child spent awake at night. This might be the product of technical or situational artefacts in the actigraphy recording. Artefacts may result when persons place their wrists on or under their head or stomach during sleep (breathing artefact) or possibly from having an unusual mattress or bed (Sadeh et al., 1995). However this does not seem to be the case because differences between sleep diary data and actigraphy recording of night wakings were also found in the case of Ben's younger brother. Also the fact that data based on 7 nights of actigraphy recordings corroborated diary data of bedtime, wake up time and sleep duration confirmed that discrepancy were due to maternal misperception of Ben's night waking problems rather than sleep recording artefacts.

Also the mother's failure to report night waking for all the post-intervention and follow up phase might account for the discrepancy between diary and actigraphy recording. The fact that Ben's mother completed the diary only for 4 days at post-intervention and gradually skipped items she should have completed on the daily log is likely to have caused the discrepancy with the actigraphic measures of Ben's night wakings.

The parental attrition factor described by Sadeh's (1994) study plays an important role when comparing actigraphy with daily logs and when parents are requested to document their children's sleep for prolonged periods. Sadeh (1994) studied the effect of behavioural guidance for parents of 50 sleep-disturbed infants and young children by using actigraphy and parents' subjective reports and showed that discrepancy between these measures increased during the study due to increased parental incapacity to document night wakings and the children's acquired ability to stay awake and to resume sleep without signalling. Subjects studied in laboratory for a few nights under close supervision may complete daily logs much more reliably than those who are studied at home for prolonged period of time (Sadeh et al., 1995).

Because this mother had two children taking part in this research study and given that she had to keep a daily log for each child over a long period of time and she could have lost motivation and made the diary's completion inaccurate, Ben's mother was asked to record her children's sleep only during certain period of the intervention phase. Despite these precautions, compliance with the sleep diary remained a big issue. This result also gives some insight into the problem of bedroom sharing in childhood. Despite siblings sharing a bedroom having some positive aspects (e.g children bond given their close proximity; children are given the tools they will need when sharing a room later at university, or living with roommates in private accommodation; children learn to compromise and share) this might become a problem when difficult bedtime behaviour of one can clearly impact the other, making things difficult for parents who do not have choice (e.g. lack of space in the house) but have to have siblings share a room. The fact that Ben learned to ignore the younger brother who woke up often at night and who called for his attention suggested that children as young as 6 years old can be taught basic behavioural strategies to cope with the problem of sharing a room with a sibling who has sleep difficulties.

In addition it was reported that Ben encouraged his younger brother not to disturb their parents' sleep when night waking occurred. Thus, behavioural principles might not only been used by parents to treat children sleeplessness problems but also by children themselves when they share a bedroom with a sibling with problematic sleep behaviour.

Following intervention all the creative measures of the TTCT increased. Improvement was also noted in the number of words, real fact and imaginary elements introduced in the story-telling task by Ben.

5.3.3 Summary of sleep and creativity results for TC group

The current study supports recent findings (Eckerberg, 2002) that booklet-delivered behavioural treatment is both quick and appropriate for the enhancement of sleeping difficulties in typical children.

For the five target sleep variables analysed (number of problems settling to bed, duration of settling to bed, number of night wakings, duration of night wakings and frequency of early morning wakings) visual analysis of graphs showed that four children who experienced these problems at baseline moderately or substantially improved at post intervention phase (except for Jack who suffered multiple sleep problems and showed no change in settling to sleep). In contrast, one child (Lucy) who only reported mild settling to sleep problems showed a moderate deterioration at post-intervention. When improvement was found, this was not maintained at three months follow-up (follow-up data were available only for Esme and Lucy) and was not associated with any change in the children's objective sleep patterns. The principal difference between actigraphy and sleep logs was the fact that the mother reported shorter night waking duration at baseline and post-intervention phase. Because parents are most aware of their child's sleep problems if the child signals, discrepancies between parents' logs and actigraphy are expected (Sadeh et al., 1995). However sleep diary data were corroborated by children's sleep questionnaire results. Only three children were able to complete the MSDQ. Two TC (Teddy and Ben) reported feeling slightly more relaxed at bedtime following intervention whereas one child (Lucy), consistent with the results of reduced sleep according to sleep diary and actigraphy data, reported increased mental activity at post-intervention.

Typical children's creativity measures in general improved following intervention. All TC, in fact, reported higher creativity score in the TTCT and showed a general tendency to perform above normal level of performance at post intervention (except for elaboration score which remained very low for all children) .

The same pattern of results was found in the story-telling task. Children used a greater number of words and added more real facts following intervention. On the contrary no changes were found in terms of imaginary elements. Children introduced none or few imaginary elements in their story.

It is important to state that enhanced creativity seen in TC cannot be simply attributed to improvement in sleep behaviour. In fact higher creativity scores were also found in one child (Lucy) who showed moderate sleep deterioration following treatment. Creativity results are discussed in more details in section 5.4.

5.3.4 Children in the HFA group

Case 6: Dominic

This child was a 4:10 year old boy with a diagnosis of autism which was further confirmed by the ASQ completed by his mother. He was assigned a score of 22. Since the first visit Dominic showed a strong oppositional behaviour which made any form of interaction with him extremely difficult for the duration of the study. He appeared to have a strong bond with his mother, who was the only person whose words he would listen to. He was living at home with her. She was a single parent and unemployed. During the day Dominic attended a nursery school where he received special support.

Based on the BASII, Dominic obtained a GMA score above the average level of performance (107). The child's scores on the Verbal Ability and Spatial Ability scale were very different, 84 and 127 respectively. Whereas his verbal ability score was in the below average range, his spatial ability score was classified as very high. The BASII indicates that a difference of 18 points between verbal and spatial scores is significant at the .05 level. The difference of 43 standard score points between these scores is therefore significant and it might be concluded that the child's spatial ability was a strength compared with verbal ability. The fact that Dominic excelled in the spatial task is not surprising. A common theme from the autistic literature is the heightened abilities in visual spatial functioning in individuals with autism. It is in fact long established that they do extremely well on embedded figure tasks and block design tests relative to non-autistic control groups (Mitchell & Ropart, 2004) and that they might have impaired verbal abilities (Wing & Gould, 1979).

His mother reported that he presented problems in settling to sleep, night waking and co-sleeping which were lifelong and rated as severe. Of specific concern was the child's sleep anxiety that often led to bouts of crying and screaming when bedtime approached. The mother reported that he would continually come out of his room, keep asking if she was going to sleep and calling her non-stop. He would sometimes fall asleep in places other than his own bedroom such as in front of his mother's bedroom door. The only way she could reduce his anxiety was to sleep with him. Often this led to the child being put into his mother's bed and transferred into his own bed when asleep. Consequently, when night wakings occurred, the child called out or appeared in his mother's room and demanded to sleep with her.

The child's mother had never sought professional help for his sleep problems but used the Picture Exchange Communication System (PECS) when he was particularly anxious and showed oppositional and disruptive behaviour. PECS uses pictures instead of words to help children communicate. Dominic was given a set of pictures representing human feeling. When the child wanted to express a feeling (such as anger, happiness, sadness), he was asked to give the appropriate picture to his mother.

With respect to medication being taken, Dominic was on a stimulant laxative and calcium supplements.

Assessment of changes in sleep behaviour: Dominic

Baseline measures indicated that almost every night, Dominic was being settled to sleep in his mother's bed and then relocated into his own bed when asleep (Figure 5.20). Then he woke up every night between one and two times and disturbed his mother if not permitted to return to sleep with her (Figure 5.22 and Figure 5.23). Visual analysis of graphs show that there was a SUBSTANTIAL IMPROVEMENT in the number of times Dominic went to sleep in his mother's bed and in the frequency and duration of night wakings at intervention and post-intervention phase. However NO CHANGE were observed in the amount of time Dominic took to fall asleep. Phone contact one week after the beginning of the intervention found that co-sleeping had reduced considerably within the first few nights. After implementing the intervention for approximately two weeks, number of night wakings decreased and the boy experienced between zero and two night wakings per week for the remainder of the intervention and post-intervention phase.

As depicted in Table 5.12, average bedtime, wakeup time and sleep duration did not change from baseline to post-intervention. Prior to treatment, sleep latency was not considered a problem because the child was allowed to sleep with his mother (Figure 5.21). During baseline sleep latency averaged 40.80 minutes (sd 26.1) and during intervention Dominic fell asleep with an average of 27.20 minutes (sd15.6). During intervention, on the second night of diary recording Dominic took over an hour to fall asleep because he was not feeling well.

Actigraphy data are not available because the monitor did not retain the data at baseline and Dominic's mother did not attempt to put it on her child wrist at post intervention.

Data of the MSDQ were also not collected as Dominic's mother believed the questionnaire was too difficult for a child of his age and with autism.

Additionally, three months follow-up diary was never returned despite it being sent two times in a two week period and one phone call being made to encourage Dominic's mother to find the time to complete it.

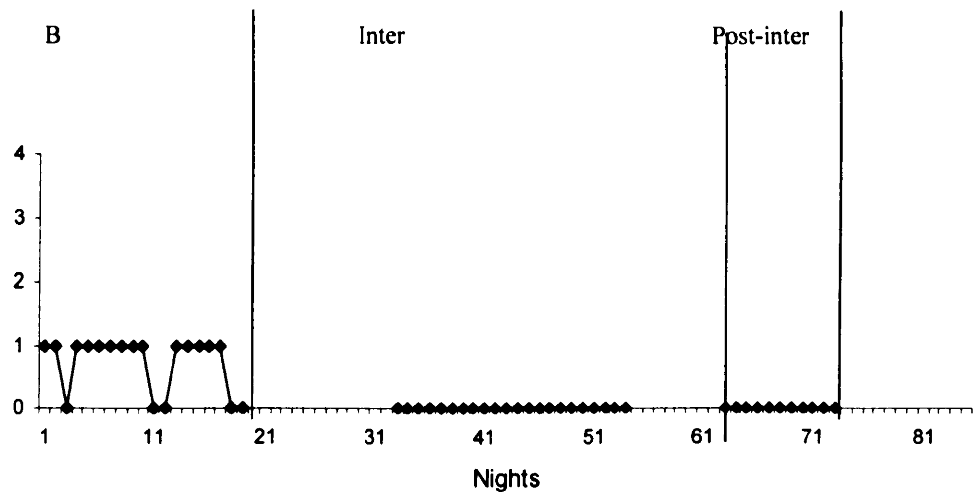

Figure 5.20 Number of times per night that Dominic fell asleep in his own bed during baseline (B) intervention (inter) and post-intervention (post-inter) based on diary recording.

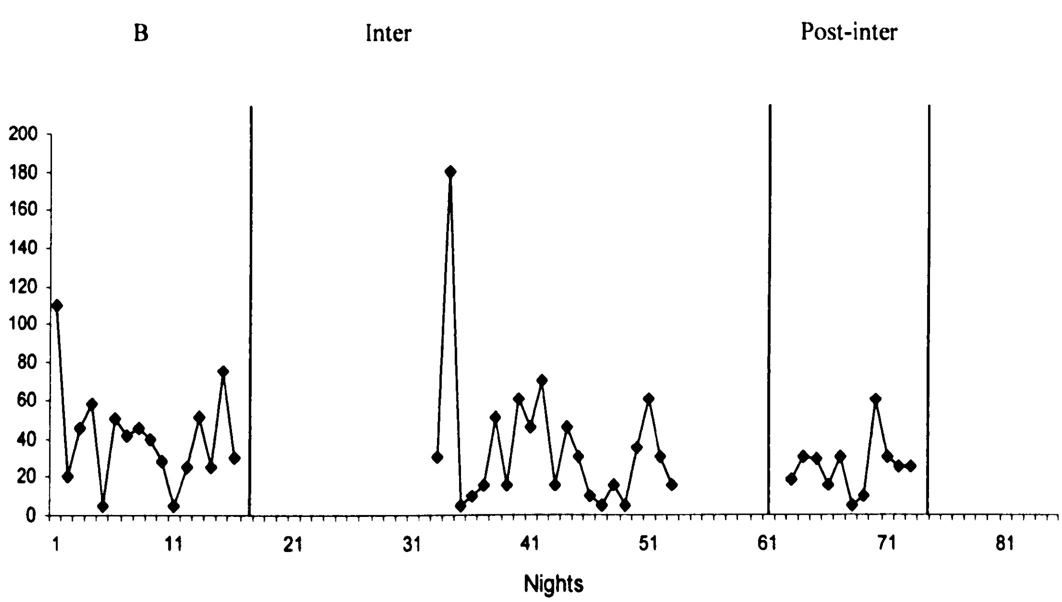

Figure 5.21 The amount of time (minutes) taken to fall asleep during baseline (B) and intervention (inter), 1 week post intervention (post-inter) based on diary recording, for Dominic.

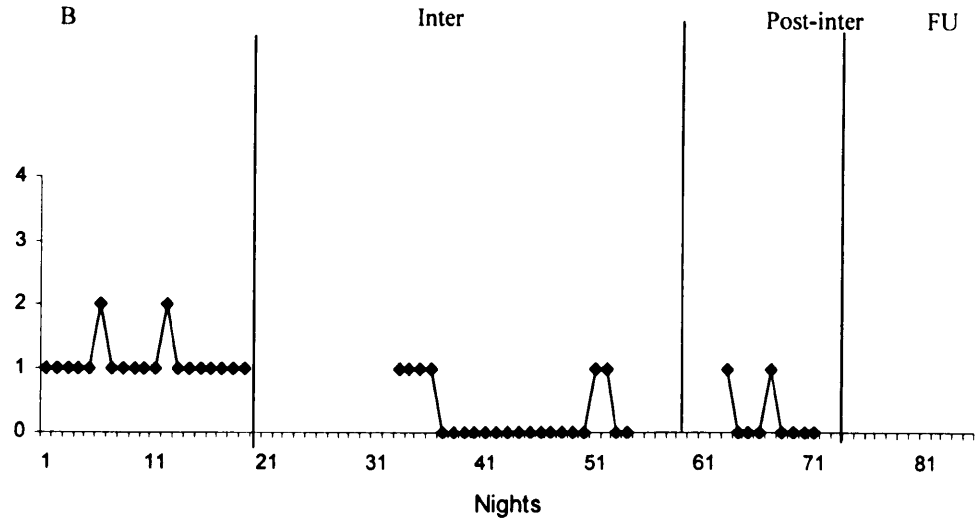

Figure 5.22 Number of night wakings during baseline (B), intervention (Inter) and 1 week post-intervention (Post-inter) base on diary recording, for Dominic.

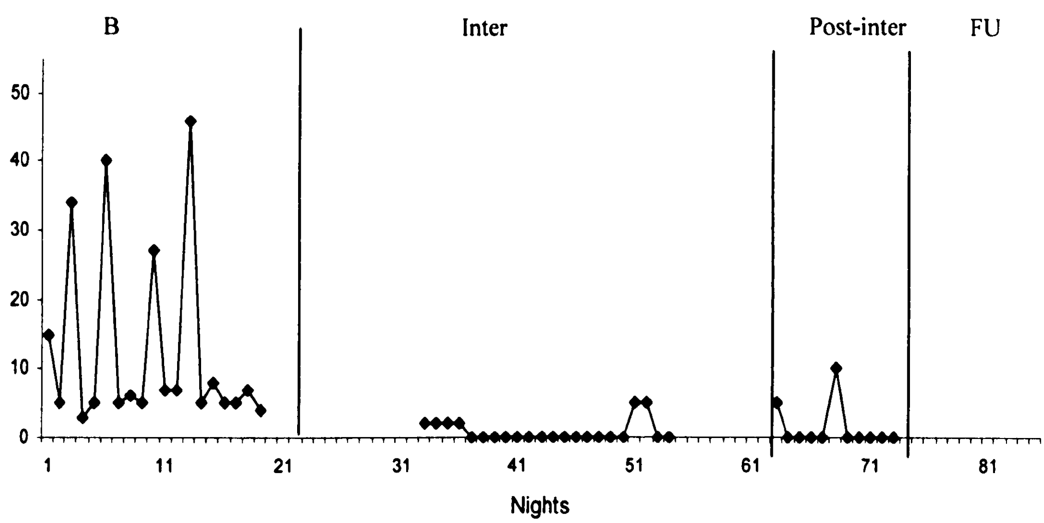

Figure 5.23 Duration (minutes) of night wakings per night during baseline (B), intervention (inter) and 1 week post-intervention (post-inter) based on diary recording, for Dominic.

Table 5.12 Mean (sd) of sleep diary data for Dominic (there are no actigraphy data)

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	19.50 (0.63)	-	40.80 (26.1)	-	10.80 (18.2)	-	12.50 (9.4)	-	6.64 (0.65)	-
Post-intervention	20.33 (0.48)	-	27.20 (15.6)	-	10.10 (0.90)	-	0.90 (1.6)	-	6.66 (0.42)	-

Assessment of change in creativity: Dominic

Creativity data are available for baseline period only as Dominic refused to collaborate in any of the activity at post-intervention (Table 5.13) His behaviour was extremely oppositional. Despite being encouraged by his mother and being offered rewards for the completion of the tasks he did not respond positively. He cried, screamed and moved to a different room. At baseline he agreed to collaborate for a limited time and after numerous attempts. He performed below the average level of performance in the TTCT reporting a score of 64 in total creativity, a score of 28 in originality, a score of 16 in fluency, a score of 10 in flexibility and a score of 18 in elaboration. He also performed poorly in the story-telling task. He did not respond to any of the questions asked at the pre-test phase. When he was presented with the real card, he was not able to create a story and repeated the statement "*I don't know*" 10 times. The imaginary card did not inspire him. He produced only 14 words and did not introduce any new concepts nor imaginary elements. However Dominic was able to recognise all the unreal cards.

Table 5.13 Creativity scores based on TTCT at baseline (there are not post-intervention data for this child) compared to normative means(standarddeviation) for Dominic

	NORMATIVE SAMPLE (n=1365) Mean (sd)	Baseline score	Post-intervention score
Originality score	**28 (10)**	20	-
Fluency score	**21 (6)**	16	-
Flexibility score	**16 (5)**	10	-
Elaboration score	**68 (26)**	18	-

Case consideration: Dominic

These results confirm that behavioural intervention can successfully be used in the treatment of sleep problems in young children with autism. At an initial assessment, Dominic's mother appeared very concerned about her child's sleep behaviour as she made the following note on the diary: "*Dom is very anxious and distracted and finds it hard to go to sleep. He continuously cries and asks if mum is going to bed*". Settling to sleep difficulties were also accompanied by severe night waking problems. Dominic came out of his room every night and required insistently to sleep with his mother and caused her stress. She wrote on the diary the following comment: "*...in his own bed he won't resettle so he comes into mum's bed every night as that is the only way I get sleep*".

According to his mother, settling difficulties, night waking and co-sleeping were no longer a problem by the end of the study. However data were not collected immediately after baseline so it is not possible to delineate the changes in terms of sleep that occurred during the first 10 days of the treatment period. She reported that a reward was used to encourage him to stay in his own bedroom throughout the night as she wrote: "*Routine is exactly the same. No changes to anything except ice-lolly reward in the morning if Dominic stays in his bed all night*". The last day of the treatment period, she wrote the following note on the diary: "*This morning he did not want ice-lolly even though he stayed in his bed. I think now he will just stay in his bed* *and I don't need to use the reward anymore"*. Despite the amount of time taken to fall asleep did not change at the two time points according to visual analysis of graph, Dominic learned to soothe himself to sleep. The fact that Dominic responded so well to the treatment might be due to the determination of his mother to stop his bad sleep behaviour in conjunction with rewarding his positive conduct. She was exhausted by his long lasting sleep problems and was resolute to stop them. Thus it might be possible that Dominic was more responsive to his mother who was now adopting a more authoritative parenting style. This result is in line with a number of studies with atypically developing children which have stressed the importance of reward systems to promote independent sleep and encourage positive night routines (Wood and Sacks, 2004). Whether or not maternal report was congruent with objective sleep measures remains unknown since there was a technical failure with the first recording. This was an unusual occurrence and actigraphy has been used successfully in previous research with children with autism (Allik et al., 2006; Hering et al., 1999; Wiggs & Stores, 2004). It is worth noting the possibility that Dominic did not wear the monitor at post-intervention because his mother did not attempt to put it on his wrist since it failed to work at baseline.

The MSDQ was also not completed. Dominic's mother wrote the following note on the questionnaire: *"The concept of this questionnaire is too complex for Dominic (5yrs)... usually, sometimes, rarely. It doesn't seem for children (autistic or not) of his age. I asked him the questions but he couldn't answer orally either and lost interest totally. It's too hard for him."*

Given the potential importance of children's beliefs about their own sleep, the MSDQ was developed for use with school age typical children and it was adopted in the current study in an exploratory manner to investigate its use with early age typical children and children with autism.

Change scores of the creativity measures are not available as Dominic refused to complete the tasks at post-intervention. At baseline, he performed below the average level of performance in the TTCT and he did not introduce any imaginary elements in both the real and unreal story.

Case 7: Andrew

This participant was a 10:3 year old boy with autism who lived with his parents and his older sister. His mother worked as a primary school teacher and his father as a policeman.

Andrew scored 30 in the ASQ completed by his mother. He showed behaviour that might cause others to feel uncomfortable. In fact he made very inappropriate comments to which his mother tended to diminish the improperness by laughing as though they were funny jokes.

The boy obtained a GMA score (97) and a spatial ability score (106) classified in the average range according to the classification system of the BASII. Verbal ability was categorised as below average (89) but it was in the higher end of that score range. As indicated in the BASII a difference of 17 points between verbal and spatial score is significant at the .05 level. For Andrew, spatial ability was therefore a strength compared to verbal ability.

Andrew's mother reported that her child complained of having settling to sleep difficulties. These sleep problems appeared in the last four years and were rated as severe. The mother stated that her child had a good sleep routine, went to bed every night at the same time and did not have problems being left alone in his own room. Although he remained in his bedroom for all the night, the next morning he would complain about feeling tired and report to have taken a long time to fall asleep. The child also informed his mother that he could not get comfortable in bed nor empty his mind of thoughts at bedtime. Although the mother reported that her own sleep was not being disturbed, she was becoming increasingly concerned about her child's sleep complaints. For this reason she was looking for professional help via her GP.

At that time Andrew was not on medication.

Assessment of changes in sleep behaviour: Andrew

Settling to sleep difficulties were rated as having NO CHANGE from baseline level. At three months follow up no changes were noted.

Visual analysis of graphed data showed that Andrew had settling to sleep difficulties occurring three or more nights a week and presented a great variability in sleep latency across all the phases of the study (Figure 5.25). Night waking data were not recorded as his mother was not aware of any nocturnal waking episodes. When the diary required indicating time and length of any waking during the night, she wrote on the diary the following statement: "*I don't know*" for the duration of the study.

Change score of the MSDQ are not available, as Andrew's mother returned the questionnaire only at baseline and failed to send it back at post-intervention phase (though she was encouraged and provided with a pre-paid addressed envelope).

At baseline Andrew's reported very high scores in the total and derivate 4 Factors (Total 34, Factor one 18, Factor two 9, Factor three 4, Factor four 3).

Means and standard deviation of diary and actigraphy data are depicted in Table 5.14. Objective sleep data are not available for Andrew as the monitor did not retain the data at baseline and the child wore the monitor after the 21st day at post intervention (the Mini Motionlogger actigraph used in this study records data for up to 21 days) despite this his mother was encouraged to put the actigraph on her child's wrist immediately after the end of treatment phase.

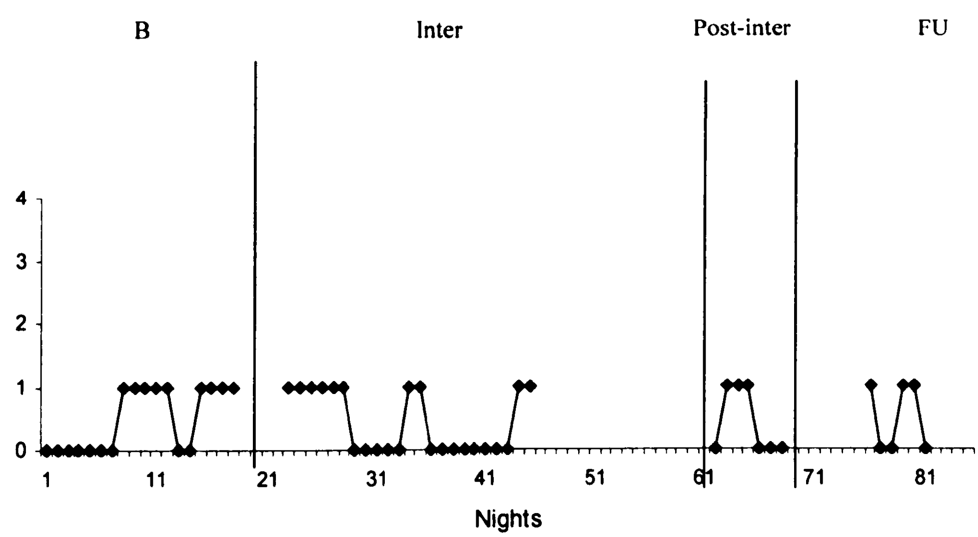

Figure 5.24 Number of problems settling to sleep during Baseline (B), intervention (inter) 1 week post intervention (post-inter) and 1 week follow-up (FU) based on diary recording, for Andrew.

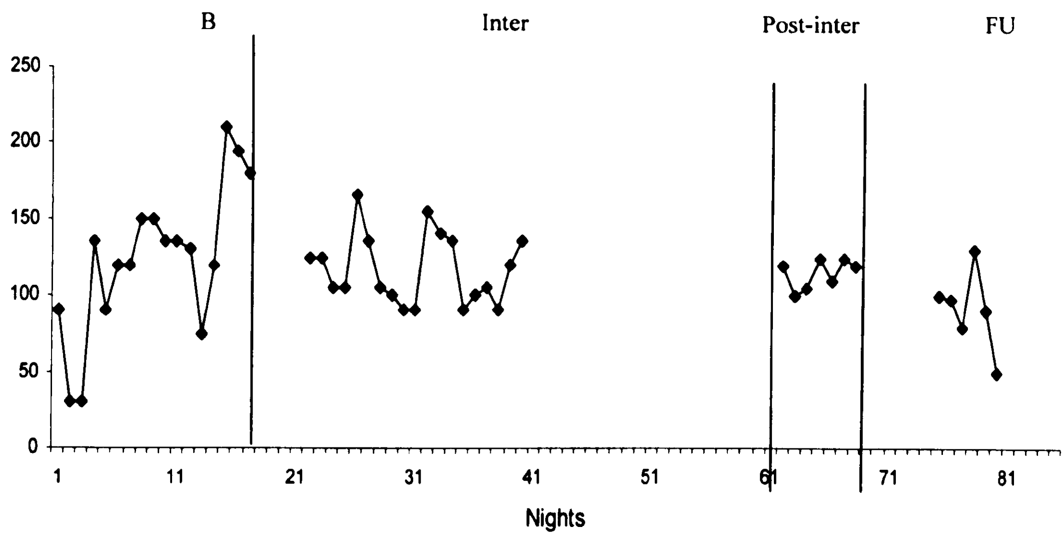

Figure 5.25 The amount of time (minutes) taken to fall asleep during baseline (B) and intervention (inter), 1 week post intervention (post-inter) and 1 week follow up (FU) based on diary recording, for Andrew.

Table 5.14 Mean (sd) of sleep variables assessed by diary for Andrew. (There are no actigraphy data available for Andrew).

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.78 (0.37)	-	123 (49.84)	-	9.99 (0.77)	-	-	-	7.08 (0.39)	-
Post-intervention	21.15 (0.58)	-	115 (10.00)	-	10.25 (0.21)	-	-	-	7.34 (0.39)	-

Assessment of change in creativity: Andrew

Table 5.15 shows the creative measure of the TTCT for Andrew.

He appeared interested in all creative tasks. He was collaborative and showed a generally positive attitude towards the test. He completed all the tasks and never complained or wanted to interrupt the activity.

All the creativity measures of the TTCT slightly increased from baseline to post intervention and were above the average level of performance (except for elaboration). The total creativity score increase by 32 points, originality and flexibility by 2 points, fluency by 10 points and elaboration by 18 points. However, there was a decline in the story-telling task for Andrew. With respect to the pre-test, there was a reduction of 14 words and 1 real fact following intervention. Also, in the reality based story, Andrew used 21 fewer words, 2 fewer real facts and number of imaginary elements remained at 0. When he was asked to invent a story based on an imaginary card, impaired performance was also noted. The number of words decreased by 8, and the number of real facts and imaginary elements introduced did not change. Andrew was able to distinguish between real and unreal cards.

Table 5.15: Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Andrew.

	NORMATIVE SAMPLE (n=1365) Mean (sd)	**Baseline score**	**Post-intervention score**
Originality score	**28 (10)**	32	34
Fluency score	**21 (6)**	20	30
Flexibility score	**16 (5)**	15	17
Elaboration score	**68 (26)**	12	30

Case consideration: Andrew

This result indicated that behavioural treatment delivered by written advice booklet might be not sufficiently effective in resolving severe settling to sleep problems in school age children with high functioning autism. Several explanations may account for this result. It is possible that Andrew's mother thought that her child's sleep problems could not be treated and it was part of the child's autistic condition. For instance, two weeks after the beginning of intervention phase, Andrew's mother wrote on the diary: *"I'm sceptical that this is going to work... you know we have struggled with sleep issues for years with Andrew and it just doesn't seem to change...he is*

autistic". Other studies with children with a learning disability have also suggested that parental beliefs that the child's sleep problem is permanent or that is due to a medical condition or is a direct result of the child's disability might all be possible reasons why some parents do not believe in the effectiveness of behavioural interventions for sleep problem and do not seek help (Bramble, 1996; Robinson & Richdale, 2004). It appeared that the interest of Andrew's mother in undertaking the intervention progressively decreased. At baseline, she was more participative, completed and returned all the questionnaires, and put the actigraphy monitor on her child's wrist. At intervention and post-intervention phases, she seemed less keen in collaborating, cancelled some of the telephone consultations planned, did not return Andrew's sleep questionnaire, did not reply to emails and did not put the monitor on her child's wrist before the actigraphy recording time expired. It also might be possible that she felt disappointed with the booklet because her initial expectations of change were unrealistic. This is unlikely due to a lack of information offered to parents during recruitment as they were all provided with information sheets where explanations and descriptions of the study were given in detail. They were also given contact numbers of the researchers and invited to call and have further information about the study. An additional reason of why the intervention was not effective might simply be because of the mother's lack of willingness to complete the treatment. She possibly was not able to commit to the intervention because it ran for 6 weeks.
Another possible reason for the lack of treatment-related change could be the older age of Andrew for whom the treatment was ineffective. The fact that an age factor might play an important role is also suggested by the steady improvement found in Dominic who had severe sleep problems and was aged 4:10 years old. On the contrary a similar improvement was not found in Andrew, who was 10:3 years old. This result is in line with a number of studies which have empirically supported the effectiveness of behavioural treatment for sleep problems in young children with autism (Weiskop et al., 2001) (Weiskop et al., 2001) and one study where it was speculated that behavioural treatment such extinction was too difficult and stressful to implement with older autistic children (Weiskop et al., 2005).

As shown in the MSDQ, at baseline sleeplessness problems appeared to be associated with pre-sleep cognitive arousal in Andrew (the MSDQ was not returned at post-intervention so change scores are not available). This suggested that cognitive factors were responsible for the maintenance of sleeplessness in Andrew. As previously mentioned, his mother reported that Andrew told her he had uncontrolled thoughts at bedtime. He often complained of having difficulties in finding a way to relax. Thus, it might be possible that the booklet-delivered behavioural intervention was not effective with him because it did not specifically address these issues.

Creative measures of the TTCT improved at post-intervention but deteriorated scores both in the real and imaginary story were found following intervention.

Case 9: Joseph

This was a 7:10 year old boy recently diagnosed with autism who scored 20 in the ASQ completed by his mother. Joseph lived with his parents (his mother was housewife and his father a website developer) and two younger brothers. Since the first visit Joseph presented a lack of eye contact and atypical fixed gaze but he appeared friendly and affable.

The boy obtained a GMA score (119), a verbal ability score (118), and spatial ability score (115) above average. Verbal and spatial scores were not significantly different at the .05 probability level.

He was reported to have had severe settling problems in settling to sleep since he was a baby. The mother described him as having a good sleep routine. He would go to bed at the same time every night after taking a bath and would stay in bed without any problems. However, his mother would hear him talking quietly to himself and see him staying still in bed with open eyes looking wide awake. During this time, he would not disturb the members of the household but the mother was concerned that her child was not getting enough sleep. At bedtime she quickly fell fast asleep whereas her child was still awake, she felt somewhat guilty about this the following morning. For this reason the mother had recently begun to look for someone who could help her child to sleep better.

Joseph was not on medication.

Assessment of changes in sleep behaviour: Joseph

Also for Joseph, sleep problems were rated as having NO CHANGE at intervention and post-intervention phase (Figure 5.26 and Figure 5.27). Three months after post-intervention, the one week sleep diary was not returned so follow-up data are not available for this child.

Change score of the MSDQ are available for Joseph since he completed the questionnaire at the two time points (baseline and post-intervention). Total MSDQ and Factor one (attribution concerning restless and agitation) reduced by 1 point, Factor two (attribution concerning mental overactivity) and Factor three (attribution concerning the consequences of insomnia) did not change and Factor four (attribution concerning lack of sleep readiness) increased by 1 point at post-intervention.

Sleep diary recording was corroborated by actigraphy data except for sleep latency with Joseph mother's tending to overestimate the time her child took to fall asleep (see Table 5.16).

According to actigraphy data there was no substantial change in total sleep time and sleep efficiency from baseline (9.90 hours and 99%) to post intervention (8.78 hours and 98%) for this child.

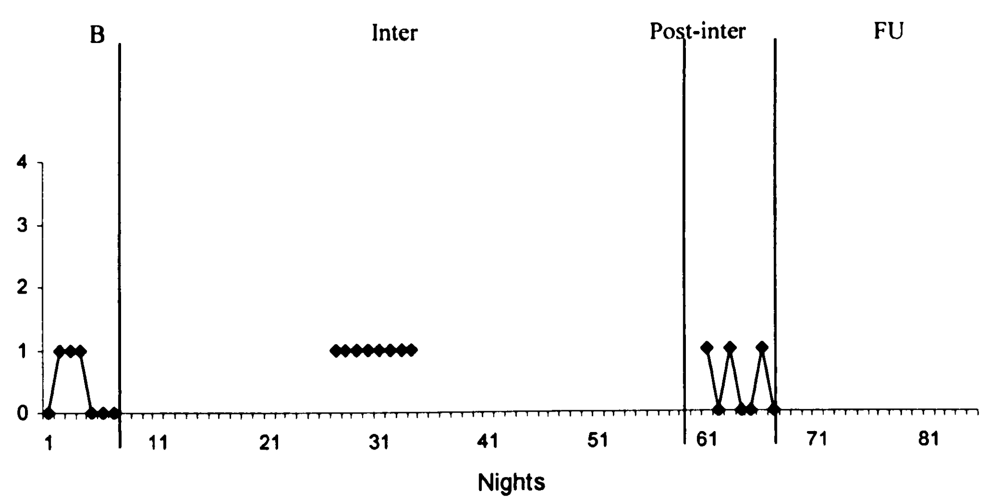

Figure 5.26 Number of problems settling to sleep during Baseline (B) , intervention (inter) and 1 week post intervention (post-inter) based on diary recording, for Joseph.

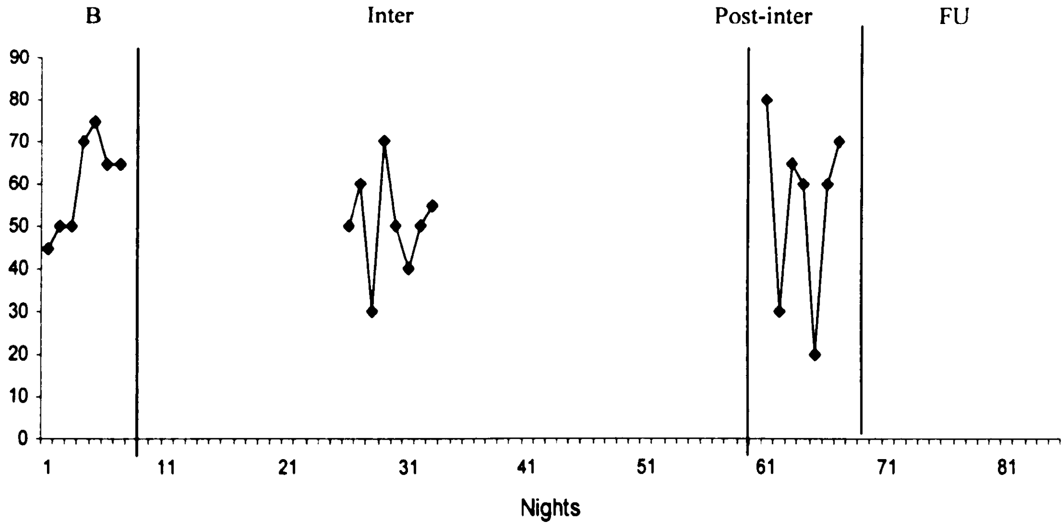

Figure 5.27 The amount of time (minutes) taken to fall asleep during baseline (B) and intervention (inter) and 1 week post intervention (post-inter) based on diary recording, for Joseph.

Table 5.16 Mean (sd) of sleep variables assessed by diary and actigraphy for Joseph

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	21.13 (0.11)	21.10 (18.02)	53.75 (11.00)	12.00 (10.89)	10.10 (0.09)	10.05 (37.50)	0 (0.00)	3.90 (3.78)	7.30 (0.40)	7.30 (30.14)
Post-intervention	21.10 (.070)	21.22 (17.29)	54.16 (23.75)	31.33 (17.31)	9.70 (0.37)	8.92 (39.86)	0 (0.00)	9.17 (6.11)	6.94 (0.32)	6.48 (34.47)

Assessment of change in creativity: Joseph

Joseph appeared interested in all creative tasks. He was collaborative and seemed to enjoy performing the drawing tasks of the TTCT and telling stories. Joseph used statements such as *"this is fun"* or *"I like drawing"*.

At post- intervention, there was a reduction in all the creative measures of the TTCT except for elaboration (see Table 5.17). Total creativity and originality scores declined quite sharply by 24 points and 20 points respectively whereas fluency decreased by 4 points and flexibility by 3 points only. Elaboration score increased slightly by 3 points at post intervention but remained far below the normal value.

Only flexibility and fluency scores stayed above the average level of performance following intervention.

However there was some improvement in the story-telling task for Joseph. With respect to the pre-test, performance improved slightly. The number of words rose by 16 and number of real facts used by 1. At post intervention Joseph used 25 more words and introduced 5 more real facts when a real card was used. In addition, Joseph used 7 imaginary elements at baseline (*1. "a blue window had turned into an oil painting", 2. "the window in the oil painting flapped and flapped", 3. "birds flew out of the painting", 4. "the super turtles", 5. "seagulls flew out of shell bashes", 6. "the boy tuned into a stone", 7. "the boy got sucked away")* did add 2 unreal events to his account at post intervention (*1. "the milkshake got bigger and bigger", 2. "a really powerful hoover that could hoover up anything"*).

When the child was asked to invent a story based on an imaginary card, impaired performance was noted. Number of words significantly decreased by 53 and number of real facts introduced reduced by 3. However, number of imaginary elements increased by 2 (*1. "the sky is full of flying guys", 2. "the guys had on their heads a black propeller"*). Joseph was able to distinguish real and unreal cards.

Table 5.17: Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Joseph

	NORMATIVE SAMPLE (n=1365) Mean (sd)	Baseline score	Post-intervention score
Originality score	**28 (10)**	41	21
Fluency score	**21 (6)**	28	24
Flexibility score	**16 (5)**	22	19
Elaboration score	**68 (26)**	12	30

Case consideration: Joseph

Again, results suggest that booklet-delivered behavioural treatment might be not effective in resolving severe sleep problems in HFA children. However, the reasons why the behavioural treatment delivered by booklet was not effective with Joseph are not completely clear. External factors might have interfered with the accomplishment of the intervention.

Behavioural treatment was, in fact, implemented during the summer period when Joseph and his family went to France for two weeks. During this period of time, phone calls and any other form of contact with the investigator were interrupted. Phone calls served several purposes: to check progress, obtain data, answer questions, assist with problems parents were experiencing, prompt appropriate behaviour and to praise mothers' success. Thus, the lack of support for two weeks might have reduced the treatment effects with Joseph. Previous studies have reported that phone calls and contact with a therapist were one of the strengths and most important aspects of a behavioural programme for sleep problems in children with an intellectual disability (Thackeray & Richdale, 2002). However another study found that a booklet, containing specific advice about behavioural techniques, was effective with and without telephone support for typically developing infants (Seymour et al., 1989). Time restriction did not allow for postponing the intervention phase until after the holiday. Before leaving for France, Joseph's mother was recommended to maintain regular sleep routines for the child. However, she confessed that being on holiday in a different country made it difficult to implement the usual sleep regime.

The mother's psychological state might also account for the fact that the behavioural treatment for sleeplessness lacked effectiveness. Joseph had recently been diagnosed with autism and his parents were not familiar with the autistic condition. At the first home visit, when she described receiving Joseph's diagnoses of autism, she reported: *"I felt in a state of shock! ... we don't know about autism"* and also in a subsequent email during post intervention phase, she wrote: *"Do you have some more special recommendations about ways of helping Joseph?"* Perhaps these statements indicate that, for Joseph, whose parents were puzzled and did not know what to do with their child's autistic condition, the behavioural intervention for sleep problem delivered by booklet was not suitable. Maybe they needed more psychological support, more information about autism and a more active form of intervention for their child's sleeplessness problem than the booklet could provide.

Alternatively, there may have been some other reasons that explain the lack of effectiveness of the intervention for Joseph. Perhaps, because the child's settling problems were not severe (according to actigraphy recording, at baseline Joseph took a mean of 12 minutes to fall asleep) it might have made it difficult to detect a substantial sleep change in this child. In terms of sleep latency, actigraphy data were not congruent with maternal report. Maybe, having recently found out that Joseph had an autistic condition made his mother oversensitive to any event during the night. However evidence is not available to support this hypothesis.

At post-intervention, substantial changes were not noted in the MSDQ scores in Joseph. Again, this suggests that the booklet-delivered behavioural treatment for sleep problems leads to changes in children's sleep habits but do not address the cognitive factors that appeared to cause and maintain these problems in Joseph.

Joseph's creativity performance in the TTCT deteriorated at post-intervention however fluency and flexibility remained in the normal range.

With regards to the story-telling task, Joseph appeared to have slightly improved in terms of number of words and real facts presented in the reality story and in terms of imaginary elements introduced in the unreal story. Given that change was not noted in terms of sleep, better creative scores cannot be attributed to improved sleep following intervention.

Case 10: Ross

This child was a 7:11 year old male with a diagnosis of autism which was also confirmed by the ASQ completed by his mother. He was assigned a score of 25. Ross was living at home with his adoptive parents and two older sisters, one of whom had Down's syndrome. Ross's mother was a housewife and his father was a taxi driver. At the beginning of the first visit, Ross displayed a lack of interest for the tasks. He appeared sad and spoke with a passive voice. After nearly 30 minutes he started moving around and showing signs of inattention. Many times he repeated, *"this is boring"*. He displayed this pattern of behaviour for the remaining three visits.

Ross obtained a GMA score (84) below the average range. When subscale scores were evaluated, the child's spatial ability (77) was classified in the low range and his verbal ability in the average range (96). A difference of 19 points between spatial and verbal ability was significant at the .05 level. Such a difference was unexpected as children with autism are usually more skilled in visual-spatial tasks than in tasks requiring skill in verbal reasoning (Sadock & Kaplan, 2008).

The mother and his older sister described him as exhibiting severe settling difficulties and a mild form of night waking. These problems appeared when the child was a few months old. He was described as being hyperaroused at bedtime, refusing to go to sleep and running away from his mother. The mother reported that it took a long time to convince him to go to his room where he spent a long time in bed engaging in writing activities or stimulating games before settling to sleep. When night waking occurred he wandered around the house, went into another room or into his sister's bed. In addition, he sometimes suffered from bedwetting and called out to his sister to be changed. The child also struggled to wake up in the morning to go to school. The mother seemed to be very frustrated with her child's sleep behaviour and was considering the possibility of looking for professional help.

Ross was not taking any medication.

<u>Assessment of changes in sleep behaviour: Ross</u>

Visual analysis of graphs showed that Ross's sleep problems had NO CHANGE at intervention, post-intervention phase and at three months follow-up.

At baseline, settling to sleep difficulties occurring three or more nights a week (Figure 5.28) and the amount of time taken to fall asleep presented a great variability from baseline to follow-up phase (Figure 5.29).

Frequency of night waking and the amount of time spent awake during each night waking episode remained the same across all phases (Figure 5.30 and Figure 5.31). According to maternal report, he continued to wake up in the night two times a week for approximately 30 minutes. During intervention period two nocturnal enuresis episodes were recorded.

Ross completed the MSDQ at baseline and post-intervention. Total MSDQ score, Factor two (attribution concerning mental overactivity) and Factor four (attribution concerning lack of sleep readiness) increased by 1 point whereas the remaining two Factors (Factors one: attribution concerning restless and agitation and Factor three: attribution concerning the consequences of insomnia) reduced by 1 point at post-intervention.

Means and standard deviation of diary and actigraphy data are presented in Table 5.18.

Except for wake up time, a substantial discrepancy between objective and subjective sleep measures were found for Ross at baseline and post-intervention. According to diary recording Ross went to bed earlier, slept longer and had shorter nocturnal waking episodes than actigraphy data indicated. This discrepancy was due to the fact that Ross was playing around with the monitor as his mother wrote on the diary several times (e.g "*took monitor off for about 10-15 minutes said he did not want it on*" or "*did not want to wear monitor had it on for a short time only*" or "*took monitor off half way, put it back in when I went to bed at about 11.45 pm*"). Difference between maternal report and objective sleep measures was very evident at post-intervention for the sleep latency parameter. Ross mother's reported a 125 minutes sleep latency whereas actigraphy recording suggested 86. This large discrepancy was due to the fact that Ross mother's put the monitor on her child's wrist when he was already asleep as he did not want to wear it.

On the basis of actigraphy recording there was no significant change in total sleep time and sleep efficiency from baseline (7.95 hours and 92%) to post intervention (7.55 hours and 95%) for Ross.

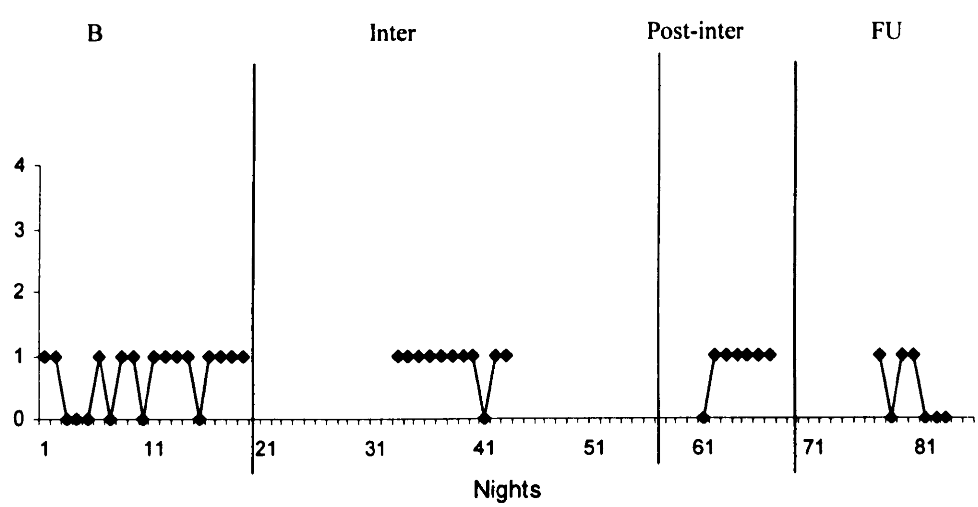

Figure 5.28 Number of problems settling to sleep during Baseline (B), intervention (inter) 1 week post intervention (post-inter) and 1 week follow-up (FU) based on diary recording, for Ross.

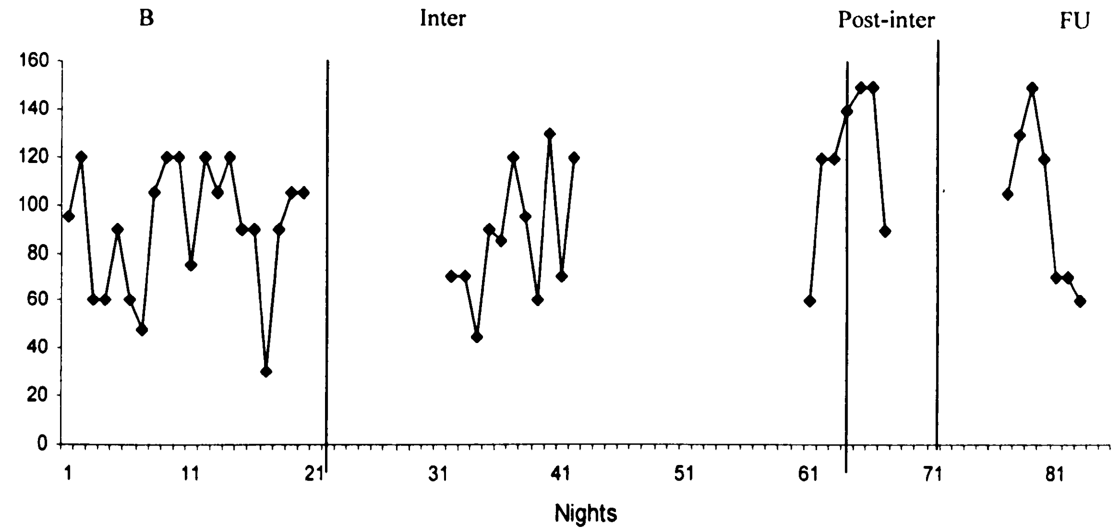

Figure 5.29 The amount of time (minutes) taken to fall asleep during baseline (B) and intervention (inter), 1 week post intervention (post-inter) and 1 week follow up (FU) based on diary recording, for Ross.

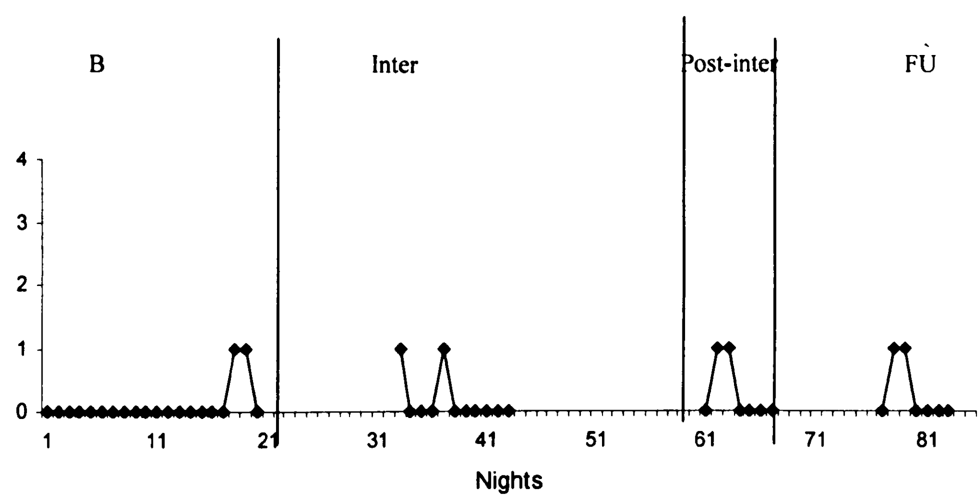

Figure 5.30 Number of night waking during Baseline (B), intervention (inter) 1 week post-intervention (post-inter) and 1 week follow up (FU) based diary recording for Ross.

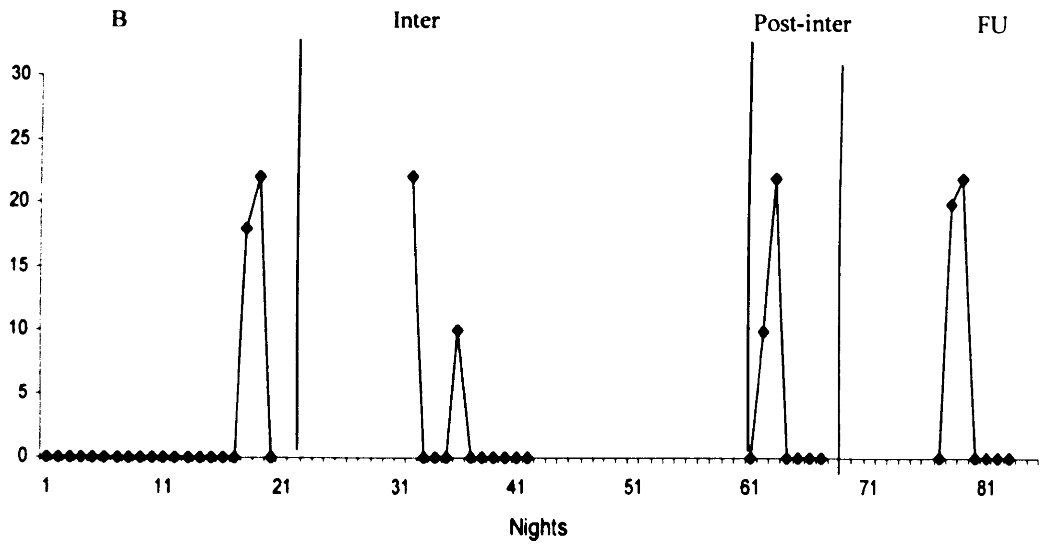

Figure 5.31 Duration of night waking per night during baseline (B), intervention (inter), 1 week post intervention (post-inter) and 1 week follow-up (FU) based on diary recording, for Ross.

Table 5.18 Mean (sd) of sleep variables assessed by diary and actigraphy for Ross

	Bedtime		**Sleep latency (minutes)**		**Sleep Duration (hours)**		**Night waking (minutes)**		**Wake up time**	
	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph	Diary	Actigraph
Baseline	20.81 (0.45)	22.08 (73.66)	100 (8.66)	65.33 (52.37)	9.82 (0.75)	8.46 (20.82)	11.50 (10.40)	39.00 (6.08)	6.69 (0.68)	7.40 (17.93)
Post-intervention	20.62 (0.79)	23.38 (34.30)	124 (43.05)	86 (1.46)	10.49 (0.74)	7.93 (44.85)	4.28 (7.86)	22.86 (14.50)	7.43 (0.52)	7.34 (26.70)

Assessment of change in creativity: Ross

Table 5.19 shows the creative measure of the TTCT for Ross.

He was reluctant to finish the tasks and he did not show enthusiasm or interest in any of the activities presented.

There were small changes from baseline to post intervention in all the creativity scores of the TTCT. Total score, originality and flexibility increased by 1 point, 2 points and 3 points respectively whereas there was a reduction by 3 points for fluency and 1 point for elaboration at post intervention. All scores remained below the average level of performance.

A decrease in the story-telling scores was also noted in both the real and unreal condition. By contrast, pre-test performance slightly improved. This might be a sign that the child felt more familiar and so relaxed with the investigator at visit three. In fact, for Ross the number of words rose by 9 and real facts introduced by 2.

At post-intervention Ross used 34 fewer words, 1 less real fact and did not introduced imaginary elements in the reality based story. Impaired performance was also noted when Ross was asked to invent a story based on an imaginary card. Number of words and imaginary elements decreased by 29 and 1 respectively and number of real facts introduced did not change. Ross was capable of differentiating between real and unreal cards.

Table 5.19: Creative scores based on TTCT at baseline and post-intervention compared to control means (standard deviation) for Ross

	NORMATIVE SAMPLE (n=1365) Mean (sd)	**Baseline score**	**Post-intervention score**
Originality score	**28 (10)**	23	25
Fluency score	**21 (6)**	18	15
Flexibility score	**16 (5)**	11	14
Elaboration score	**68 (26)**	3	2

Case consideration: Ross

Behavioural treatment delivered by written advice booklet was not effective in resolving sleep problems in Ross.

The characteristics of Ross's family in whom treatment effects were disappointing need to be considered. In conventional delivery of behavioural treatment the quality of parental relationship and emotional resources in the family as well as socioeconomic status are thought to be important factors of success (Acebo et al., 2005; Quine, 1991). For instance, family factors might have interfered with treatment success with Ross. Literacy levels of Ross's mother were low as she reported needing her older daughter's help to complete the diary and questionnaires. Perhaps a superficial understanding of the booklet's contents might have made it more difficult for Ross's mother to apply the behavioural principles described in the booklet. On the contrary, a study involving sleep disordered clinic patients with limited literacy skills found that a booklet for improving knowledge was effective (Murphy et al., 2000). However, it is important to say that the current study used a different booklet that did not just aim to alter parents' knowledge of childhood sleep problems but to change their children's sleep behaviour.

Socioeconomic status might also influence how parents regulated their child's sleep. Children in working class families tend to be out of bed later in the morning, spend more time in bed during the night, have more nocturnal wakings, lower sleep efficiency and shorter total sleep time than children in families with higher socioeconomic status (Acebo et al., 2005). In the same way, Ross who lived in a working class family went to bed later, spent more time in bed, had more night wakings (according to actigraphy recording in Table 5.18) than Joseph whose father had a professional occupation (see actigraphy data Table 5.16). Also, in line with the study by Acebo et al. (2005), Ross had lower sleep efficiency, shorter total sleep time and more nightly variability in bedtime at baseline and intervention phase than Joseph. This result might reflect differences in parents' attitudes, including attention to scheduling regular sleep periods and variable financial and social pressures.

In addition, Ross's mother was caring not only for him but also for an older daughter with Down's syndrome. Ross's mother was struggling with the complicated situation of having two children with special needs and she declared on several occasions that she felt overwhelmed. Perhaps this made it difficult for her to properly implement the behavioural treatment. In addition, she did not have the benefit of her partner's support. He was not very supportive of his wife in any of the phases of the study and when attempts were made to get him involved in the decisions about Ross's sleep scheduling, he referred the problem to his wife. Successful parenting of a child with special needs is closely intertwined with the integrity of the family itself. Maintaining a harmonious marital relationship is critical. It could be, therefore, speculated that Ross's parents needed to communicate effectively and to be able to offer support and respite for each other for the sleep treatment to be effective.

Family problems might account in part for the child's enuresis sleep episodes during treatment phase. Anxiety and stress have been associated with bedwetting, and many investigators have looked towards the family of enuretic children to locate the possible source of the problem (Goin. 1998). One study found that enuretic children were more likely to have parents with a dysfunctional relationship than non-enuretic children (Moilanen. Jarvelin. Vikevainen-Tervonen, & Huttunen, 1987). A study has also found that living with a disrupted family environment increases a child's chances of developing nocturnal incontinences (Dimitriou, Kontas, & Logothetis, 1976).

It could be also speculated that the use of a new behavioural regime provoked two nocturnal enuresis episodes seen in the child during intervention, but this is unlikely to be the case as Ross's sleep regime did not change from baseline to post-intervention. Although Ross's mother was encouraged to take her child to a physician to rule out any possible physiological cause for Ross's bedwetting problem, the child did not visit a doctor at any phase of the study.

According to the MSDQ at baseline, sleeplessness problems appeared to be associated with pre-sleep cognitive arousal in Ross. At post-intervention, substantial changes were not noted in the MSDQ scores. As for Andrew, this may suggest that the booklet addressed the behavioural component of sleep problem rather than impacting on cognitive aspects.

Creative measures of the TTCT remained below the average level of performance.

Also in the story-telling task, Ross showed deteriorated scores both in the real and imaginary story following intervention.

5.3.5 Summary of sleep and creativity results for the HFA group

According to sleep diary data, of the 4 children with HFA involved in the booklet behavioural intervention, only the 5 year old child (Dominic) who suffered multiple sleep problems showed a substantial improvement in co-sleeping and night waking problems at post intervention while no change was noted in the amount of time the child spent falling asleep. The mother of Dominic did not return the diary at follow-up, so visual analysis for follow-up data was not depicted.

For the remaining three children the intervention was not effective since no change was observed in the sleep problems reported.

Objective sleep data were available for only 2 children (Ross and Joseph) and showed no change from baseline to post-intervention. However, while a substantial discrepancy between objective and subjective sleep measures were found for Ross, sleep diary data were corroborated by actigraphy recording for Joseph (except for sleep latency which was overrated by Joseph's mother).

Booklet-delivered behavioural intervention produced very little to no improvement in the MSDQ. Children who were able to complete the questionnaire (Andrew, Joseph and Ross) displayed irregular change with total and factors scores slightly increasing or reducing within a range of 0 to 1 point. Andrew, who completed the questionnaire at baseline only, reported high total and factors scores. This suggested that cognitive components were associated with sleeplessness in this child.

Children's performance on the creative measures was poor at baseline and post-intervention. Except for one child (Andrew) who showed improved creativity scores on the TTCT, all children performed below the normal level. The same pattern of results was found for the story telling task which did not enhance following intervention. Despite children showing impaired performance in all the creative measures, these results showed that all HFA children were able to complete a figural and verbal creativity task. Creativity results are discussed in section 5.4

5.4 DISCUSSION

Booklet-delivered behavioural intervention proved to be a useful method of delivering treatment for sleeplessness in school age typical children and in an early age autistic child.

However whilst mothers no longer considered their child's sleep to be problematic, objective sleep measures did not change at post intervention. This finding suggests that booklet-delivered behavioural treatment for childhood sleep problems affects child behaviour during wakefulness and/or maternal recognition of a child sleep difficulty. Thus, children who learnt appropriate sleep behaviour were less likely to disturb their mother when they had difficulties falling asleep or when waking at night. Improvements were seen in the creativity measures in most TC but this cannot be considered a direct effect of the behavioural treatment since higher creativity scores were obtained by a child (Lucy) who reported a moderate sleep deterioration. This suggests that other factors and mechanisms account for the relationship between sleep and creativity or that multiple processes exist effecting individuals differently.

On the contrary, HFA children's creativity did not improve at post-intervention. This is not surprising since no sleep change was observed following treatment and given that poor imagination is a trait of autism.

Discussion of sleep results

The booklet-based behavioural treatment was effective with children who suffered from mild to severe sleep problems and who had multiple sleep difficulties.

Although previous studies have shown the effectiveness of written information for the treatment of sleep disturbance in infants (Weymouth et al., 1987) and pre-school children with learning disabilities (Montgomery et al., 2004), this was the first study to show the efficacy of a self-administered booklet in older TC and in a young child with HFA. This result has important clinical implications. It suggests that mothers who were reluctant to seek professional help could successfully utilise self-help information about sleep related behavioural interventions without the need for face-to-face advice. Perhaps mothers who worked outside the family home or who had more than one child to look after did not have time or economic resources to be involved in conventional therapist-driven face-to-face delivered therapy but were seeking a quick, easy and economical solution for their child's sleep disturbance. Perhaps, this is a reason why mothers took part in this study.

This is not the only work to have shown that parents do not look for professional help to resolve their child's sleep difficulties. Previous research studies have reported low treatment rates for sleep problems in children with intellectual disability (Robinson & Richdale, 2004). This finding is remarkable given the persistence of sleep problems in these children. Besides the lack of time and economic resources, there may be other reasons why mothers do not seek for professional help. Perhaps, it is because of the lack of suitably qualified health professionals in the field of sleep problems (Stores & Wiggs, 1998a) or because professionals rarely address the issue of sleep disturbance (Chervin, Archbold, Panahi, & Pituch, 2001) the mothers assume that this cannot be treated and are therefore less inclined to try to seek help. Thus, a self-administered booklet might be a valid solution for those parents who are willing to resolve their child's sleep difficulties but are not correctly informed about the possibilities of behavioural treatment for childhood sleeplessness. In fact, sleep problem education for parents is required in order to ensure that treatment is adequately implemented by families who have a child with sleep difficulties. A previous study has reported that parents who were given incorrect treatment advice did not help to improve their child's sleep (Wiggs & Store, 1996b).

Although, at post intervention the mothers no longer considered their child's sleep to be a problem, objective sleep recording suggested that children still had a sleep difficulty. This result indicates that parent recognition of childhood sleep disturbances can be influenced in the short term by factors other than objective improvement of the problems. A factor leading to a change in parent recognition may be the fact that children no longer called for attention at bedtime or when a night waking occurred. Other factors not associated with the intervention might have also changed parental perception of their child's sleep disruption. For instance, these parents may have become concerned about other, more prevalent issues (such as child illness or family issues) and their child's sleep was no longer a major concern.
However, the possibility that mothers overlooked the sleep problems at post intervention appears to be remote since they were encouraged to keep a daily sleep diary to monitor sleep changes. The fact the booklet addressed the behavioural component of sleep problems rather than impacting on objective sleep is consistent with Wiggs and Stores' (1998b) recommendation to distinguish between intervention which leads to objective changes in a child's sleep versus those that do not objectively change sleep, but which result in the child no longer disturbing their parents. It also might be that 6 weeks were not a sufficient period of time to expect behavioural therapy to lead to objective change. Future studies should implement the booklet behavioural therapy for a longer time to verify whether or not children's objective sleep changed over a prolonged intervention period.
This result suggests that behavioural intervention might still be a useful treatment component in school age typical children whose sleeplessness appeared to be more related to cognitive factors rather than behavioural. In fact, a mild reduction in pre-sleep arousal problems was reported at post intervention.

There is a vast amount of sleep literature for adults where the behavioural component together with cognitive factors is considered essential in the treatment of insomnia (Sadeh, 2005). The behavioural therapy component involves a combination of stimulus control and sleep restriction procedures which are designed to eliminate sleep incompatible behaviour and to consolidate sleep over a shorter period of time in bed. The number of school age typical children, in this study, was not large to allow a definitive conclusion on the effect of the treatment on the cognitive component associated with sleep problems. In addition, improvements in sleep were not seen in all TC and in older HFA children and when sleep enhancement was observed it was not maintained at three months follow up.

This result is in apparent disagreement with the conclusions of Montgomery et al. (2004) and Seymour et al (1989) who suggested that delivery of behavioural treatment using a booklet is effective, with benefits maintained at six months and three months post intervention respectively. It might be that, in the current study, that the positive outcomes were not maintained because mothers no longer received telephone support after the six week intervention. It is not clear how important the use of therapist support is for the effectiveness of self-administered interventions. Seymour et al (1989) failed to show any effects of adding telephone support to their trial of a booklet-delivered behavioural treatment, whereas Weymouth et al. (1987) found that some parents needed additional clinical support.

However, in the current study, is not possible to draw definitive conclusions about the impact of telephone support upon the effectiveness of the treatment because it was not always possible to provide additional help (e.g. parents were not at home or they were busy and could not speak on the telephone) and also the number of telephone support calls provided to each family was not monitored and analysed as this was not an aim of the study. Perhaps future studies, with a large sample size, could divide the children according to the severity or the type of sleep problems and investigate the effects of booklet behavioural treatment with and without telephone support on pre-sleep arousal in older children only.

Sleep disturbance in school age children with autism was less amenable to management with booklet-delivered behavioural strategies. Despite children with autism appear to have the same sleep problems as TC, these problems did not change at post-intervention. Perhaps, the severe baseline level of problems, children's older age, cognitive factors and the autistic condition might account for the lack of treatment success. Some special considerations, in fact, should be given to the application of behavioural management strategies with children with autism. The severity and chronicity of sleep problems in all these children may have necessitated long-term treatment and utilisation of different treatment strategies. For instance, the use of pharmacological intervention in conjunction with behavioural techniques has been shown to be effective in some case (Mindell & Owens, 2003). Also collaboration with a behavioural therapist or sleep specialist in designing and implementing treatment plan is necessary in case of complex, chronic or multiple sleep problems.

In fact, when treating children with autism, it is particularly important to tailor the behavioural intervention to the needs and special circumstances of the individual child and family. For instance, the lack of knowledge about autism of Joseph's mother or apparently low literacy levels of Ross's mother might have interfered with the correct treatment implementation. Given that cognitive components were associated with children's sleeplessness, it would be interesting for future studies to assess the benefits of including cognitive self-help strategies in any booklet. MSDQ appeared to be an acceptable method of assessment of mental over-activity in typical children as young as 6 years old and in school age autistic children. In spite of considerable evidence of the importance of thoughts and believe in insomnia in adults (Lichstein & Rosenthal, 1980) and children (Gregory et al., 2008) there is not gold standard measure of cognitive activity in children.

This research, therefore, provides evidence that the MSDQ can be successfully used to assess children's pre-sleep cognition. The MSDQ can be applied in future studies and in clinical settings for the effective targeting of children with therapy. It is important to highlight that MSDQ scale, being only 12 items in length, and using concise and easy to understand statements can be complete quickly by children and can also be recommended to those clinicians who want to know what children think about their own sleep difficulties. However future studies should be implemented to further assess the validity and reliability of this questionnaire.

To conclude, the booklet used in the current research appeared to be a valid instrument for the treatment of sleep disturbances in school age typical children and perhaps young children with autism whose sleep problems seemed to be more related to behavioural than cognitive factors.

Discussion of creativity results

Typical children's performance on the creative measures generally improved following intervention. Given that objective sleep did not change after the 6 week

sleep treatment, it is not clear if improved creativity is related to behavioural sleep treatment outcomes. Different interpretations can be given of this result.
It is possible that children's creative performance enhanced at post intervention because of maturational factors. Young children's brains are in fact more open to learning and enriching influences than those of adults. There are certain windows of time during which young children are especially sensitive to their environment and they can significantly change in terms of cognitive abilities (Shore, 1997).
However, it is unlikely that all children would experience such a maturational change at the same time so this explanation does not itself rule out other possible reasons for improvement found in TTCT scores following intervention.

For instance, motivational factors are especially important in the measurement of abilities involved in creative thinking. Torrance (1965) has demonstrated that a variety of motivational aspects affect creative functioning. Thus, children might have felt the need to perform better than at baseline to please the researcher, and this might have contributed in enhancing the creative score. On the other hand, this seems not to be the case since the same procedure was used at two time points and no special encouragement was given to them at the post-intervention phase.

Also the effect of practice might have biased the results. The fact that the children, at post intervention period, were familiar with the task might have made them feel more confident and might have made them perform tasks faster and consequently to have more time to add ideas to the activities proposed.
For all children, elaboration scores remained far below the average range of performance at the two time points. Because the same results were found repeatedly in all children (autistic and non autistic) it might be that this was due to the instruction used, though these were standard instructions.

Similar improvements were found in the story telling task. All children (except for Teddy whose change scores are not available) were more fluent and added more real facts at post-intervention than at baseline. However improvements were not found in the number of imaginative elements. Children introduced none or few imaginary elements in their stories. This might be due to the prompts used which were perhaps more appropriate to grown-ups (cards representing adults and/or landscapes in a real and unreal setting). It is suggested that future research introduce more suitable prompts for young children such as cards representing animals or children.

However these explanations do not clarify a) why for two TC (Esme and Teddy), TTCT creativity scores (except for flexibility and fluency) were below the average level of performance at the two time points, b) why all children show very poor elaboration scores despite the instructions according to Torrance being rigorously applied, and c) why all children showed a lack of imagination in the story-telling task. Perhaps the children were not meeting their daily sleep requirement which might have prevented them performing at a normal levels in the creativity task. Thus, reduced total sleep time and sleep efficiency according to actigraphy recording found in TC group might be associated with poor elaboration, imagination and creativity scores. This result is corroborated by a study that showed that sleep deprived participants, when tested on figural and verbal form of the TTCT had severe and persistent impairment on their performance (Horne, 1988). Another study that analysed the link between creativity, dreams, and sleep behaviours discovered that participants who were classified as "fast sleepers" (those who fell asleep in a few minutes) were more likely to score highly on a creativity test (Sladeczek & Domino, 1985).

Importantly, all children with autism except for Dominic, completed the creative tasks. This indicates that creativity assessment can be practicable with autistic children. Children showed irregular creative scores at the two time points but there was a general tendency to perform below a normal level. This confirmed previous studies that found poor creative ability in children with autism compared to typical children and children with moderate learning disability (Craig & Baron-Cohen, 1999). The theory best explaining the abnormality in the functioning of the imaginative creativity in children with autism still needs to be established. A school of thought explains this lack of creativity as an indication of executive problems (Leevers & Harris, 1998), with others explaining it as a problem with the theory of mind (Scott & Baron-Cohen, 1996).

However, Andrew's performance improved at post intervention and was above normative values. The reasons why creativity improved only for one child are obscure. It is unlikely that better creative performance was associated with improved sleep since no sleep changes were equally noted for all children with autism following intervention (except for Dominic who sleep problems improved according to diary data). As for the typical children, motivational factors might account for enhanced performance in Andrew. Creativity tests were performed by Andrew with his mother present at post intervention. Perhaps, this might have made Andrew feel more secure and perform better in order to impress his mother.

An alternative explanation is based on the creative insomnia theory (Healey & Runco, 2006). Andrew's MSDQ scores were far above the other children (with and without autism) and were at or very close to the maximum score possible. Elevated MSDQ scores indicate high mental over activity which, on the basis of the creative insomnia theory, enhances the creative process.

A further explanation might be based on maturational factors. Andrew was more than two years older than Ross and Joseph and more than five years older than Dominic. However chronological differences are not sufficient to explain differences in term of creativity scores found between these children as they had all an average or above average level of general mental ability according to the BASII.

Finally, it might be possible that creativity scores are related to the severity of the autistic condition itself. Andrew was the child with the highest score of the ASQ. Although impoverished imaginative creativity has been identified as one of the main autistic impairments by Wing and Gould (1979), high functioning autistic children might also be very creative and talented (Fizgerald, 2002).

Consistent with the low level of performance in the TTCT found in children with HFA, the story-telling scores also did not improve following intervention. Only Joseph who had the highest verbal creativity score of the BASII showed some improvement and introduced imaginary elements in his account, compared with the other autistic children.

Verbal tasks have been used successfully to investigate creative ability with 12 year old children with autism (Craig & Baron-Cohen, 2000) and with school age autistic children in the current study but the story-telling task appeared to be not very appropriate with Dominic who was aged 4:10 and who refused to invent an account at post intervention.

What is important to highlight is that all children (with and without autism) showed a severe lack of imagination and creativity. This result is in line with studies reporting a loss of originality and creativity during the developmental process. For instance

Torrance (1968) showed that creativity in children began to decline around age 6 and slumped further at age 10. Perhaps reduced creativity in children is additionally impaired by inadequate sleep which is becoming the norm in children.

Given the variation of the results with some children showing improved creativity after intervention and others showing no change, future studies need to investigate further the effect of successful behavioural treatment for sleeplessness problems on creativity using randomised controlled trial designs, a larger sample, a homogeneous group, smaller age range, numerous and more rigorous creative assessment tools. Experimental manipulation of sleep and relationship with creativity in children is an additional and different important area for future research.

5.4.1 Limitations

In this study, the small number of participants was a limitation as this prevented an initial intention to use a randomised controlled trial to assess the efficacy of the booklet-delivered behavioural treatment. Some of the reasons given by mothers for not wanting to take part in the intervention phase included not being able to commit for several weeks, fear of further deteriorating their child's sleep behaviour or considering their child's sleep problem not sufficiently severe to undertake a treatment. Perhaps lack of motivation might also account for low rate of participation. For instance, previous research with children with intellectual disability has been conducted with small sample sizes due to a lack of motivation from mothers to seek for help for their child's sleep problems (Didden, Curfs, van Driel, & de Moor, 2002).

Another limitation of the study was the relatively large age range and the heterogeneity of the sample which included typical children and children with autism. These limited interpretation regarding the efficacy of the treatment. The different findings obtained across the sample suggest that is important for samples to be diagnostically specific and of a restricted age range group to better understand what typology of children could gain the most benefit from booklet-delivered behavioural interventions. Although the small sample might not be reflective of the child population at large, issues specific to age and diagnosis were investigated with the adoption of a multiple baseline design.

A further methodological disadvantage is related to the use of the sleep diary. Parents did not complete the diary for the duration of the treatment so results are based on fragmentary information although when possible telephone support was provided to check on diary completion. Previous studies using this methodology have found similar difficulties with parents not always able to fill in a diary on a regular basis for long periods of time (Weiskop et al., 2005). Parents, in the current study, were asked to complete the diary for the duration of the study. However, when full completion was considered unrealistic, they were encouraged to complete the diary for all of the baseline period, at least during the third week of intervention period and during all of the post-intervention and follow-up period. This decision was taken to improve the quality of the mother-completed sleep diaries, since it is known that the number of omitted items from parental daily logs increases over time (Sadeh, 1994).
Another drawback of this study is that the actigraphy recording was not always possible. In some case, data were lost because the actigraph failed or the monitor's battery expired prematurely. In other cases the child refused to wear the monitor. Future study implementing a more resistant device is needed to investigate whether objective sleep changes occur following intervention in children. Given the reluctance of some children to wear the monitor and given that some children with autism were reported as not tolerating the monitor on their wrist, future studies could investigate the optimal placement for actigraphy devices (dominant vs non-dominant wrist, ankle, or torso).

A further problem was that the amount of variability in the sleep diary data made it difficult to draw conclusions from this resource. In some case, there was large variability in either baseline or in the remaining three phases which made it difficult to determine if there was a cause-effect relationship between introduction of the treatment and sleep changes. However graphing data and analysing them visually appears to be a conservative way of making judgments. The data are, in fact, raw data untouched by statistical "cooking" and when a mistake occurs, it is likely to be in the direction of saying there was no effect rather than identifying one (Bailey & Burch, 2002). Because visual analysis is open to interpretation and professionals looking at the same data might draw different conclusions, in the current study, percentage agreement between two independent raters was calculated and when disagreements occurred, these were discussed.

Furthermore, the lack of paternal participation in the study is also a limitation. Only two fathers were reported by children's mothers to have helped to implement the treatment but they never discussed possible problems or met the investigator. The reasons why they did not participate in the study are not clear. Perhaps, either lack of time or interest might be possible explanations. Fathers would have given an important contribution to the understanding of these results and future studies are needed to find new strategies to describe and explore their role and involvement in their children's sleep problems.

As discussed in section 4.3, there were limitations in terms of the new measures developed to assess children's creativity and imagination. The combining and flexibility task lacked in variability and seemed to be insufficiently challenging for school age children whereas the cards used for the story-telling task and based on Rene Magritte's pictures appeared to be unable to stimulate imagination in children. Perhaps these limitations were due to the fact that newly developed creativity measures were piloted only with 5 year old typically developing children, given the difficulties to recruit children with HFA and older TC. A further drawback was the fact that creativity scores obtained in the TTCT by children were compared to a normative sample from the USA and that norms were not available for children with autism.

5.5 MOTHERS EVALUATION OF INTERVENTION

The booklet-delivered behavioural intervention was implemented for 3 TC by mothers only and for the remaining 2 children (Esme and Lucy) by their mothers and fathers in collaboration. With regards to HFA children group, only mothers carried out the treatment. Following intervention, they evaluated their experience with the booklet by answering five questions and giving comments of what, in their opinion, were the positive and negative aspects of the treatment.

Table 5.20 shows responses to the intervention evaluation form given by mothers.

Table 5.20 : Responses to the intervention evaluation form given by mothers of TC and HF

Mothers of :	TC+S	HFA+S	Total
Who has read the Booklet?			
Mother	2	4	6
Father			
Both	3		3
Other			
No-one			
Did you start following the advice described in the booklet...			
The same night	5	3	8
After a few nights		1	1
After one week			
After two weeks			
Never			
Since the first assessment do you think that your child's sleep is ...			
A lot better	3	1	4
A little bit better	1		1
About the same	1	3	4
A little bit worse			
A lot worse			
How satisfied are you with your child's current sleep pattern?			
Completely satisfied	2		2
Mostly satisfied although some aspects still cause problems	3	3	6
Neither satisfied nor dissatisfied			
Mostly dissatisfied, although some aspects are OK			
Completely dissatisfied		1	1
Do you think that the treatment was ...			
Very helpful	2		2
A little bit helpful	3	2	5
Neither particularly helpful, nor unhelpful		2	2
Slightly unhelpful			
Very unhelpful			

5.5.1 Discussion of booklet evaluation

All mothers of TC and 2 mothers of HFA children thought that the booklet treatment was helpful to some extent. The remaining 2 mothers of HFA reported that the behavioural treatment delivered by written information was neither particularly helpful, nor unhelpful.

The responses to the open questions indicated that one of the best aspects of the treatment was to have acknowledged the good things that they were already doing with their children. For instance, mothers reported: *"The booklet reinforced the fact that we need to be consistent and follow things through"* (mother of Jack and Ben) or *"I liked the fact that we were already doing most of it"* (mother of Teddy) or *"I found it helpful to confirm we had been doing things right"* (mother of Andrew) or " *I have read books about the importance of routine at bedtime and being consistent. So from the time he was a baby I've followed the advice in the booklet and it is good to know that we did the right things"* (mother of Joseph). Thus, it might be that the reasons why the booklet was not helpful with some mothers was that it did not change parental practice.

However the booklet also proved to be insightful as mothers commented that this gave them ideas that they had never thought of before, such as: *"I had not thought of the light issue before and it explains why my child has problems with light evenings in the summer"* (mother of Lucy) or *"I liked the idea of waiting until the child was drowsy to put her to bed"* (mother of Esme) or *"The most useful advice was thinking about no stimulation before bedtime. I now make sure not to do homework with my child before bedtime or start chatting when it's bedtime"* (mother of Joseph) or *"Saying about getting your child up at the same time every day even at weekends"* (mother of Ross).

Studies have also reported that parents who used an advice booklet for management of children's night-time behaviour problems, found written information easy to understand, appropriate and helpful (Montgomery et al., 2004b) and would recommended the booklet to other parents in similar situations (Weymouth et al., 1987). However, mother's evaluations of the intervention differed, in some aspects, from what parents reported in other behavioural intervention studies. For instance, one study found that behavioural treatment delivered by booklet was helpful with or without telephone support (Seymour et al., 1989), whereas, in this study, five mothers commented that the weekly phone calls were one of the strengths of the intervention. Also a previous study identified record keeping as the least-liked by mothers (Weiskop et al., 2005) while one of the participants in the current study commented: *"It has been a very useful exercise doing a sleep diary and writing daily times"* (mother of Lucy).

Mothers of HFA children were less satisfied with the booklet and also with their child's sleep pattern post-intervention. This is congruent with the more minor improvements found in the HFA group, according to both objective and subjective sleep measures.

Some of the parents believed that the booklet was not appropriate for their child's sleep problem as they reported: *"Not enough information on ways to help children who wake up during the night"* (mother of Jack and Ben) or *"I think the treatment was helpful but I don't think was right for my child"* (mother of Ross) or *"Doesn't altogether apply. My child had a sleep issue rather than a going to bed/ staying in bed problem"* (mother of Joseph). In addition one mother believed that the booklet was not appropriate for her child's age: *"It would have been helpful to have received this when my child was younger"* (mother of Andrew).

This highlights the difficulty of trying to standardise treatment when children all have differing individual needs.

5.6 SUMMARY

The intervention study shows the effectiveness of conventional behavioural treatment for sleep problems in older typical children and one young child with autism and confirms that brief delivery of this treatment using a booklet appeared to not reduce its effect, though benefits were not maintained at three months. The booklet had an immediate good effect on children who rapidly learned to soothe themselves and not to disturb their mothers at bedtime, but did not objectively change sleep in children who perhaps tend to relapse into bad sleep habits when the treatment is applied with minor parental rigor or external support to the family is discontinued. In addition, older children, especially those with autism showed they had cognitive factors associated with their sleep difficulties which might require a different form of intervention to have a long lasting resolution of this problem.

As expected, mothers of typical children were more satisfied with the booklet than mothers of children with autism. The mothers rated highly the fact that the booklet reinforced the positive things they were doing to help their children to sleep better. The booklet made specific reference to the needs of older children which is rare in the self-help field and could be more easily followed equally by both parents so that the handling of the child was more consistent. However, in this study only two mothers who evaluated the booklet reported that both parents used it, and only one mother in the HFA group thought that the booklet was more appropriate to younger children.

Improved creativity found at post intervention in typical children is likely to be due to many factors rather than the booklet-delivered behaviour treatment being the sole or major reason for children enhanced creativity. Other variables within the child and its family need to be considered in conjunction. However reduction in sleeplessness problems is suggested as being possibly associated with children's creativity for TC. On the contrary, children with autism showed no change in the creative ability following intervention.

The results of the booklet-delivered behavioural intervention and the descriptive study reported in chapter 4 of this thesis produced some overall main conclusions which will be stated in the next and final chapter.

CHAPTER SIX

Overall conclusions

The experimental work presented and discussed in chapters four and five has a number of theoretical, methodological and clinical implications relevant for future research.

Theoretical implications

The current study suggests, as others have found (Randazzo et al. 1998), that sleep is associated with creativity hence it is important to explore the possibilities for this link. One explanation might be that sleeplessness impaired the pre-frontal cortex function seen associated with creativity. Theorists are increasingly calling for investigation on the prefrontal lobes and their specific role in creativity (Ariety, 1976; Elliott, 1986; Norlander, 1976). The evidence that frontal lobes are involved in the creative acts is shown by the fact that the prefrontal cortex is important to execute new behaviour which might involve a novel sequence of acts (Fuster, 1989) and to perform effectively neuropsychological tests of divergent thinking (Kolb & Whishaw, 1985; Milner & Pedrides, 1984; Struss & Benson, 1986). The normal functioning of the frontal lobes appears to be damage by insufficient sleep. Few studies have in fact shown deficits in the executive function of the frontal lobes following sleep deprivation in adults (Horne & Pettitt, 1985). Sleep deprived subjects have difficulties thinking of imaginative words or ideas. Instead, they tend to choose repetitious words or clichéd phrases. Also, sleep deprived people do not have the speed or creative abilities to cope with making quick but logical decisions, nor do they have the ability to implement them well (Horne, 1993).

The prefrontal cortex has also been extensively implicated in autism to explain deficits in executive and other higher-order functions related to cognition. Studies have used the executive dysfunction hypothesis to explain the lack of creativity in children with autism (Leevers & Harris 1998). Executive function, in fact, referred to high level cognitive capacities such flexibility, fluency and originality which are impaired in autism and might be further damage by lack of sleep. However, in the descriptive part of this study objective sleep did not appear to be associated with creativity in TC and HFA children. In addition, creativity improvement seen in TC

following treatment was not accompanied by changes in objective sleep. This might be due to the limitation of actigraphy as a measure in that it does not provide detailed information about sleep, other than identifying basic sleep/wake states, and it may be that more sophisticated analysis of aspects of sleep (e.g. sleep stages or arousals) would be linked to prefrontal cortex functions such as creative thinking. There are in fact a few objective studies of sleep that have shown REM sleep in particular as an enhancing factor for divergent thinking (Lewing and Glaubman, 1974) and have demonstrated that aminergic neurotransmitters which reduce cognitive flexibility are inhibited during sleep (Beversdorf et al., 1999) still others have show that all sleep phases are important for the creative process (Wagner et al., 2004).
REM sleep might be important also in the creativity of subjects with autism. For instance, REM sleep disruption (Godbout, et al., 2000) hypoactivation of REM sleep (Limoges et al., 2005), and lower beta activity during REM sleep (Daoust et al., 2004) observed in subjects with autism possibly play a key role in the impaired creativity seen in this clinical group. Thus, there is a need for more PSG studies to clarify what are the aspects of sleep to improve creativity in TC and whether or not the abnormal sleep architecture seen in autistic persons (Stores & Wiggs, 1998b) impairs their ability to be creative. To have a better understanding of the nature of this relationship, additional study should also be carried out with children with autism who are not affected by sleeplessness .

Given that objective sleep, as assessed by actigraphy, did not appear to be linked with creativity further possibilities for the mechanism need to be considered. The enhanced creativity seen post-intervention in some TC might be due to decreased arousal levels. A state of hyper-arousal suggested by the sleepless children's self-report data (but not necessarily picked up by actigraphy recording) was perhaps reduced by behavioural intervention resulting in a more tranquil sleep period for the TC. In contrast the lack of change in the arousal level seen in the HFA children did not lead to enhancement in their creativity ability. Various investigations have showed that creativity is more likely to occur in low-arousal state. Studies of creativity (Dentler & Mackler, 1964; Krop, Alegre, & Williams, 1969) have demonstrated that stress produces a decrease in originality. Even the excitement/arousal caused by rewards seems to decrease creativity (Amabile, 1983). Martindale and Hasenfus (1978) showed that creative persons, when asked to be original, exhibited defocused attention accompanied by low levels of cortical activation. On the other hand, non- creative people focused their attention too much, and this prevented them from thinking of original ideas. However, no difference was found between creative and non-creative people when they were not asked to be original. Thus, the authors proposed that low cortical arousal would allow defocused attention which would, in turn, facilitate the associations that provide original insights.

The results of the current study support the notion that reduction of arousal in children with sleeplessness problems can be beneficial for their creativity.

Beside these theoretical explanations for why sleep might be directly linked to creativity there are also a number of indirect factors which perhaps are responsible for this relationship. For instance uneven cognitive abilities as reported in the British Ability Scale second edition (BASII) in TC and HFA with sleeplessness problems might underlie both sleep and creativity. As explained in section 4.3 discrepancy in the cognitive skills might be indicative of cognitive problems and is frequently found in children with developmental disorders (Mervis et al., 1999) and neurological disease (Brookshire at al.,1995). Thus, children with uneven intellectual ability might find more difficult to learn appropriate sleep behaviour, they might be less independent and require parental attention at bedtime. On the other hand, children who are less skilled on one area of competence (e.g verbal skills or drawing abilities) might have problems in executing a verbal or figural creativity task, might be more insecure and stressed and consequently less original. Additional studies are needed to investigate if sleeplessness is associated with cognitive discrepancies favouring spatial or verbal skills and if reduced ability in one area is more linked with impaired creativity than the other.

It might also be possible that TC and HFA with sleeplessness problems have reduced creativity because these problems made mothers feel stressed, tired and negative towards the child rather than the sleep problem being directly associated with the child's impaired creativity ability. The fact that sleep problems in children might negatively affect the mother-child relationship has been well established. For instance a study reported that American mothers admitted fantasies of aggression towards their child who had sleep problems (Levitzky & Cooper, 2000). Given that family and education are important component in the development of creative ability in children

(Feldman, 1999), this suggests that the relationship parent-child might be a third variable underlying sleep and creativity.

Also other variables such as mothers' personality or coping style, not investigated in this thesis, may be an important factor associated with sleep and creativity in children. For instance mothers who are not capable to deal with their child's sleep problems and are stressed by the difficulties associated with these problems might be not able to create a stimulating, supportive and nurturing environment for their child and consequently child's creativity is affected. Additional research in relation to intrinsic parent factors related to specific characteristics of mothers (e.g personality, educational level, perceive control) that might affect the development of creativity in children who have sleep problems is warranted.

Methodological implication

The studies reported here also have some methodological implications for future studies assessing sleep in childhood. As discussed in section 4.3.2 this research constitutes one of the few attempts to examine children's self reports of sleep. Resistance to the use of questionnaires with children has had its roots in genuine concerns about whether younger people have the cognitive and communicative skills necessary for providing good quality responses to survey questions (Bell, 2007). Only in the last 10–15 years have social researchers come to acknowledge the importance of conducting survey research with children directly, rather than relying on the data collected from adults (Scott, 1997).

In the current study, the good correspondences between parental and child questionnaire data confirmed school age children as an important source of information and suggest that children's self report could be used more extensively in sleep research. There is a growing body of evidence that shows that children are able to provide more reliable information about themselves with respect to a range of issues than adults who know them well (Tizard, 1986). The Children's Sleep Habits Questionnaire (child version) (CSHQ) and the Modified Sleep Disturbance Questionnaire (MSDQ), in fact, generated a more in-depth understanding of what keeps children awake at night. Sleep questionnaires for young people might be especially useful with school age children who have difficulties going to sleep rather than problems going to bed. In this study the MSDQ showed that children experienced excessive cognitive activity while trying to get to sleep which would not have been picked up if children's sleep was evaluated with the parents' questionnaire and objective measures only. Insomnia is in fact a subjectively defined problem that only those personally affected might be able to reveal. Older children with sleeplessness problems may not necessarily make their sleep difficulties known to parents and even objective measurement of sleep, by means of actigraphy might not be a reliable measure of insomnia if a child is kept awake by intrusive thoughts but remains quietly in bed.

Children's self report appeared to be useful also with HFA children. Children with autism might reasonably be expected to experience difficulties in expressing their feelings and thoughts. Hence, a structured questionnaire with short sentences and straightforward language such as CSHQ and MSDQ might have helped them to identify the factors associated with sleeplessness difficulties and encouraged them to discuss these problems with parents. In fact it might be especially difficult for children to communicate in their own words, the thoughts or bodily sensations that accompany insomnia. Parents themselves might find children's questionnaire responses to be informative and consequently they might be more able to provide more appropriate help. Sleep questionnaires for children could be, in fact, a way to enhance awareness in those parents who do not realise their child has difficulty falling asleep.

The lack of correspondence between subjective and objective sleep measures found in this study highlights the complex nature of childhood insomnia and raise important questions about the usefulness of actigraphy for the evaluation of sleeplessness in childhood. What makes this condition particularly challenging to evaluate is that sleeplessness is more than a physiological phenomenon. There is a vast sleep literature with adults which documents how some people complain of insomnia but do not show any objective evidence of poor sleep, while others may experience sleep disruptions, but are not concerned and do not complain about them (Morin, 1993). The most central feature of insomnia is the subjective perception of inadequate sleep duration or quality hence its clinical significance in childhood should be judged by its impact on daytime functioning, school performance, mood, peer and family relationship. However, it is only by including multiple measures of sleep that this disparity becomes apparent and, with this, our understanding of the nature of insomnia is improved.

Future studies should try to clarify the degree of discrepancy between subjective and objective sleep measures in children. Studies with adults show that usually individuals fall along a continuum, including at one end, cases of pure subjective complaint of insomnia without any objective finding and, at the other end, subjective complaint fully corroborated by objective findings. Similar research into the degree of correspondence between measures has not been carried out with children. For example the proportion of cases with purely subjective complaints of insomnia is relatively low among treatment-seeking adults (Edinger & Fins, 1995) but similar information is not available in the sleep literature for children.

The methodological implications of the research go beyond the area of sleep and sleeplessness. In this study novel tasks (combining and flexibility test and story telling task) were designed to assess creativity in childhood. While these tests have some limitations as discussed in section 4.3, they constitute an attempt to better understand creativity in children. Although creative thinking is an important "natural resource" (Guilford, 1950) allowing us to adapt and properly respond to the environment, the abilities involved in creativity (e.g sensitivity to problems, capacity to produce many ideas, the ability to change one's mental set and the ability to be original) might be difficult to measure. In contrast to many standardized tests of convergent thinking which require only one correct answer, there are few standardized and internationally used tests of creativity. New creativity tests used in the current study were also designed to be administered to children with autism. The tests were in fact simple, easy to understand and based on previous tasks used with children with HFA (Craig et al., 2001; Craig & Baron-Cohen, 2000). There are very few experimental studies in autism that have included the development of creativity tests. For instance, in Frith's (1972) study different coloured rubber stamps were used with autistic children to assess creativity. Children produced less varied patterns relative to control showing lack of creativity. A similar conclusion was reached by Lewis and Boucher (1991) who utilized a drawing task and found that the content of the drawings of children with autism was less varied. More recently Craig and Baron-Cohen (1999) using a test of imaginative fluency (children were given a three-dimensional shape and asked what it could be) found out that children with autism exhibited reduced fluency as well as generated fewer imaginative responses. While all these studies indicated that creativity was impaired in autism there is still today a lack of appropriate measures to establish the nature of the creativity deficit in autism. For instance, it remains unclear if children with autism have a specific impairment in the ability to imagine and draw unreal things as suggested by Scott & Baron-Cohen, (1996) or they have difficulties in the planning or execution of pictures both of real or unreal entity as indicated by Leevers and Harris (1998). The discrepancy between Scott & Baron-Cohen, (1996) and Leevers and Harris (1998) theories' might reflect a methodological limitation in the tests used to assess creativity. In addition, in these two studies children performed different tasks and so results are difficult to compare. Developing appropriate tasks to assess these two hypotheses is a priority for future research in this area. There is also a need to replicate the current study with the use of both the same and different creativity measures to allow comparisons.

Clinical implications

The results of the current study show that booklet-delivered behavioural advice might be a successful method for improving the sleeping difficulties in school age TC. This suggests that mothers of TC can successfully use self-help information without the need for professional help and that TC can learn appropriate sleep behaviours (e.g do not disturb parents at bedtime, sleep in their own bed) with only the support of written advice given to their parents. Thus, a booklet-based behavioural intervention appeared to be a quick, easy and economical solution for the child's sleep disturbance. At present, many health care professionals overlook sleep problems in children and do not offer treatment (Chervin et al., 2001). Although this is clearly poor practice the results of the current study suggest that, for some children at least, parental education by means of a booklet might be a way to overcome some of the problems with accessing treatment.

However, it is perhaps not surprising that the booklet was able to reduce HFA children's sleep difficulties only in one case. Perhaps HFA children who were reported to have challenging behaviour, had severe baseline levels of sleep problems and needed extra support or a different form of intervention. The booklet information used in the current study with school-age children was based on a booklet written for addressing the sleeplessness problems of younger children, written by the Child Psychology Department of Dudley Road Hospital, West Birmingham. Although the text, images and the information included was then modified so as to be more appropriated to the sleep issues of older children and those with autism, some mothers reported that the intervention did not meet the specific needs of their children. Children with autism appeared to have sleep issues which went beyond the behavioural.

Future research could consider using this booklet together with professional help. Despite the intention to support and encourage mothers who took part in this study with weekly telephone calls, this was not always possible (e.g. a family went on holiday for two weeks). The positive effect of the advice booklet for sleep problems in children might be enhanced by adding telephone support. This is documented by a study (Wymouth et al., 1987) that showed that some parents could reduce their child's sleep problems with the use of a self-administered treatment only, whereas others required additional clinical support.

Given that there appears to be a relationship between sleep and creativity (Horne, 1988a), it was thought that it may be possible to improve creativity by means of enhancing sleep. As the objective sleep of children (with and without HFA) did not improve, it is perhaps not surprising that children with HFA showed no change in the creativity measures following treatment. In contrast, the enhanced creativity seen in TC at post intervention is less easy to explain (given that there was no objective change in their sleep patterns either). Thus it appeared that other factors, rather than improved objective sleep, are responsible for improved creativity found at post intervention in these children. Future research is required to fully understand the mechanism underlying this result. As outlined above it is possible that successful treatment changed the subjective child's experience of being awake, resulting in reduced nocturnal arousal. High arousal has been seen to be associated with compromised creativity (Dentler & Mackler, 1964; Krop, Alegre, & Williams, 1969).

It is also possible that behavioural treatment, whilst not having a direct effect on the children's objective child sleep, improved the relationship between mother and child which .had indirect benefits for the child's creativity For example, the treatment might have resulted in a child with night waking problems continuing to wake but no longer signalling this awake state to parents and remaining silent. This would improve parent sleep quality, resulting in the parent feeling less tired and so better able to interact with the child during the day time. Thus, it might be possible that booklet behavioural intervention indirectly affected children's creativity by improving the relationship between mother and child.

Improved creativity in TC might be due to other factors not necessarily associated with the behavioural intervention. For instance, situational factors related to the child, parent or family context might have affected the creativity results. For instance, the post test for one child took place just before his birthday party. This could have made a positive impact on his mood and consequently improved performance on the creativity test. It would be useful to conduct a randomised control trial to have a full understanding of the mechanisms underlying treatment for sleep problems and the effect these have on creativity.

Given that sleep changes were observed only in the TC group, it is not surprising that while mothers of TC believed that the booklet treatment was somehow helpful, mothers of children with autism were less satisfied with the intervention. It is also noteworthy that children in both groups complained of having incontrollable thoughts. Cognitive behavioral therapy might help them to identify and correct these inappropriate thoughts and beliefs that contribute to insomnia, as well as give parents the proper information about sleep, age-related sleep changes and reasonable sleep goals. For instance, self-hypnosis, thought stopping, visual imagery, in conjunction with the behavioral methods described in the booklet, may be more helpful for overcoming insomnia in school-age children than behavioral approaches in isolation. Future studies assessing the relative efficacy of behaviour therapy, cognitive therapy and combined approaches for the treatment of sleeplessness in school age children with and without autism would seem appropriate.

Overall, the experimental work presented in this thesis suggests that sleeplessness problems are associated with high level cognitive ability in children and adversely affect maternal mental health. The mechanism by which sleep and creativity are associated remains speculative although actigraphically assessed objective sleep variables or sleep times, duration and gross-continuity, seem unlikely to play a role. The results are clinically useful as they support the use of booklet-based behavioural interventions for sleeplessness in school age TC. The efficacy of such approach with children with HFA needs to be further explored. Nocturnal mental over-activity appeared to play a key maintenance role in sleeplessness in both TC and HFA. This finding strongly suggests that models of childhood sleeplessness need to include cognitive components and this has important clinical implications when considering appropriate intervention approaches for this age group.

REFERENCES

Acebo, C., Sadeh, A., Seifer, R., Tzischinsky, O., Hafer, A., & Carskadon, M. A. (2005). Sleep/wake patterns derived from activity monitoring and maternal report for healthy 1- to 5-year-old children. *Sleep, 28*(12), 1568-1577.

Achenbach, T. M. (1991). *Manual for Child Behavior Checklist/4-18 and 1991 Profile*. Burlington,VT: University of Vermont, Department of Psychiatry.

Adams, L. A., & Rickert, V. I. (1989). Reducing bedtime tantrums: comparison between positive routines and graduated extinction. *Pediatrics, 84*(5), 756-761.

Agnew, H. W. J., Webb, W. B., & Williams, R. L. (1966). The first night effect: an EEG study of sleep. *Psychophysiology, 2*(3), 263-266.

Akshoomoff, N. (2002). Selective attention and active engagement in young children. *Developmental Neuropsychology, 22*(3), 625-642.

Albert, R. S., & Runco, M. (1988). Independence and cognitive ability in gifted and exceptionally gifted boys. *Journal of Youth and Adolescence, 18*(3) 221-230.

Allen, R. P. (2003). Does a modest loss of sleep affect neurocognitive functioning of children? *Sleep Medicine, 4*(4), 353-355.

Allik, H. (2006). *Asperger syndrome and high functioning autism in school age children: the children's sleep and behaviour and aspects of their parents' well-being.*: Doctoral dissertation. Stockholm: Karolinska Institute.

Allik, H., Larsson, J. O., & Smedje, H. (2006). Insomnia in school-age children with Asperger Syndrome or high-functioning autism. *BMC Psychiatry, 6*(18) 1-11.

Amabile, T. (1983). *The Social Psychology of Creativity*. New York, NY: Springer-Verlag.

Anders, T. F. (2004). Sleep-wake states and problems and child psychosocial development. In *Encyclopedia on Early Childhood Development*, retrieved from http//www.excellence-earlychildhood.ca/documents/Anders/ANGxp.pdf.

Anders, T. F., Carskadon, M. A., Dement, W. C., & Harvey, K. (1978). Sleep habits of children and the identification of pathologically sleepy children. *Child Psychiatry and Human Development, 9*(1), 56-63.

Anders, T. F., & Sostek, A. M. (1976). The use of time lapse video recording of sleep-wake behavior in human infants. *Psychophysiology, 13*(2), 155-158.

APA. (1994). *DSM-IV Diagnostic and Statistical Manual of Mental Disorders, 4th edition*: Washington, DC: American Psychiatry Association.

APA. (2000). *DSM-IV Diagnostic and Statistical Manual of Mental Disorders, 4th edition, text rev*: Washington, DC: American Psychiatry Association.

Ariety, S. (1976). *The magic synthesis*. New York, NY: Basic Books.

Armour, D., & Paton, C. (2004). Melatonin in the treatment of insomnia in children and adolescents. *Psychiatric Bulletin, 28*(6), 222-224.

Armstrong, K. L., O'Donnell, H., McCallum, R., & Dadds, M. (1998). Childhood sleep problems: association with prenatal factors and maternal distress/depression. *Journal of Paediatric Child Health, 34*(3), 263-266.

Aronen, E. T., Paavonen, E. J., Fjallberg, M., Soininen, M., & Torronen, J. (2000). Sleep and psychiatric symptoms in school-age children. *Journal of the American Academy Child Adolescent Psychiatry, 39*(4), 502-508.

ASDA. (1990). *The International Classification of Sleep Disorders. Diagnostic and Coding Manual*. Lawrence, KS: Allen Press

ASDA. (1997). *The international Classification of sleep disorders. Diagnostic and coding Manual- Revised*: Rochester, MN: American Sleep Disorders Association.

ASDA. (2005). *The International Classification of Sleep Disorders: Diagnostic and Coding Manual, 2nd ed*: Westchester, IL: American Academy of Sleep Medicine

Asperger, H. (1944). Die "Autistischen Psychopathen" im Kindesalter. *Archiv fur Psychiatrie und Nervenkrankheiten, 117*, 76-136.

Attwood, T. (1998). *Aspergers Syndrome. A guide for parents and professionals.* London, UK: Jessica Kingsley Publishers.

Baddeley, A. (2000). The episodic buffer: a new component of working memory? *Trends of Cognitive Sciences, 4*(11), 417-423.

Bailey, J., & Burch, M. (2002). *Research Methods in Applied Behavior Analysis.* London, UK: Sage Publications.

Baker, B. L., Blacher, J., Crnic, K. A., & Edelbrock, C. (2002). Behavior problems and parenting stress in families of three-year old children with and without developmental delays *American Journal on Mental Retardation, 107*(6), 433-444.

Baron-Cohen, S., Richler, J., Bisarya, D., Gurunathan, N., & Wheelwright, S. (2003). The systemizing quotient: an investigation of adults with Asperger syndrome or high-functioning autism, and normal sex differences. *Philosophical transactions of the Royal Society of London. Series B, Biological sciences, 358*(1430), 361-374.

Barret, S., Prior, M., & Manjiviona, J. (2004). Children on the Borderlands of Autism Differential Characteristics in Social, Imaginative, Communicative and Repetitive Behaviour Domains. *Autism, 8*(1), 61-87.

Barron, F., & Harrington, D. M. (1981). Creativity, intelligence, and personality. *Annual Review of Psychology, 32*, 439-476.

Bearpark, H. M., & Michie, P. T. (1987). Prevalence of sleep/wake disturbance in Sydney adolescents. *Sleep Research, 16*, 304.

Beebe, D., Lewin, D., Zeller, M., MacCabe, M., MAcLeod, K., Daniels, S., et al. (2006). Sleep in overweight adolescents: shorter sleep, poorer sleep quality, sleepiness, and sleep disordered breathing. *Journal of Pediatric Psychology 32*(1), 69-79.

Been, H. (1997). Dreams: the convergence of neurobiologic and psychoanalytic perspectives.
Journal of the American Academy of Psychoanalysis, 25(4), 639-654.

Bell, A. (2007). Designing and testing questionnaires for children. *Journal of Research in Nursing.* 12 (5), 461-469

Beltramini, A. U., & Hertzig, M. E. (1983). Sleep and bedtime behavior in preschool-aged children. *Pediatrics, 71*(2), 153-158.

Bennetto, L., Pennington, B. F., & Rogers, S. J. (1996). Intact and impaired memory functions in autism. *Child Development, 67*(4), 1816-1835.

Benoit, D., Zeanah, C. H., Boucher, C., & Minde, K. K. (1992). Sleep disorders in early childhood: association with insecure maternal attachment. *Journal of the American Academy of Child and Adolescent Psychiatry, 31*(1), 86-93.

Berger, H. (1930). Uber das elektroenkephalogramm des Menschen. *Journal of Psychiatry & Neuroscience, 40*, 160-179.

Berger, R. J., & Oswald, I. (1962). Effects of sleep deprivation on behaviour, subsequent sleep, and dreaming. *The Journal of Mental Science, 108*, 457-465.

Berument, S. K., Rutter, M., Lord, C., Pickles, A., & Bailey, A. (1999). Autism screening questionnaire: diagnostic validity. *British Journal of Psychiatry, 175*, 444-451.

Beversdorf, D., Hughes, D., Steingerg, B., Lewis, L., & Heilman, K. (1999). Noradrenergic modulation of cognitive flexibility in problem solving. *Neuroreport, 10*(13), 2763-2767.

Bishop, D. V. M., & Butterworth, G. E. (1979). A longitudinal study using the WPPSI and WISC-R with an English sample. *British Journal of Educational Psychology 49*(2), 156-168.

Bishop, D., & Norbury, C. (2002). Exploring the borderlands of autistic disorder and specific language impairment: a study using standardised diagnostic instruments. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 43*(7), 917-929.

Blader, J. C., Koplewicz, H. S., Abikoff, H., & Foley, C. (1997). Sleep problems of elementary school children. A community survey. *Archives of Pediatrics & Adolescent Medicine, 151*(5), 473-480.

Bloch, V., Hennevin, E., & Leconte, P. (1981). The phenomenon of paradoxical sleep augmentation after learning: experimental studies of its characteristics and significance. In W. Fishbein (Eds.), *Sleep, dreams, and memory* (pp. 1-18): Jamaica, NY: Spectrum.

Bodary, D. L., & Miller, L. D. (2000). Neurobiological substrates of communicator style *Communication Education, 49*(1), 82-98.

Bolte, S., Crecelius, K., & Poustka, F. (2000). The Questionnaire on Behaviour and Social Communication (VSK): An autism screening instrument for research and practice. *Diagnostica, 46*(3), 149-155.

Borbely, A., & Achermann, P. (2005). Sleep homeostasis and models of sleep regulation. In R. D. Kryger (Eds.), *Principles and practice of sleep medicine 4th edition* (pp. 405-417). Philadelphia: Elsevier Saunders.

Born, J., & Fehm, H. L. (1998). Hypothalamus-pituitary-adrenal activity during human sleep: a coordinating role for the limbic hippocampal system. *Experimental and Clinical Endocrinology & Diabetes, 106*(3), 153-163.

Born, J., Muth, S., & Fehm, H. L. (1988). The significance of sleep onset and slow wave sleep for nocturnal release of growth hormone (GH) and cortisol. *Psychoneuroendocrinology, 13*(3), 233-243.

Bramble, D. (1996). Consumer opinion concerning the treatment of a common sleep problem. *Child: Care, Health and Development, 22*(6), 355-366.

Bramble, D. (2003). Annotation: The use of psychotropic medications in children: a British view. *Journal of Child Psychology and Psychiatry, 44*(2), 169-179.

Bristol, M. M. (1987). Mothers of children with autism or communication disorders: Successful adaptation and the Double ABCX model. *Journal of Autism and Developmental Disorders, 17*(4), 469-486.

Brookshire, B. L., Fletcher, J. M., Bohan, T. P., Landry, S. H., Davidson, K. C., & Francis, D. J. (1995). Verbal and Nonverbal Skill Discrepancies in Children with Hydrocephalus: A Five-Year Longitudinal Follow-Up1. *Journal of Paediatric Psychology 20*(6), 785-800.

Broughton, R. (1982). Human consciousness and sleep/waking rhythms: A review and some neuropsychological considerations. *Journal of Clinical and Experimental Neuropsychology, 4*(3), 193-218.

Bruni, O., Ottaviano, S., Guidetti, V., Romoli, M., Innocenzi, M., Cortesi, F., et al. (1996). The Sleep Disturbance Scale for Children (SDSC). Construction and validation of an instrument to evaluate sleep disturbances in childhood and adolescence. *Journal of Sleep Research, 5*(4), 251-261.

Bruni, O., Proietti, C., Ottaviano, S., Lucchetti, A., Rinna, A., Cortesi, F., et al. (1998). Trattamento non farmacologico in ambito psichiatrico. In G. Bonsignore, G. Insalaco & S. Smirne (Eds.), *Il sonno in Italia* (pp. 94-98): Milan, IT: Poletto Editore.

Cano, A., Sprafkin, R. P., Scaturo, D. J., Lantinga, L. J., Fiese, B. H., & Brand, F. (2001). Mental Health Screening in Primary Care: A Comparison of 3 Brief Measures of Psychological Distress. *Primary care companion to the Journal of clinical psychiatry, 3*(5), 206-210.

Capaldi, V. F., Handwerger, K., Richardson, E., & Stroud, L. R. (2005). Associations between sleep and cortisol responses to stress in children and adolescents: a pilot study. *Behavioral Sleep Medicine, 3*(4), 177-192.

Carskadon, M. A. (1982). The second decade. In C. Guilleminault (Eds.), *Sleeping and waking disorders: Indications and Techniques* (pp. 99-125).Menlo Park, CA: Addison-Wesley

Carskadon, M. A. (2002). *Adolescent sleep patterns: Biological, social, and psychological influences*: Cambridge, Cambridge University Press.

Carskadon, M. A., Acebo, C., Richardson, G. S., Tate, B. A., & Seifer, R. (1997). An approach to studying circadian rhythms of adolescent humans. *Journal of Biological Rhythms, 12*(3), 278-289.

Carskadon, M. A., & Dement, W. C. (1981a). Acute restriction of nocturnal sleep in children. *Perceptual and Motor Skills, 53*, 103-112.

Carskadon, M. A., & Dement, W. C. (1981b). Cumulative effects of sleep restriction on daytime sleepiness. *Psychophysiology, 18*(2), 107-113.

Carskadon, M. A., Harvey, K., & Dement, W. C. (1981c). Sleep loss in young adolescents. *Sleep, 4*(3), 299-312.

Carskadon, M. A., Vieira, C., & Acebo, C. (1993). Association between puberty and delayed phase preference. *Sleep, 16*(3), 258-262.

Chambers, M. J. (1994). Actigraphy and insomnia: a closer look; part 1. *Sleep, 17(5)*, 405-408.

Chamberlain, R. S., & Herman, B. H. (1990). A novel biochemical model linking dysfunctions in brain melatonin, proopiomelanocortin peptides, and serotonin in autism. *Biological Psychiatry, 28*(9), 773-793.

Charman, T., Howlin, P., Berry, B., & Prince, E. (2004). Measuring developmental progress of children with autism spectrum disorder on school entry using parent report. *Autism, 8*(1), 89-100.

Chervin, R. D., Archbold, K. H., Panahi, P., & Pituch, K. J. (2001). Sleep problems seldom addressed at two general pediatric clinics. *Pediatrics, 107*(6), 1375-1380.

Clements, J., Wing, L., & Dunn, G. (1986). Sleep problems in handicapped children: a preliminary study. *Journal of Child Psychology and Psychiatry, 27*(3), 399-407.

Copes, K., & Rosentswieg, J. (1972). The effects of sleep deprivation upon motor performance of ninth-grade students. *Journal of Sports Medicine and Physical Fitness, 12*(1), 47-53.

Cotton, S., & Richdale, A. (2006). Brief report: Parental descriptions of sleep problems in children with autism, Down syndrome, and Prader-Willi syndrome. *Research in Developmental Disabilities, 27*, 151-161.

Craig, J., & Baron-Cohen, S. (1999). Creativity and imagination in autism and Asperger syndrome. *Journal of Autism and Developmental Disorder, 29*(4), 319-326.

Craig, J., & Baron-Cohen, S. (2000). Story-telling ability in children with autism or Asperger syndrome: a window into the imagination. *Israel Journal of Psychiatry Related Science, 37*(1), 64-70.

Craig, J., Baron-Cohen, S., & Scott, F. (2001). Drawing ability in autism: a window into the imagination. *Israel Journal of Psychiatry Related Science, 38*(3-4), 242-253.

Cramond, B. (1993). The Torrance Test of Creative Thinking:From design through establishment of predictive validity. In R.F. Subotnik, K. D. Arnold (Eds.), *Beyhond Terman: Contemporany Longitudinal studies of giftedness and talent* (pp. 229-254): Nortwood, NJ: Alblex publishing corporation.

Cramond, B. (1994). Attention-Deficit hyperactivity Disorder and Creativity: What is the connection? *Journal of Creative Behavior, 28*, 193-210.

Crick, F., & Mitchison, G. (1983). The function of dream sleep. *Nature, 304*(5922), 111-114.

Crick, F., & Mitchison, G. (1995). REM sleep and neural nets. *Behavioural Brain Research, 69*(1-2), 147-155.

Cropley, A. (2000). Defining and measuring creativity: are creativity tests worth using? *Roeper Review, 23*(2), 72-79.

Cunningham, C., Sloper, T., Rangecroft, A., Knussen, C., Lenning, C., Dixon, I., et al. (1986). *The effect of early intervention on occurrence and nature of behaviour problems in children with Down syndrome*. Abstract Summary of Final Report, Hester Adrian Research Centre, University of Manchester.

Currie, S., Malhotra, S., & Clark, S. (2004). Agreement among objective, subjective, and collateral reports of poor sleep in recovering alcoholics *Behavioral Sleep Medicine,2* (3), 148-161.

Dahl, R. E. (1996). The impact of inadequate sleep on children's daytime cognitive function. *Seminars in Pediatric Neurology, 3*(1), 44-50.

Dahl, R. E., Pelham, W. E., & Wierson, M. (1991). The role of sleep disturbances in attention deficit disorder symptoms: a case study. *Journal of Pediatric Psychology, 16*(2), 229-239.

Daoust, A. M., Limoges, E., Bolduc, C., Mottron, L., & Godbout, R. (2004). EEG spectral analysis of wakefulness and REM sleep in high functioning autistic spectrum disorders. *Clinical Neurophysiology, 115*(6), 1368-1373.

Davis, G. (1997). Identifying creative students and measuring creativity. In N. Colangelo, G. A. Davis (Eds.), *Handbook of gifted education* (pp.269-281). Needham Heights, MA: Allyn and Bacon.

De Bruin, E. A., Beersma, D. G., & Daan, S. (2002). Sustained mental workload does not affect subsequent sleep intensity. *Journal of Sleep Research, 11*(2), 113-121.

De Giacomo, A., Lamanna, A., Martinelli, D., Spadone, M., & Margari, L. (2008). Diagnostic validity of the ASQ in an Italian sample of children affected by pervasive developmental disorders. *Giornale Italiano di Psicopatologia, 14(3)*, 252-257.

Dearing, E., MacCartney, K., Marshall, N. L., & Warner, R. (2001). Parental reports of children's sleep and wakefulness: Longitudinal associations with cognitive and language outcomes. *Infant Behaviour and Development, 24*, 151-170.

Dement, W. (1960). The effect of dream deprivation. *Science, 131(3415)*, 1705-1707.

Dentler, R. A., & Mackler, B. (1964). Originality: Some Social and Personal Determinants. *Behavioral Science, 9(1)*, 1-7.

Didden, R., Curfs, L. M., van Driel, S., & de Moor, J. M. (2002). Sleep problems in children and young adults with developmental disabilities: home-based functional assessment and treatment. *Journal of Behavior Therapy and Experimental Psychiatry, 33*(1), 49-58.

Didden, R., & Sigafoos, J. (2001). A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. *Research in Developmental Disabilities, 22* (4), 255-272.

Dimitriou, E., Kontas, K., & Logothetis, J. (1976). Relationship between parental attitude towards the emotionally disturbed child and nocturnal enuresis. *Behavioural Neuropsychiatry, 8*(1-12), 76-77.

Diomedi, M., Curatolo, P., Scalise, A., Placidi, F., Caretto, F., & Gigli, G. L. (1999). Sleep abnormalities in mentally retarded autistic subjects: Down's syndrome with mental retardation and normal subjects. *Brain & development, 21*(8), 548-553.

Domino, G. (1976). Primary process thinking in dream reports as related to creative achievement *Journal of Consulting and Clinical Psychology 44(6)*, 929-932.

Domino, G. (1982). Attitudes towards dreams, sex differences, and creativity. *Journal of Creative Behavior*, 12, 112-122.

Doo, S., & Wing, Y. K. (2006). Sleep problems of children with pervasive developmental disorders: correlation with parental stress. *Developmental Medicine and Child Neurology, 48*(8), 650-655.

Dornbusch, S. M. (2002). Sleep and adolescence: A social psychologist's perspective. In M. A. Carskadon (Eds.), *Adolescent sleep patterns: Biological, social, and psychological influences* (pp. 1-3). New York, NY: Cambridge University Press.

Duffy, J. F., Rimmer, D. W., & Czeisler, C. A. (2001). Association of intrinsic circadian period with morningness-eveningness, usual wake time, and circadian phase. *Behavioral Neuroscience, 115*(4), 895-899.

Durand, V. (2002). Treating sleep terrors in children with autism. *Journal of Positive Behavior Interventions, 4*(2), 66-72.

Durand, V., Gernert-Dott, P., & Mapstone, E. (1996). Treatment of sleep disorders in children with development disabilities. *Journal of the Association for Persons with Severe Handicaps, 21*(3), 114-122.

Dworak, M., Schierl, T., Bruns, T., & Strüder, H.,K., (2007). Impact of singular excessive computer game and television exposure on sleep patterns and memory performance of school-aged children. *Pediatrics*, 120 (5), 978-985

Eckerberg, B. (2002). Treatment of sleep problems in families with small children: is written information enough? *Acta Paediatrica, 91*(8), 952-959.

Edinger J.,D., Fins A.,I. (1995) The distribution and clinical significance of sleep state misperceptions among insomniacs. *Sleep, 18,* 232–9.

Elia, M., Ferri, R., Musumeci, S. A., Del Gracco, S., Bottitta, M., Scuderi, C., et al. (2000). Sleep in subjects with autistic disorder: a neurophysiological and psychological study. *Brain & Development, 22*(2), 88-92.

Elliot, C. (1983). *British Ability Scales Technical Handbook*: Windsor: NFER-Nelson.

Elliot, C. (1997). *British Ability Scales II Technical Manual* Windsor: NFER-Nelson.

Elliott, P., C. (1986). Right (or left) brain cognition, wrong metaphor for creative behavior: It is prefrontal lobe volition that makes the (human/humane)

difference in the release of creative potential. *The Journal of Creative Behavior, 20* (3), 202-214.
Epstein, R., Chillag, M., & Lavie, P. (1998). Starting times of school: effects on daytime functioning of fifth grade children in Israel. *Sleep, 21* (3), 250-256.
Espie, C. (2002). Insomnia: conceptual issues in the development, persistance and treatment of sleep disorder in adults. *Annual Review of Psycology, 53*, 215-243.
Espie, C., Brooks, D., & Lindsay, W. (1989). An evaluation of tailored psychological treatment of insomnia in terms of statistical and clinical measures of outcome. *Journal of Behavior Therapy and Experimental Psychiatry, 20*(2), 143-153.
Espie, C. A., Inglis, S. J., Harvey, L., & Tessier, S. (2000). Insomniacs' attributions. psychometric properties of the Dysfunctional Beliefs and Attitudes about Sleep Scale and the Sleep Disturbance Questionnaire. *Journal of Psychosomatic Research, 48*(2), 141-148.
Fallone, G., Owens, J. A., & Deane, J. (2002). Sleepiness in children and adolescents: clinical implications. *Sleep Medicine Reviews, 6*(4), 287-306.
Feinberg, I. (1974). Changes in sleep cycle patterns with age. *Journal Psychiatric Research, 10*(3-4), 283-306.
Feldman, D. H. (1999). The development of creativity. In R. J. Sternberg (Ed.), *Handbook of creativity* (pp. 169-186). New York, NY: Cambridge University Press.
Ferber, R. (1985). *Solving your chil's sleep problems*. NY: Simon and Schuster.
Ferrara, M., De Gennaro, L., Casagrande, M., & Bertini, M. (2000). Selective slow-wave sleep deprivation and time-of-night effects on cognitive performance upon awakening. *Psychophysiology, 37*(4), 440-446.
Finke, R. (1990). *Creative imagery: Discoveries and inventions in visualization.* Hillsdale, N.J: Lawrence Erlbaum Associates
Fitzgerald, M. (2004). *Autism and Creativity: is there a link between autism in men and exceptional ability?* New York, NY: Brunner-Routledge.
Flesch, R. (1948). A new readability yardstick. *The Journal of Applied Psychology, 32*, 221-233.
France, K. G., Blampied, N. M., & Wilkinson, P. (1991). Treatment of infant sleep disturbance by trimeprazine in combination with extinction. *Journal of Developmental and Behavioral Pediatrics, 12*(5), 308-314.
Frankel, B. L., Coursey, R. D., Buchbinder, R., & Snyder, F. (1976). Recorded and reported sleep in chronic primary insomnia. *Archives of General Psychiatry, 33*(5), 615-623.
Freitag, C. M. (2007). The genetics of autistic disorders and its clinical relevance: a review of the literature. *Molecular Psychiatry, 12*(1), 2-22.
Frith, U. (1972). Cognitive mechanisms in autism: experiments with colour and tone sequence production. *Journal of Autism Childhood Schizophrenia, 2*(2), 160-173.
Fuller, P., Picciotto, A., Davies, M., & McKenzie, S. (1998). Cough and sleep in inner-city children. *The European Respiratory Journal, 12*, 426-431.
Fuster, J. (1989). *The prefrontal cortex*. New York, NY: Raven Press.
Gaina, A., Sekine, M., Chen, X., Hamanishi, S., & Kagamimori, S. (2004). Validity of child sleep diary questionnaire among junior high school children. *Journal of Epidemiology, 14*(1), 1-4.
Galbraith, L., & Hewitt, K. E. (1993). Behavioural treatment for sleep disturbance. *Health Visit, 66*(5), 169-171.

Gale, A., & Lynn, R. (1972). A developmental study of attention. *British Journal of Educational Psychology, 42*(3), 260-266.

Gaina, A., Sekine, M., Chen, X., Hamanishi, S., & Kagamimori, S. (2004). Validity of child sleep diary questionnaire among junior high school children. *Journal of Epidemiology, 14*(1), 1-4.

Gardner, H. (1982). *Art, mind, and brain; A cognitive approach to creativity*. New York, NY: Basic Books.

Gardner, H. (1993). *Creating minds: An anatomy of creativity seen through the lives of Freud, Einstein, Picasso, Stravinsky, Eliot, Graham and Ghandi*. New York, NY: Harper Collins.

Garretson, H. B., Fein, D., & Waterhouse, L. (1990). Sustained attention in children with autism *Journal of Autism and Developmental Disorders, 20*(1), 101-114.

Gelman, V. S., Jory, M. K., & Macris, P. G. (1998). Pesonality factors in mothers of children who wake at night. *Australian Journal of Psychology, 50*(1), 25-28.

Giannotti, F., Cortesi, F., Cerquiglini, A., & Bernabei, P. (2006). An open-label study of controlled-release melatonin in treatment of sleep disorders in children with autism. *Journal of Autism and Developmental Disorders, 36*(6), 741-752.

Gidman, W. (2006). Under pressure to cope with demand. *The Pharmaceutical Journal, 277*(7414), 218.

Giebenhain, J. E., & O'Dell, S. L. (1984). Evaluation of a parent-training manual for reducing children's fear of the dark. *Journal of Applied Behavior Analysis, 17*(1), 121-125.

Gillberg, M., Anderzen, I., & Akerstedt, T. (1991). Recovery within day-time sleep after slow wave sleep suppression. *Electroencephalogr Clin Neurophysiol, 78*(4), 267-273.

Gilliam, J. E. (1995). *Gilliam Autism Rating Scale* Austin, TX: Pro-ed.

Glaubman, H., Orbach, I., Aviram, O., Frieder, I., Frieman, M., Pelled, O., et al. (1978). REM deprivation and divergent thinking. *Psychophysiology, 15*(1), 75-79.

Godbout, R., Bergeron, C., Limoges, E., Stip, E., & Mottron, L. (2000). A laboratory study of sleep in Asperger's syndrome. *Neuroreport, 11*(1), 127-130.

Goin, R. (1998). Nocturnal enuresis in children. *Child: Care, Health and Development, 24*(4), 277-288.

Goldberg, D. P., Gater, R., Sartorius, N., Ustun, T. B., Piccinelli, M., Gureje, O., et al. (1997). The validity of two versions of the GHQ in the WHO study of mental illness in general health care. *Psychological Meicined, 27*(1), 191-197.

Goldberg, D. P., & Williams, P. (1991). *A User's guide to the General Health Questionnaire*: Windsor: NFER Nelson Publishing Company Ltd.

Goldstein, N. A., Sculerati, N., Walsleben, J. A., Bhatia, N., Friedman, D. M., & Rapoport, D. M. (1994). Clinical diagnosis of pediatric obstructive sleep apnea validated by polysomnography. *Otolaryngology and Head and Neck Surgery, 111*(5), 611-617.

Goodline-Jones, B. L., Sitnick, S. L., Tang, K., Liu, J., & Anders, T. F. (2008). The children sleep habits questionnaire in toddlers and pres-school children *Journal of Development and Behavioral Pediatrics 29*(2), 82-88.

Goodman, R. (1997). The Strengths and Difficulties Questionnaire: a research note. *Journal of Child Psychology and Psychiatry, 38*(5), 581-586.

Gough, H. G. (1976). Studying creativity by means of word association test. *The Journal of Applied Psychology 61*(3), 348-353.

Graziano, A. M., & Mooney, K. C. (1980). Family self-control instruction for children's nighttime fear reduction. *Journal of Consulting and Clinical Psychology, 48*(2), 206-213.

Graziano, A. M., & Mooney, K. C. (1982). Behavioral treatment of "nightfears" in children: maintenance of improvement at 2 1/2- to 3-year follow-up. *Journal of Consulting and Clinical Psychology, 50*(4), 598-599.

Gregory, A. M., Willis, T. A., Wiggs, L., Harvey, G. A., & STEPS. (2008). Pre-sleep Arousal and Sleep Disturbances in Children. *Sleep, 31*(12), 1745-1747.

Gregory, A. M., Eley, T. C., O' Connor, T. C., Rijsdijk, F. V., & Polmin, R. (2005). Family influences on the association between sleep problems and anxiety in a large sample of pre-school aged twins. *Personality and individual differences, 39*, 1137-1148.

Gregory, A. M., & O' Connor, T. G. (2002). Sleep problems in childhood: a longitudinal study of developmental change and association with behavioural problems. *Journal of the American Academy Child Adolescent Psychiatry, 41*(8), 964-972.

Gudmund, J. W. S., & Ingegerd, C. (1983). Creativity and anxiety: An experimental study *Scandinavian Journal of Psychology 24*(1), 107-115.

Guilford, J. P. (1950). Creativity. *American Psychologist, 5*(9), 444-454.

Guilleminault, C., Korobkin, R., & Winkle, R. (1981). A review of 50 children with obstructive sleep apnea syndrome. *Lung, 159*(5), 275-287.

Hall, A., & Nathan, P. (1992). *The managment of night waking and settling problems in young children: efficacy of a behavioural group parent training programme.* Paper presented at the Fourth World Congress of Behaviour Therapy, Queensland, Austalia.

Hall, M., Buysse, D. J., Nowell, P. D., Nofzinger, E. A., Houck, P., Reynolds, C. F., et al. (2000). Symptoms of stress and depression as correlates of sleep in primary insomnia. *Psychosomatic Medicine, 62*, 227-230.

Hankins, M. (2008). The reliability of the twelve-item General Health Questionnaire (GHQ-12) under realistic assumptions. *BCM Public Health, 14*(8),355.

Happe', F. G. E. (1994). Wechsler IQ profile and theory of mind in autism: A research note *Journal of Child Psychology and Psychiatry,35*, 1461-1471.

Harvey, A. G. (2000). Pre-sleep cognitive activity in insomnia: A comparison of sleep-onset insomniacs and good sleepers. *British Journal of Clinical Psychology, 39*(3), 274-286.

Harvey, A. G. (2002). A cognitive model of insomnia. *Behaviour Research and Therapy, 40*(8), 869-893.

Harvey, A. G. (2003). Attempted suppression of pre-sleep cognitive activity in insomnia. *Cognitive Therapy and Research, 27*(66), 2003.

Harvey, A. G., Gregory, A. M., & Bird, C. (2002). The role of cognitive processes in sleep disturbance: A comparison of japane and english university students. *Behavioral and Cognitive Psychotherapy, 30*(3), 259-270.

Harvey, A. G., Mullin, B. C., & Hinshaw, S. P. (2006). Sleep and circadian rhythms in children and adolescents with bipolar disorder. *Development and Psychopathology, 18*(4), 1147-1168.

Hauri, P. J., & Olmstead, E. M. (1989). Reverse first night effect in insomnia. *Sleep, 12*(2), 97-105.

Haythornthwaite, J., Hegel, M., & Kerns, R. (1991). Development of a sleep diary for chronic pain patients. *Journal of Pain and Symptom Management 6*, 65-72.

HBO, (2007). Interview with Alan Berliner. Retrieved May, 13, 2008 from http://www.hbo.com/docs/programs/wideawake/interview.html

Healey, D., & Runco, M. (2006). Could creativity be associated with insomnia? *Creativity Research Journal, 18*(1), 39-43.

Hering, E., Epstein, R., Elroy, S., Iancu, D. R., & Zelnik, N. (1999). Sleep patterns in autistic children. *Journal of Autism and Developmental Disorders, 29*(2), 143-147.

Hiscock, H., & Wake, M. (2001a). Infant sleep problems and postnatal depression: A community based-study. *Pediatrics, 107*(6), 1317-1322.

Hiscock, H., & Wake, M. (2001b). Preventing toddler externalizing behaviour problems: pilot evaluation of a universal parenting program. *International Journal of Mental Health Promotion, 7*(2), 52-58.

Hobson, J., A. (1989). *Sleep*. New York, NY: Scientific America Library.

Hoddes, E., Zarcone, V., Smythe, H., Phillips, R., & Dement, W. C. (1973). Quantification of sleepiness: a new approach. *Psychophysiology, 10*(4), 431-436.

Hoffman, D. C., Sweeney, D. P., Gilliam, J. E., Apocada, D. D., Lopez-Wagner, M. C., & Castillo, M. M. (2005). Sleep problems and symptomology in children with autism. *Focus on autism and other developmental disabilities, 20*(4), 194-200.

Hofman, W. F., & Steenhof, L. (1997). Sleep characteristics of Dutch adolescents are related to school performance. *Sleep Wake Research in the Netherlands, 8*, 51-55.

Honomichl, R. D., Goodlin-Jones, B. L., Burnham, M., Gaylor, E., & Anders, T. F. (2002). Sleep patterns of children with pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 32*(6), 553-561.

Horne, J. A. (1988). Sleep loss and "divergent" thinking ability. *Sleep, 11*(6), 528-536.

Horne, J. A. (1989). *Why we sleep?* Oxford, UK: Oxford University Press.

Horne, J. A. (1992). Human slow wave sleep: a review and appraisal of recent findings, with implications for sleep functions, and psychiatric illness. *Experientia, 48*(10), 941-954.

Horne, J. A. (1993). Human sleep, sleep loss and behaviour. Implications for the prefrontal cortex and psychiatric disorder. *British Journal of Psychiatry, 162*, 413-419.

Horne, J. A., & Minard, A. (1985). Sleep and sleepiness following a behaviourally 'active' day. *Ergonomics, 28*(3), 567-575.

Horne, J. A., Pakhurst, F. L., Reyner, L. A., Hume, K., & Diamond, I. D. (1994). A field study of sleep disturbances: effect of aircraft noise and other factors on 5,742 nights of actimetrically monitored sleep in a large subject sample. *Sleep, 17*(2), 146-159.

Horne, J. A., & Pettitt, A. N. (1985). High incentive effects on vigilance performance during 72 hours of total sleep deprivation. *Acta Psychologica (Amst), 58*(2), 123-139.

Hoshino, Y., Watanabe, H., Yashima, Y., Kaneko, M., & Kumashiro, H. (1984a). An investigation of sleep disturbance of autistic children. *Folia Psychiatrica et Neurologia Japonica, 38*(1), 45-52.

Hoshino, Y., Yamamoto, T., Kaneko, M., Tachibana, R., Watanabe, M., Ono, Y., et al. (1984b). Blood serotonin and free tryptophan concentration in autistic children. *Neuropsychobiology, 11*(1), 22-27.

Howlin, K. (1984). A brief report on elimination of long-term sleeping problems in a 6-year old autistic boy. *Behavioural Psychotherapy, 12*, 257-260.

Hutt, S. J., & Hutt, C. (1968). Stereotypy, arousal and autism. *Human Development, 11*(4), 277-286.

Iber C., Ancoli-Israel S.,Chesson A.L., Quan S.F. (2007). *The AASM Manual for the Scoring of Sleep and Associated Events: Rules, Terminology, and Technical Specifications*. Westchester: American Academy of Sleep Medicine.

Inanuma, K. (1984). Sleep-wake patterns in autistic children. *Japanese Journal of Child and Adolescent Psychiatry, 25*(4), 205-217.

Iranzo, A., Santamaria, J., & de Riquer, M. (2004). Sleep and sleep disorders in Don Quixote. *Sleep Medicine, 5*(1), 97-100.

Ishihara, K. (2002). Relationship between amount of sleep and daytime sleepiness in three cases. *Psychiatry and Clinical Neurosciences, 56*(3), 253-254.

Ishihara, K., & Miyake, S. (1998). A longitudinal study of the development of daytime sleepiness in children. *Psychiatry and Clinical Neurosciences, 52*(2), 178-181.

Jamison, K. (1994). *Touched with fire: maniac- depressive illness and the artistic temperament*. New York: Free Press Paperbacks.

Jan, J. E., Freeman, R. D., & Fast, D. K. (1999). Melatonin treatment of sleep-wake cycle disorders in children and adolescents. *Developmental Medicine and Child Neurology, 41*(7), 491-500.

Jean-Louis, G., Von Gizycki, H., Zizi, F., Spielman, A., Friedman, K., Nunes, J., et al. (1996). Actigraphic correspondence as a measured on the dominant and nondominant wrist. *Sleep Research, 25*, 500.

Jenkins, S., Owen, C., Bax, M., & Hart, H. (1984). Continuities of common behaviour problems in preschool children. *Journal of Child Psychology and Psychiatry, 25*(1), 75-89.

Jenni, O. G., Fuhrer, H. Z., Iglowstein, I., Molinari, L., & Largo, R. H. (2005). A longitudinal study of bed sharing and sleep problems among Swiss children in the first 10 years of life. *Pediatrics, 115*(1), 233-240.

Johns, M. W. (1991). A new method for measuring daytime sleepiness: the Epworth sleepiness scale. *Sleep, 14*(6), 540-545.

Johnson, C. M. (1991). Infant and toddler sleep: a telephone survey of parents in one community. *Journal of Developmental and Behavioral Pediatrics, 12*(2), 108-114.

Johnson, C. M., Brandley-Johnson, S., & Stack, J. (1981). Decreasing the frequency of infants nocturnal crying with the use of scheduled awakenings. *The Family Practice Research Journal, 1*, 98-104.

Johnson, C. R. (1996). Sleep Problems in Children with Mental Retardation and Autism. *Child and adolescent psychiatric clinics of North America, 5*(3), 673-683.

Johnson, L. C. (1973). Are stages of sleep related to waking behavior? *American Scientist, 61*(3), 326-338.

Johnson, L. C., Naitoh, P., Moses, J. M., & Lubin, A. (1974). Interaction of REM deprivation and stage 4 deprivation with total sleep loss: experiment 2. *Psychophysiology, 11*(2), 147-159.

Joseph, R. M., Tager-Flusberg, H., & Lord, C. (2002). Cognitive profiles and social-communicative functioning in children with autism spectrum disorder. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 43*(6), 807-821.

Jouvet, D., Valatx, J. L., & Jouvet, M. (1961). Polygraphic study of sleep in the kitten. *Comptes rendus des séances de la Société de biologie et de ses filiales, 155*, 1660-1664.

Jouvet, M. (1962). Research on the neural structures and responsible mechanisms in different phases of physiological sleep. *Archives Italiennes De Biologie, 100*, 125-206.

Jouvet, M. (1999). Sleep and serotonin: An unfinished story. *Neuropsychopharmacology, 21(2),24-27.*

Kahn, A., Mozin, M. J., Groswosser, J., Sottiaux, M., Dan, B., Scaillet, S., et al. (2001). Sleep disorders and childhood allergy. In L. Wiggs & G. Stores (Eds.), *Sleep disturbance in children and adolescents with disorders of development: Its significance and management* (pp. 137-139). New York, NY: Cambridge University Press.

Kahn, A., Van de Mereckt, C., Rebuffat, E., Mozin, M., J., Sottiaux, M., Blum, D., et al. (1989). Sleep problems in healthy pre-adolescents. *Paediatrics* (84), 542-546.

Kanner, L. (1943). Autistic disturbance of effective contact. *Nervous Child, 2*, 217-250.

Kataria, S., Swanson, M. S., & Trevathan, G. E. (1987). Persistence of sleep disturbances in preschool children. *Journal of Pediatrics, 110*(4), 642-646.

Kavanau, J. L. (1997). Memory, sleep and the evolution of mechanisms of synaptic efficacy maintenance. *Neuroscience, 79*(1), 7-44.

Keen, D., & Ward, S. (2004). Autistic spectrum disorder: A child population profile. *Autism, 8*(1), 39-48.

Keener, M., Zeanah, C., & Anders, T. (1988). Infant temperament ,sleep organization and nightime parental interventions. *Pediatrics, 81(6)*, 762-771.

Kerr, S. M., Jowett, S. A., & Smith, L. N. (1996). Preventing sleep problems in infants: a randomized controlled trial. *Journal of Advanced Nursing, 24*(5), 938-942.

Kim, K. (2006). Can we trust creativity tests? A review of Torrance Test of Creativity Thinking (TTCT). *Creative Research Journal, 18*(1), 3-14.

King, N., Ollendick, T. H., & Tonge, B. J. (1997). Children's nighttime fears. *Clinical Psychology Review, 17*(4), 431-443.

Kingshott, R. N., Cosway, R. J., Deary, I. J., & Douglas, N. J. (2000). The effect of sleep fragmentation on cognitive processing using computerized topographic brain mapping. *Journal of Sleep Research, 9*(4), 353-357.

Kirmil-Gray, K., Eagleston, Gibson, & Thoresen. (1984). Sleep disturbans in adolescents: Sleep quality, sleep habits, beliefs about sleep and daytime functioning. *Journal of Younth and Adolescence, 13*(5), 375-384.

Klackenberg, G. (1982). Sleep behaviour studied longitudinally. Data from 4-16 years on duration, night-awakening and bed-sharing. *Acta Paediatrica Scandinavica, 71*(3), 501-506.

Klimes-Dougan, B., Hastings, P. D., Granger, D. A., Usher, B. A., & Zahn-Waxler, C. (2001). Adrenocortical activity in at-risk and normally developing adolescents: individual differences in salivary cortisol basal levels, diurnal variation, and responses to social challenges. *Development and Psychopathology, 13*(3), 695-719.

Kobayashi, T., Ishikawa, T., & Arakawa, K. (1998). Effects of daytime activity upon the timing of REM sleep periods during a night. *Psychiatry and Clinical Neurosciences, 52*(2), 130-131.

Koegel, R. L., Schreibman, L., Loos, L. M., Dirlich-Wilhelm, H., Dunlap, G., Robbins, F. R., et al. (1992). Consistent stress profiles in mothers of children with autism. *Journal of Autism and Developmental disorders, 22*(2), 205-216.

Kolb, B., & Whishaw, I. (1985). *Fundamentals of human neuropsychology*. San Francisco, CA: Freeman.

Kovacs, M. (1992). *Manual for the Children's Depression Inventory*. North Tonawanda, NJ: Multi-Health Systems.

Krop, H. D., Alegre, C. E., & Williams, C. D. (1969). Effect of induced stress on convergent and divergent thinking. *Psychological Reports, 24*(3), 895-898.

Kuhn, B., & Weidinger, D. (2000). Interventions for infant and toddler sleep disturbance: A review. *Child & Family Behavior Therapy, 22*(2), 33-50.

Laberge, L., Carrier, J., Lesperance, P., Lambert, C., Vitaro, F., Tremblay, R. E., et al. (2000). Sleep and circadian phase characteristics of adolescent and young adult males in a naturalistic summertime condition. *Chronobiology International, 17*(4), 489-501.

Laberge, L., Petit, D., Simard, C., Vitaro, F., Tremblay, R. E., & Montplaisir, J. (2001). Development of sleep patterns in early adolescence. *Journal of Sleep Research, 10*(1), 59-67.

Lam, P., Hiscock, H., & Wake, M. (2003). Outcomes of infant sleep problems: a longitudinal study of sleep, behavior, and maternal well-being. *Pediatrics, 111*(3), e203-207.

Lawton, C., France, K., & Blampied, N. (1991). Treatment of infant sleep disturbance by graduate extinction. *Child & Family Behavior Therapy, 13*(1), 39-56.

Leevers, H. J., & Harris, P. L. (1998). Drawing impossible entities: a measure of the imagination in children with autism, children with learning disabilities, and normal 4-year-olds. *Journal of Child Psychology and Psychiatry, 39*(3), 399-410.

Levitzky, S., & Cooper, R. (2000). Infant colic syndrome--maternal fantasies of aggression and infanticide. *Clinical Pediatrics (Phila), 39*(7), 395-400.

Lewin, I., & Glaubman, H. (1975). The effect of REM deprivation: is it detrimental, beneficial, or neutral? *Psychophysiology, 12*(3), 349-353.

Lewis, V., & Boucher, J. (1991). Skill, content and generative strategies in autistic children's drawings. *British Journal of Developmental Psychology, 9*(3), 393-416.

Lichstein, K. L., & Rosenthal, T. L. (1980). Insomniacs' perceptions of cognitive versus somatic determinants of sleep disturbance. *Journal of Abnormal Psychology, 89*(1), 105-107.

Limoges, E., Mottron, L., Bolduc, C., Berthiaume, C., & Godbout, R. (2005). Atypical sleep architecture and the autism phenotype. *Brain, 128*(5), 1049-1061.

Lincoln, A. J., Courchesne, E., Kilman, B. A., Elmasian, R., & Allen, M. (1988). A study of intellectual abilities in high-functioning people with autism. *Journal of Autism and Developmental Disorders, 18*(4), 505-523.

Lissitz, R., & Willhoft, J. (1985). A methodological study of the Torrance Test of Creativity. *Journal of Educational Measurement, 22*(1), 1-11.

Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: a revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 24*(5), 659-685.

Lotter, V. (1966). Epidemiology of autistic conditions in young children: I prevalence. *Social Psychiatry, 1*(24), 124-137.

Lubin, A., Moses, J. M., Johnson, L. C., & Naitoh, P. (1974). The recuperative effects of REM sleep and stage 4 sleep on human performance after complete sleep loss: experiment I. *Psychophysiology, 11*(2), 133-146.

Manni, R., Ratti, M. T., Marchioni, E., Castelnovo, G., Murelli, R., Sartori, I., et al. (1997). Poor sleep in adolescents: a study of 869 17-year-old Italian secondary school students. *Journal of Sleep Research, 6*(1), 44-49.

Martindale, C. & Hasenfus, N. (1978). EEG differences as a function of creativity, stage of the creative process, and effort to be original. *Biological Psychology, 6*(3), 157-167.

Matty, M., Dahl, R., & Al-Shabbout, M. (1994). Actigraphy as a method to document adolescent sleep patterns. *Journal of Sleep Research, 23*, 452.

McCall, W. V., & Edinger, J. D. (1992). Subjective total insomnia: an example of sleep state misperception. *Sleep, 15*(1), 71-73.

McGarr, R., & Hovel, M. (1980). In search of the sandman: Shaping an infant to sleep. *Education and treatment of children, 3*,173-182.

Meltzer, H., Gatward, R., Goodman, R., & Ford, F. (2000). *Mental health of children and adolescents in Great Britain* London: The Stationery Office

Meltzer, L. J., & Mindell, J. A. (2004). Nonpharmacologic treatments for pediatric sleeplessness. *Pediatric Clinics of North America, 51*(1), 135-151.

Mervis, C., Morris, C. A., Bertrand, J., & Robinson, B. F. (1999). Williams syndrome: Findings from an integrated program of research. In H. Tager-Flusberg (Eds.), *Neurodevelopmental disorders* (pp. 65-110). Cambridge, MA: MIT Press

Milan, M., Mitchell, Z., & Berger, M. (1981). Positive routines: a rapid alternative to extinction for the elimination of the bedtime tantrum behavior. *Child & family Behavior Therapy, 3* (1), 13-24.

Millar, G. (2002). *The Torrance kids at mid-life*: Westport, CT: Ablex.

Milner, B., & Pedrides, M. (1984). Behavioural effects of frontal lobe lesions in man. *Trends in Neurosciences, 7*(11), 403-407.

Minde, K., Faucon, A., & Falkner, S. (1994). Sleep problems in toddlers: effects of treatment on their daytime behavior. *Journal of the American Academy of Child and Adolescent Psychiatry, 33*(8), 1114-1121.

Mindell, J. A. (1993). Sleep disorders in children. *Health Psychology, 12*(2), 151-162.

Mindell, J. A. (1999). Empirically supported treatments in pediatric psychology: bedtime refusal and night wakings in young children. *Journal of Pediatric Psychology, 24*(6), 465-481.

Mindell, J. A., Kuhn, B., Lewin, D. S., Meltzer, L. J., & Sadeh, A. (2006). Behavioral treatment of bedtime problems and night wakings in infants and young children. *Sleep, 29*(10), 1263-1276.

Mindell, J. A., & Owens, J. A. (2003). *A clinical guide to pediatric sleep: diagnosis and managment of sleep problems*. Philadelphia, PA: Lippincott Williams & Wilkins.

Mitchell, P., & Ropart, D. (2004). Visual-Spatial abilities in autism: A review. *Infant and Child Development, 13*(3), 185-198.

Moes, D., Koegel, R. L., Schreibman, L., & Loos, L. M. (1992). Stress profiles for mother and fathers of children with autism. *Psychological Reports, 71*, 1272-1274.

Moffitt, A., Kramer, M., & Hoffman, R. (1993). *The functions of dreaming*. Albany, NY: State University of New York Press.

Moilanen, I., Jarvelin, M., Vikevainen-Tervonen, L., & Huttunen, N. (1987). Personality and family characteristic of enuretic children. *Psychiatric Fennica, 18*, 53-61.

Montgomery, P. (2001). Review: self help, media-based therapy is effective for behavioural problems in children. *Evidence Based Nursing, 5*(1),11.

Montgomery, P., Stores, G., & Wiggs, L. (2004). The relative efficacy of two brief treatments for sleep problems in young learning disabled (mentally retarded) children: a randomised controlled trial. *Archive of Diseases Childhood, 89*(2), 125-130.

Moore, B. A., Friman, P. C., Fruzzetti, A. E., & MacAleese, K. (2007). Brief report: evaluating the Bedtime Pass Program for child resistance to bedtime--a randomized, controlled trial. *Journal of Pediatric Psychology, 32*(3), 283-287.

Moore, M., Meltzer, L., & Mindell, J. (2007). Bedtime problems and night wakings in children. *Sleep Medicine Clinics, 2*(3), 377-385.

Morgenthaler, T., Kramer, M., Alessi, C., Friedman, L., Boehlecke, B., Brown, T., et al. (2006). Practice parameters for the psychological and behavioral treatment of insomnia: an update. An american academy of sleep medicine report. *Sleep, 29*(11), 1415-1419.

Morgenthaler, T. I., Owens, J., Alessi, C., Boehlecke, B., Brown, T. M., Coleman, J., Jr., et al. (2006). Practice parameters for behavioral treatment of bedtime problems and night wakings in infants and young children. *Sleep, 29*(10), 1277-1281.

Morin CM. (1993). Insomnia: psychological assessment and management. New York (NY): Guilford Press.

Morrell, J. (1999). The infant sleep questionnaire: a new tool to assess infant sleep problems for clinical and research purpose. *Journal of Pediatric Psychology, 4*(1), 20-26.

Morrell, J. M. (1999). The role of maternal cognitions in infant sleep problems as assessed by a new instrument, the maternal cognitions about infant sleep questionnaire. *Journal of Child and Psychology and Psychiatry, 40*(2), 247-258.

Morrison, D. N., McGee, R., & Stanton, W. R. (1992). Sleep problems in adolescence. *Journal of America Academy of Child and Adolescent Psychiatry, 31*(1), 94-99.

Murphy, P. W., Chesson, A. L., Walker, L., Arnold, C. L., & Chesson, L. M. (2000). Comparing the effectiveness of video and written material for improving knowledge among sleep disorders clinic patients with limited literacy skills. *The Southern Medical Journal, 93*(3), 297-304.

Muzet, A., Libert, J. P., & Borbely, A. A. (1988). Variation in daily body motility under high ambient temperature in a confined environment with repetitive and monotonous activity. In J. P. Leonard (Eds.), *Methods, models and regulations* (pp. 137-145). Frankfurt: Peter Lang.

Nation, K., Clarke, P., & Snowling, M. J. (2002). General cognitive ability in children with reading comprehension difficulties. *British Journal of Educational Psychology, 72*(4), 549-560.

Nir, I., Meir, D., Zilber, N., Knobler, H., Hadjez, J., & Lerner, Y. (1995). Brief report: circadian melatonin, thyroid-stimulating hormone, prolactin, and cortisol levels in serum of young adults with autism. *Journal of Autism and Developmental Disorders, 25*(6), 641-654.

O'Brien, L. M., Holbrook, C. R., Mervin, C., Klaus, C.,J. Bruner, J. L., Troy, J., et al. (2003). Sleep and Neurobehavioral Characteristics of 5- to 7-Year-Old Children With Parentally Reported Symptoms of Attention-Deficit/Hyperactivity Disorder. *Pediatrics, 111,* (3) 554-536

Ollendick, T. H., Hagopian, L. P., & Huntzinger, R. M. (1991). Cognitive-behavior therapy with nighttime fearful children. *Journal of Behavior Therapy and Experimental Psychiatry, 22*(2), 113-121.

Ophir-Cohen, M., Epstein, R., Tzschinsky, E., Tirosh, E., & Lavie, P. (1993). Sleep patterns of children sleeping in residential care, in kibbutz dormitories and at home- a comparative study. *Sleep, 16* (5), 428-432.

Oskar, G., & Heidi, Z. (2005). A longitudinal study of bed sharing and sleep problems among swiss children the first 10 years of life. *Pediatrics, 115*(1), 233-240.

Ostrosky-solis, F., & Lozano, A. (2006). Digit span: Effect of education and culture. *International Journal of Psychology, 41*(5), 333-341.

Oswald, I. (1980). Sleep as restorative process: human clues. *Progress in Brain Research, 53,* 279-288.

Ott, P. (2007). *Teaching Children with Dyslexia. A practical guide.* Oxford, UK : Routledge Education.

Owens, J., France, K., & Wiggs, L. (1999). Behavioural and cognitive-behavioural interventions for sleep disorders in infants and children: A review. *Sleep Medicine Reviews, 3*(4), 281-302.

Owens, J. A., Spirito, A., & McGuinn, M. (2000). The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. *Sleep, 23*(8), 1043-1051.

Owens, J. A., Spirito, A., McGuinn, M., & Nobile, C. (2000). Sleep habits and sleep disturbance in elementary school-aged children. *Journal of Developmental and Behavioural Pediatrics, 21*(1), 27-36.

Oyane, N. M., & Bjorvatn, B. (2005). Sleep disturbances in adolescents and young adults with autism and Asperger syndrome. *Autism, 9*(1), 83-94.

Pagel, J., & Kwiatkowski, C. (2003). Creativity and Dreaming: Correlation of reported dream incorporation Into waking behavior with level and type of creative interest. *Creative Research Journal, 15*(2-3), 199-205.

Patzold, L. M., Richdale, A. L., & Tonge, B. J. (1998). An investigation into sleep characteristics of children with autism and Asperger's Disorder. *Journal of Paediatrics and Child Health, 34*(6), 528-533.

Penzel, T., & Peter, J. (2007). Ambulatory systems. In M. Billiard (Eds.), *Sleep, physiology, investigations and medicine.* New York, NY: Kluwer Accademic Plenur Publisher.

Piazza, C. C., & Fisher, W. W. (1991). Bedtime fading in the treatment of pediatric insomnia. *Journal of Behavior Therapy and Experimental Psychiatry, 22*(1), 53-56.

Piazza, C. C., Fisher, W. W., & Sherer, M. (1997). Treatment of multiple sleep problems in children with developmental disabilities: faded bedtime with response cost versus bedtime scheduling. *Developmental Medicine and Child Neurology, 39*(6), 414-418.

Piazza, C. C., Hagopian, L. P., Hughes, C. R., & Fisher, W. W. (1998). Using chronotherapy to treat severe sleep problems: a case study. *American Journal of Mental Retardation, 102*(4), 358-366.

Pilcher, J. J., & Huffcutt, A. I. (1996). Effects of sleep deprivation on performance: a meta-analysis. *Sleep, 19*(4), 318-326.

Polimeni, M. A., Richdale, A. L., & Francis, A. J. P. (2005). A survey of sleep problems in autism, Asperger's disorder and typically developing children. *Journal of Intellectual Disability Research, 49*(4), 260-268.

Pollock, J. I. (1994). Night-waking at five years of age: predictors and prognosis. *Journal of Child Psychology and Psychiatry, 35*(4), 699-708.

Prieto, M., Parra, J., Ferrando, M., Ferrandiz, C., Bermejo, M., & Sanchez, C. (2006). Creative ability in early childhood. *Journal of Early Childhood Research, 4*(3), 277-290.

Quine, L. (1991). Sleep problems in children with mental handicap. *Journal of Mental Deficiency Research, 35* (4), 269-290.

Quine, L. (1992). Severity of sleep problems in children with severe learning difficulties: Description and correlates *Journal of Community & Applied Social Psychology, 2*(4), 247-268.

Quine, L. (2001). Sleep problems in primary school children: comparison between mainstream and special school children. *Child Care Health and Development, 27*(3), 201-221.

Quine, L., & Pahl, J. (1985). Examining the causes of stress in families with severely mentally handicapped children. *British Journal of Social Work, 15*(5), 501-517.

Ramchandani, P., Wiggs, L., Webb, V., & Stores, G. (2000). A systematic review of treatments for settling problems and night waking in young children. *British Medical Journal, 320*(7229), 209-213.

Randazzo, A. C., Muehlbach, M. J., Schweitzer, P. K., & Walsh, J. K. (1998). Cognitive function following acute sleep restriction in children ages 10-14. *Sleep, 21*(8), 861-868.

Rapin, I., & Katzman, R. (1998). Neurobiology of autism. *Annals of Neurology, 43*(1), 7-14.

Rapoff, M. A., Christophersen, E. R., & Rapoff, K. E. (1982). The management of common childhood bedtime problems by pediatric nurse practitioners. *Journal of Pediatric Psychology, 7*(2), 179-196.

Rechtschaffen, A., & Kales, A. (1968). *A manual of standardized terminology, techniques and scoring system for sleep stages of human subject*. Washington, DC: US Government Printing Office.

Rechtschaffen, A. (1998). Current perspectives on the function of sleep. *Perspective in Biology and Medicine, 41*(3), 359-390.

Rechtschaffen, A., Gilliland, M. A., Bergmann, B. M., & Winter, J. B. (1983). Physiological correlates of prolonged sleep deprivation in rats. *Science, 221*(4606), 182-184.

Reid, M. J., Walter, A. L., & O'Leary, S. G. (1999). Treatment of young children's bedtime refusal and nighttime wakings: a comparison of "standard" and graduated ignoring procedures. *Journal of Abnormal Child Psychology, 27*(1), 5-16.

Reijneveld, S., Van der wal, M. F., Brugman, E., Hira Sing, R. A., & Verloove Vanhorick, S. P. (2004). Infant crying and abuse. *The lancet, 364*(9442), 1340-1342.

Revonsuo, A. (2000). The reinterpretation of dreams: an evolutionary hypothesis of the function of dreaming. *The Behavioral and brain sciences, 23*(6), 877-901.

Reynolds, C. R., & Richmond, B. O. (1985). *Revised children's manifest anxiety scale manual*. Los Angeles, CA: Western Psychological Services.

Richdale, A. L. (1992). *An investigation of circadian rhythm in chilhood autism.* Melbourne, Australia: La Trobe University.

Richdale, A. L. (1999). Sleep problems in autism: prevalence, cause, and intervention. *Developmental Medicine and Child Neurology, 41*(1), 60-66.

Richdale, A., Francis, A., Gavidia-Payne, S., & Cotton, S. (2000). Stress, behaviour, and sleep problems in children with an intellectual disability. *Journal of Intellectual and Development Disability, 25*(2), 147-161.

Richdale, A. L., & Prior, M. R. (1995). The sleep/wake rhythm in children with autism. *European Child & Adolescent Psychiatry, 4*(3), 175-186.

Richdale, A. L., & Wiggs, L. (2001). Sleep in autism and Asperger's syndrome. In L. Wiggs, Stores G. (Eds.), *Sleep disturbance in children and adolescents with Disorders of development: Its significance and management* (pp. 181-191). London, UK: Mac Keith press, London.

Richdale, A. L., & Wiggs, L. (2005). Behavioural approaches to treatment of sleep problems in children with developmental disorders: What is the state of art? *International Journal of Behavioural and Consultation therapy, 1*(3), 165-189.

Richman, N. (1981). A community survey of characteristics of one- to two- year-olds with sleep disruptions. *American Academy of Child Psychiatry, 20*(2), 281-291.

Richman, N., & Graham, P. J. (1971). A behavioural screening questionnaire for use with 3-year-old children: Preliminary findings. *Journal of Child and Psychology and Psychiatry, 12*(1), 5-33.

Richman, N., Stevenson, J., & Graham, P. (1982). *Preschool to school: A Behavioural Study*. London, UK: Academic Press.

Rickert, V. I., & Johnson, C. M. (1988). Reducing nocturnal awakening and crying episodes in infants and young children: a comparison between scheduled awakenings and systematic ignoring. *Pediatrics, 81*(2), 203-212.

Riedel, B. W., Winfield, C. F., & Lichstein, K. L. (2001). First night effect and reverse first night effect in older adults with primary insomnia: does anxiety play a role? *Sleep Medicine, 2*(2), 125-133.

Riemann, D., Spiegelhalder,K., Feige, B., Voderholzer, U., Berger, M., Perlis, M., et al. (2010). The hyperarousal model of insomnia: A review of the concept and its evidence. *Sleep Medicine Reviews, 14*(1), 19-31.

Ritvo, E. R., Ritvo, R., Yuwiler, A., Brothers, A., Freeman, B. J., & Plotkin, S. (1993). Elevated daytime melatonin concentrations in autism: A pilot study. *European Child & Adolescent Psychiatry,2*(2), 75-78.

Robinson, A. M., & Richdale, A. L. (2004). Sleep problems in children with an intellectual disability: parental perceptions of sleep problems, and views of treatment effectiveness. *Child: Care, Health and Development, 30*(2), 139-150.

Roffwarg, H. P., Muzio, J. N., & Dement, W. C. (1966). Ontogenetic development of the human sleep-dream cycle. *Science, 152*(3722), 604-619.

Runco, M. (1999). A longitudinal study of exceptional giftness and creativity. *Creativity Research Journal, 12*, 161-164.

Runco, M. A., & Sakamoto, S. O. (1999). Experimental studies of creativity. In R. J. Sternberg (Eds.), *Handbook of creativity* (pp. 62-92). New York, NY: Cambridge University Press.

Rutter, M. (2005). Incidence of autism spectrum disorders: changes over time and their meaning. *Acta Paediatrica, 94*(1), 2-15.

Sack, R., Blood, M., Percy, D., & Pen, J. (1995). A comparition of sleep onset latency measured by PSG, actigraphy and beahvioural response. *Sleep Research, 24*, 540.

Sadeh, A. (1990). *Actigraphic home monitoring of sleep-disturbed infants: comparison to controls and assessment of intervention.* Paper presented at the Sleep 1990: Proceedings of the 10th Congress on Sleep Research, Bochum, Germany.

Sadeh, A. (1994). Assessment of intervention for infant night waking: parental reports and activity-based home monitoring. *Journal of Consulting and Clinical Psychology, 62*(1), 63-68.

Sadeh, A. (2004). A brief screening questionnaire for infant sleep problems: validation and finding for an Internet sample. *Pediatrics, 113*(6), 570-577.

Sadeh, A. (2005). Cognitive-behavioral treatment for childhood sleep disorders. *Clinical Psychology Review, 25*(5), 612-628.

Sadeh, A., Alster, J., Urbach, D., & Lavie, P. (1989). Actigraphically based automatic bedtime sleep-wake scoring: validity and clinical application. *Journal of Ambulatory Monitoring, 2*(3), 209-216.

Sadeh, A., Gruber, R., & Raviv, A. (2002). Sleep, neurobehavioral functioning, and behavior problems in school-age children. *Child Development, 73*(2), 405-417.

Sadeh, A., Gruber, R., & Raviv, A. (2003). The effects of sleep restriction and extension on school-age children: what a difference an hour makes. *Child Development, 74*(2), 444-455.

Sadeh, A., Hauri, P. J., Kripke, D. F., & Lavie, P. (1995). The role of actigraphy in the evaluation of sleep disorders. *Sleep, 18*(4), 288-302.

Sadeh, A., Lavie, P., Scher, A., Tirosh, E., & Epstein, R. (1991). Actigraphic home-monitoring sleep-disturbed and control infants and young children: a new method for pediatric assessment of sleep-wake patterns. *Pediatrics, 87*(4), 494-499.

Sadeh, A., Raviv, A., & Gruber, R. (2000). Sleep patterns and sleep disruptions in school-age children. *Developmental Psychology, 36*(3), 291-301.

Sadeh, A. (2008). Commentary: Comparing Actigraphy and Parental Report as Measures of Children's Sleep. *Journal of Pediatric Psychology*, 33(4), 406-407.

Sadock, A. V., & Kaplan, H. I. (2008). *Kaplan and Sadock's Concise Textbook of Clinical Psychiatry.* Philadelphia, PA: Lippincott Williams & Wilkins.

Schreck, K. A., Mulick, J. A., & Rojahn, J. (2003). Development of the Behavioral Evaluation of Disorders of Sleep Scale. *Journal of Child and Family Studies, 12*(3), 349-359.

Schreck, K. A. (2001). Behavioral treatments for sleep problems in autism: Emipirically supported or just universally accepted. *Behavioral Interventions, 16*(4), 265-278.

Schreck, K. A., & Mulick, J. A. (2000). Parental report of sleep problems in children with autism. *Journal of Autism and Developmental Disorders, 30*(2), 127-135.

Schreck, K. A., Mulick, J. A., & Smith, A. F. (2004). Sleep problems as possible predictors of intensified symptoms of autism. *Research in Developmental Disabilities, 25*(1), 57-66.

Scott, F. J., & Baron-Cohen, S. (1996). Imagining real and unreal things: Evidence of a dissociation in Autism. *Journal of Cognitive Neurosciences, 8*(4), 371-382.

Scott, G., & Richards, M. P.M. (1990). Night waking in infants: effects of providing advice and support for parents. *Journal of Child Psychology and Psychiatry, 31*(4), 551-567.

Scott, G., & Richards, M. P. M. (1990). Night waking in 1-year-old children in England. *Child Care Health and Development, 16*(5), 283-302.

Scott, J. (1997). Children as respondents: Methods for improving data quality. In: Lyberg, L.E., Biemer, P.,Collins, M., De Leeuw, E., Dippo, C., Schwarz, N., Treman, D. (Eds). *Survey Measurement and Process Quality* (pp 331-350). NewYork, NY: John Wiley & Sons.

Segawa, M., & Nomura, Y. (1992). Polysomnography in the Rett syndrome. *Brain & Development, 14*, 46-54.

Sejnowski, T. J., & Destexhe, A. (2000). Why do we sleep? *Brain Res, 886*(1-2), 208-223.

Seymour, F. W., Bayfield, G., Brock, P., & During, M. (1983). Management of night waking in young children. *Australian Journal of Family Therapy, 4*(4), 217-222.

Seymour, F. W., Brock, P., During, M., & Poole, G. (1989). Reducing sleep disruptions in young children: evaluation of therapist-guided and written information approaches: a brief report. *Journal of Child Psychology and Psychiatry, 30*(6), 913-918.

Shallice, T. (1984). More functionally isolable subsystems but fewer "modules"? *Cognition, 17*(3), 243-252.

Shallice, T. (2002). Fractionation of the supervisory system. In D. T. Stuss & R. T. Knight (Eds.), *Principles of frontal lobe function* (pp. 261-277). New York, NY: Oxford University Press.

Shapiro, C. M., Bortz, R., Mitchell, D., Bartel, P., & Jooste, P. (1981). Slow-wave sleep: a recovery period after exercise. *Science, 214*(4526), 1253-1254.

Sheldon, S., H, Dekker, A., Levy, H., B. (1991). Sleep patterns in homeless children. *Sleep Research, 20*, 334.

Shneerson, J., K. (2005). *Sleep medicine: A guide to sleep and its disorders 2nd edition.* Oxford, UK: Blackwell Publishing.

Shore, R. (1997). *Rethinking the Brain: New Insights into Early Development.* New York, NY: Families and Work Institute.

Siegel, J. M. (2001). The REM sleep-memory consolidation hypothesis. *Science, 294*(5544), 1058-1063.

Simonds, J., F., & Parraga, H. (1982). Prevalence of sleep disorders and sleep beahviours in children and adolescents. *Journal of the American Academy of Child and Adolescent Psychiatry, 21*(4), 383-388.

Singh, V. K., Singh, E. A., & Warren, R. P. (1997). Hyperserotoninemia and serotonin receptor antibodies in children with autism but not mental retardation. *Biological psychiatry,* 41(6), 753-755.

Sladeczek, I., & Domino, G. (1985). Creativity, sleep and primary process thinking in dreams. *Journal of Creative Behavior, 19*(1), 38-46.

Smedje, H., Broman, J., & Hetta, J. (1998). Sleep disturbances in swedish pre-school children and their parents *Nordic Journal Of Psychiatry, 52*(1), 59-67.

Smedje, H., Broman, J. E., & Hetta, J. (2001). Associations between disturbed sleep and behavioural difficulties in 635 children aged six to eight years: a study based on parents' perceptions. *European Child & Adolescent Psychiatry, 10*(1), 1-9.

Sohlberg, M., & Mateer, C. (1989). *Introduction to cognitive rehabilitation.* New York, NY: The Guilford Press.

Spielman, A., J., Yang, C., & Glovinsky, P. B. (2005). Assessment technique for insomnia. In M. H. Kryger, T. Roth, W. C. Dement (Eds.), *Principles and Practice of Sleep Medicine (1239-1350).* New York, NY: Elsevier/Saunders.

Steenari, M. R., Vuontela, V., Paavonen, E. J., Carlson, S., Fjallberg, M., & Aronen, E. (2003). Working memory and sleep in 6- to 13-year-old schoolchildren. *Journal of the American Academy Child and Adolescent Psychiatry, 42*(1), 85-92.

Stein, M. A., Mendelsohn, J., Obermeyer, W. H., Amromin, J., & Benca, R. (2001). Sleep and behavior problems in school-aged children. *Pediatrics, 107*(4), E60.

Stores, G. (1996). Practitioner review: assessment and treatment of sleep disorders in children and adolescents. *Journal of Child Psychology and Psychiatry, 37*(8), 907-925.

Stores, G. (2001a). Basic aspect of sleep and its disorders. In G. Stores (Eds.), *A clinical guide to sleep disorders in children and adolescents* (pp. 25-54). Cambridge, UK: Cambridge University Press.

Stores, G. (2001b). Sleeplessness. In G. Stores & L. Wiggs (Eds.), *Sleep disturbance in children and adolescents with disorders of development: its significance and management* (pp.24-29). London,UK :Mac Kith Press,

Stores, G., & Wiggs, L. (1998a). Clinical services for sleep disorders. *Archives of Disease in Childhood, 79*(6), 495-497.

Stores, G., & Wiggs, L. (1998b). Abnormal sleep patterns associated with autism. A brief review of research findings, assessment methods and treatment strategies. *Autism, 2*(2), 157-169.

Stores, G., & Wiggs, L. (2001a). Normal sleep including developmental aspects. In G. Stores, Wiggs, L. (Eds.), *Sleep disturbance in children and adolescents with disorders of development:its significance and management* (pp. 10-14). London, UK: Mac Kith Press.

Stores, G., & Wiggs, L. (2001b). Sleep problems and sleep disorders: general. In G. Stores, Wiggs, L. (Eds.), *Sleep disturbance in children and adolescents with disorders of development: its significance and management* (pp. 15-23). London,UK: Mac Kith Press.

Struss, D., & Benson, D. (1986). *The frontal lobes.* New York, NY: Raven Press.

Sverrisson, G., & Bjarnason, H. K. (1990). A survey of sleep habits of Icelandic children. *Icelandic Medical Journal, 76,* 357-361.

Szyndler, J., & Bell, G. (1992). Are groups for parents of children with sleep problems effective? *Health Visit, 65*(8), 277-279.

Taira, M., Takase, M., & Sasaki, H. (1998b). Sleep disorders in children with autism. *Psychiatry and Clinical Neurosciences, 52*(2), 182-183.

Takahashi, M., & Arito, H. (1994). Suppression of electroencephalogram delta power density during non-rapid eye movement sleep as a result of a prolonged cognitive task prior to sleep onset. *European Journal of Applied Physiology and Occupational Physiology, 68*(3), 274-280.

Takase, M., Taira, M., & Sasaki, H. (1998). Sleep-wake rhythm of autistic children. *Psychiatry Clinical Neurosciences, 52*(2), 181-182.

Tazawa, Y., & Okada, K. (2001). Physical signs associated with excessive television-game playing and sleep deprivation. *Pediatrics International, 43*(6), 647-650.

Thackeray, E., & Richdale, A. (2002). The behavioural treatment of sleep difficulties in children with an intellectual disability. *Behavioural Interventions, 17*(4), 211-231.

Thorleifsdottir, B., Bjornsson, J. K., Benediktsdottir, B., Gislason, T., & Kristbjarnarson, H. (2002). Sleep and sleep habits from childhood to young adulthood over a 10-year period. *Journal of Psychosomatic Research, 53*(1), 529-537.

Thunstrom, M. (1999). Severe sleep problems among infants in a normal population in Sweden: prevalence, severity and correlates. *Acta Paediatrica, 88*(12), 1356-1363.

Tizard, B. (1986) The impact of nuclear threat on children's development. In: M. Richards, P. Light, (Eds). *Children of Social Worlds*. Cambridge,UK: Polity press.

Toren, P., Sadeh, M., Wolmer, L., Eldar, S., Koren, S., Weizman, R., et al. (2000). Neurocognitive correlates of anxiety disorders in children: a preliminary report. *Journal of Anxiety Disorders, 14*(3), 239-247.

Torrance, E. (1962). *Guiding creative talent*. Englewood Cliffs, NJ: Prentice Hall.

Torrance, E. (1965). *Rewarding creative behavior*. Englewood Cliffs, NJ: Prentice Hall.

Torrance, E. (1968). A longitudinal examintion of the fourth grade slump in creativity. *Gifted Child Quarterly, 12*(4), 195-199.

Torrance, E. (1974). *Torrance Test of Creative Thinking*. Lexington, MA: Personel Press.

Tynjala, J., Kannas, L., & Valimaa, R. (1993). How young Europeans sleep. *Health Education Research, 8*(1), 69-80.

Van Den Bulk, J. (2004). Television viewing, computer game playing, and Internet use and self-reported time to bed and time out of bed in secondary-school children *Sleep, 27*(101-4).

Van Houten, R., & Rolider, A. (1984). The use of response prevention to eliminate nocturnal thumbsucking. *Journal of Applied Behavior Analysis, 17*(4), 509-520.

Volkmar, F. R., & Cohen, D. J. (1985). The experience of infantile autism: A first-person account by Tony W. *Journal of Autism and Developmental Disorders, 15*(1), 47-54.

Wagner, U., Gais, S., Haider, H., Verleger, R., & Born, J. (2004). Sleep inspires insight. *Nature, 427*(6972), 352-355.

Waldhauser, F., & Steger, H. (1986). Changes in melatonin secretion with age and pubescence. *Journal of Neural Transmission. Supplementum, 21*, 183-197.

Waldman, M., Nicholson, S., Adilov, N. (2006). Does Television Cause Autism? NBER's *Bulletin on Aging and Health, 18.*

Walker, J., & Berger, R., J. (1980). Sleep as an adaptation for energy conservation functionally related to hibernation and shallow torpor. In P. S. McConnell, G. J. Boer, H Romijn, N. E. Van de Poll, M. A. Corner (Eds.), *Adaptive capabilities of the nervous system* (pp. 255-278). Amsterdam: Elsevier.

Walker, P., Liston, C., Hobson, J., & Stickgold, R. (2002). Cognitive flexibility across the sleep-wake cycle: REM-sleep enhancement of anagram problem solving. *Cognitive Brain Research, 14*(3), 317-324.

Ward, T. M., Brandt, P., Archbold, K., Lentz, M., Ringold, S., Wallace, C. A., et al. (2008). Polysomnography and Self-reported Sleep, Pain, Fatigue, and Anxiety in Children with Active and Inactive Juvenile Rheumatoid Arthritis. *Journal of Pediatric Psychology 33*(3), 232-241.

Ware, J. R. (1961). Effects on intelligence on signal detection in visual and auditory monitoring. *Perceptual and motor skills, 3*, 99-102.

Watts, F. N., Coyle, K., & East, M. P. (1994). The contribution of worry to insomnia. *British Journal of Clinical Psychology, 33*, 211-220.

Weiskop, S., Matthews, J., & Richdale, A. (1999). *A parent training program for parents of young children with autism and sleep problems.* Paper presented at the National Autism Conference, Hobart

Weiskop, S., Matthews, J., & Richdale, A. (2001). Treatment of sleep problems in a 5-year-old boy with autism using behavioural principles. *Autism, 5*(2), 209-221.

Weiskop, S., Richdale, A., & Matthews, J. (2005). Behavioural treatment to reduce sleep problems in children with autism or fragile X syndrome. *Developmental Medicine and Child Neurology, 47*(2), 94-104.

Werner H., Molinari, L.,Guyer C., Jenni, O.,G. (2008). Agreement rates between actigraphy, diary, and questionnaire for children's sleep patterns. *Archives Pediatrics & Adolescent Medicine*, 162(4), 350-8.

Weschler, D. (2004). *Wechsler Intelligence Scale for Children, 4th UK edition.* London, UK: Harcourt Assessment.

Wesensten, N. J., Balkin, T. J., & Belenky, G. (1999). Does sleep fragmentation impact recuperation? A review and reanalysis. *Journal of Sleep Research, 8*(4), 237-245.

Weymouth, J., Hudson, A., & King, N. (1987). The management of children's nighttime behaviour problems: Evaluation of an advice booklet. *Behavioral Psychoterapy, 15*(2), 123-133.

WHO (1992). *The tenth revision of the international classification of diseases and related health problems (ICD-10).* Geneva: World Health Organization.

Wiater, A., Mitschke, A., Widdern, S., Fricke, L., Breuer, U., & Lehmkuhl, G. (2005). Sleep disorders and behavioural problems among 8- to 11-year-old children. *Somnologie, 9*, 210-214.

Wiggs, L. (1996). Sleep problems and daytime behaviour in children with severe learning disabilities. *Journal of Intellectual Disability Research, 40* (6), 518-28.

Wing, L. (1998). *The autistic spectrum. A guide for parents and professionals.* London, UK: Constable.

Wiggs, L. (2001). Sleep problems in children with developmental disorders. *The Journal of the Royal Society of Medicine, 94*(4), 177-179.

Wiggs, L. (2007). Are children getting enough sleep? Implications for parents. In *Sociological Research Online 12*(5), retrieved May 10 2008 from http://www.socresonline.org.uk/12/5/13.html>.

Wiggs, L., & France, K. (2000). Behavioural treatments for sleep problems in children and adolescents with physical illness, psychological problems or intellectual disabilities. *Sleep Medicine Reviews, 4*(3), 299-314.

Wing, L., & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: epidemiology and classification. *Journal of Autism and Developmental Disorders, 9*(1), 11-29.

Wiggs, L., Montgomery, P., & Stores, G. (2005). Actigraphic and parent reports of sleep patterns and sleep disorders in children with subtypes of attention-deficit hyperactivity disorder. *Sleep, 28*(11), 1437-1445.

Wiggs, L., & Stores, G. (1995). Children's sleep: how should it be assessed? *ACPP Newletter, 17*(3), 152-157.

Wiggs, L., & Stores, G. (1996a). Severe sleep disturbance and daytime challenging behaviour in children with severe learning disabilities. *Journal of Intellectual Disability Research, 40* (6), 518-528.

Wiggs, L., & Stores, G. (1996b). Sleep problems in children with severe intellectual disabilities: What help is being provided? *Journal of Applied Research in Intellectual Disabilities, 9*(2), 159-164.

Wiggs, L., & Stores, G. (1998a). Behavioural treatment for sleep problems in children with severe learning disabilities and challenging daytime behaviour: effect on sleep patterns of mother and child. *Journal of Sleep Research, 7*(2), 119-126.

Wiggs, L., & Stores, G. (1998b). Factors affecting parental reports of the sleep patterns of children with severe learning disabilities. *British Journal of Health Psychology, 3*(4), 345-359.

Wiggs, L., & Stores, G. (1999). Behavioural treatment for sleep problems in children with severe learning disabilities and challenging daytime behaviour: effect on daytime behaviour. *Journal of Child Psychology and Psychiatry, 40*(4), 627-635.

Wiggs, L., & Stores, G. (2001). Sleeplessness. In L. Wiggs & G. Stores (Eds.), *Sleep disturbance in children and adolescents with disorders of development: Its significance and management* (pp. 137-139). New York, NY: Cambridge University Press.

Wiggs, L., & Stores, G. (2002). Child and Maternal factors associated with sleeplessness in children with autism. *Journal of Sleep Research, 1* (11), 252.

Wiggs, L., & Stores, G. (2004). Sleep patterns and sleep disorders in children with autistic spectrum disorders: insights using parent report and actigraphy. *Developmental Medicine and Child Neurology, 46*(6), 372-380.

Williams, C. D. (1959). The elimination of tantrum behaviour by extinction procedures. *Journal of Abnormal and Social Psychology, 59*, 269.

Williams, P., Sears, L., & Allard, A. (2004). Sleep problems in children with autism. *Journal of Sleep research, 13*(3), 265-268.

Williams, S., Lowe, P., & Griffiths, F. (2007). Embodying and embedding children's sleep: some sociological comments and observations. In *Sociological Research Online* 12(5), retrieved July 2 2008 from http://www.socresonline.org.uk/12/5/6.html.

Wing, L., & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: epidemiology and classification. *Journal of Autism and Developmental Disorders, 9*(1), 11-29.

Wolf, M., Risley, T., & Mees, H. (1964). Applications of Operant Conditioning Procedures to the behavior problem of an autistic child. *Behaviour Research and Therapy, 1*, 305-312.

Wolfson, A., Lacks, P., & Futterman, A. (1992). Effects of parent training on infant sleeping patterns, parents' stress, and perceived parental competence. *Journal of Consulting and Clinical Psychology, 60*(1), 41-48.

Wolfson, A. R., & Carskadon, M. A. (1998). Sleep schedules and daytime functioning in adolescents. *Child Development, 69*(4), 875-887.

Youngstedt, S. D., O'Connor, P. J., & Dishman, R. K. (1997). The effects of acute exercise on sleep: a quantitative synthesis. *Sleep, 20*(3), 203-214.

Zizi, F., Jean-Luois, G., Von Gizycki, H., Spielman, A., Friedman, K., Nunes, J., et al. (1996). Ankle versus wrist acigraph placement: assessment of sleep-wake activity in healthy individuals. *Sleep Research, 25*, 250.

Zuckerman, B., Stevenson, J., & Bailey, V. (1987). Sleep problems in early childhood: continuities, predictive factors, and behavioral correlates. *Pediatrics, 80*(5), 664-671.

Zwi, R., Shawe-Taylor, M., & Murray, J. (2005). Cognitive process in the maintenance of insomnia and co-morbid anxiety. *Behavioral and Cognitive Psychotherapy, 33*(3), 333-342.

APPENDICES

APPENDIX ONE

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER: [redacted]

EMAIL: [redacted]

Dear

My name is Claudia Cipolla and I am a post-graduate psychologist carrying out a doctoral research at Oxford Brookes University in the Department of Psychology.

My research is concerned with examining the relationship between sleep patterns and creative thinking in children and adolescents. Having worked previously as a teacher (and as a support assistant for children with special needs), I have been made aware of how creative and divergent thinking ability may influence many areas of children's functioning. Research with adults has also suggested that sleep patterns at night may influence an individual's ability to think creatively during the daytime and so I am intending to explore this link in children. Because difficulties with sleep are common in children and adolescents, I also intend to find out if giving advice to parents about things they can do to try and help their children to sleep better is useful and improves children's creative thinking.

As you may know, sleep problems and difficulty with creative thinking are even more prevalent amongst children with high functioning autism and Asperger syndrome. I am therefore intending to examine these issues in this group of children too.

I am writing to you to see if your school might be able to help me with this research by distributing information about the project to parents of children attending your school so that families who are interested in taking part could contact me. With your agreement, we would package up individual letters of invitation and information sheets for each family and ask for you to arrange that these are sent home with the children. I would be most grateful for your help. I do appreciate how busy teachers are and would like to reassure you that the research itself would be conducted in the homes of participating families and would not involve any school staff or resources.

We hope that taking part in the project would be fun and interesting for the children and their parents and, for those families where children have sleep problems, that we might be able to help improve the sleep patterns of the children, to the benefit of the child and their family! We could, of course, make the results of the study available to you and the school staff and members.

To give you some more details about the work I am enclosing draft information sheets for your perusal. I hope you will not mind if I telephone your secretary to see if it might be possible to come and see you (or speak to you on the telephone) to explain about the study in more detail and to see if there is a possibility of the school being able to help with the work in the way outlined. Please do not hesitate to contact me if, in the meantime, you have any queries.

Yours sincerely

APPENDIX TWO

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

Date

Dear Parents

My name is Claudia Cipolla and I am a doctoral research student at Oxford Brookes University Department of Psychology. I am investigating links between sleep patterns and creative thinking in children and adolescents and I am writing to you to see if you and your child might be able to help me with this research.

I enclose an information sheet which explains in detail about the study I am conducting. If, after reading the information sheet, you think that you and your child might be interested in helping me to understand more about creative thinking and sleep patterns you can fill in the enclosed brief questionnaire which asks about your child's sleep and behaviour. The questionnaire can be returned to your child's class teacher and I will get in touch with you once I have received your questionnaire.

If you have any questions about anything please do not hesitate to contact me on [redacted]

Thank you very much for taking the time to read through the information sheet. I hope to speak to you soon!

Yours faithfully

Claudia Cipolla

APPENDIX THREE

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

INFORMATION SHEET FOR PARENTS

TITLE OF PROJECT: SLEEP PATTERNS AND CREATIVITY IN CHILDREN AND ADOLESCENTS WITH AND WITHOUT HIGH FUNCTIONING AUTISM: A DESCRIPTIVE STUDY

Name of researchers: Claudia Cipolla and Luci Wiggs

You are being invited to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Do not hesitate to ask me if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for your attention.

What is the purpose of this study?
The purpose of this study is to learn more about links between sleep patterns and creative thinking in children and adolescents, including those with high functioning autism and Asperger's syndrome. There have been a few studies which suggest that, in both adults and children, loss of sleep reduces people's ability to think creatively. Creative thinking is important as it allows us to solve problems, has an impact on performance at school and affects our capacity to be independent of others. Children and adolescents with high functioning autism and Asperger's syndrome often have particular difficulty with creative thinking. Children and adolescents with autism and Asperger's syndrome commonly have difficulty sleeping but so do many typically developing children! We are therefore interested to see if the common types of sleeplessness problems experienced by children and

adolescents (both with and without autism/Asperger's syndrome) have an impact on their creative thinking. If they do, future studies could explore whether improving children's sleep patterns can help them to think more creatively.

To do this we are asking parents of children and adolescents both with and without sleeplessness problems and with and without autism/Asperger's syndrome to help us with our research. We would like to find out more about your child's sleep pattern and their creative thinking skills so that we can see if there is a link between sleep patterns and creativity.

In order to look at links between sleep patterns and creativity we need to include children who DO and DO NOT have any sleep problems. So, if your child does not have any sleep problems it would still be really helpful if he/she could take part in the research.

Why have I been chosen?
Local mainstream schools, the Oxfordshire Education Autism Support Service and parent groups are helping us with this study and we are writing to parents of children and adolescents aged 4 to 16 who are known to them. We hope to find about 80 families in total who will help with this research.

Do I have to take part?
It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason.

What will happen to me if I take part?
We would ask you to fill in the enclosed questionnaire and to return it to your child's class teacher. Claudia would then telephone you to give you more details about the project and to answer any questions. If you have any queries or would like to chat to her before you fill in the questionnaire please telephone her on the number given on the front page of this sheet.

If you decided to take part Claudia would come and visit you twice in your home, at times which are convenient for you.

At the first visit she would ask you about your child's health and sleep pattern, give you a short questionnaire which asks about your child's sleep and behaviour and about how your mood and health has been in general (this takes about 30 minutes to fill-in) and ask you to keep a diary of your child's sleep pattern for one week. In the diary you can note things like the time that you child went to sleep, woke up, if he/she woke up during the night etc. Your child can help you to fill in the diary and, if they would like to, we have a questionnaire for them to complete which asks them to tell us about their sleep.
She would also give you a little monitor to use for about 5 nights which records your child's body movements when he/she is asleep. From looking at the pattern of movement we can tell when your child is sleeping and when he/she is awake. The

monitor looks and is worn like a watch. These monitors have been used a lot with children including children with autism and Asperger's syndrome.

During this visit she would spend some time with your child to assess his/her verbal and spatial abilities using several activities which take the form of games. The session with your child should take about 40/50 minutes so the whole visit should take about an hour.

One week later, she would visit you again to collect the diary, body movement monitor and questionnaire(s) and to assess your child's creative abilities. The creativity assessment involves a series of fun activities including asking your children to draw pictures, tell stories and make objects with different shapes. This creativity assessment would last about 1½ hours and would be audio-recorded.
As well as conducting this study to look at links between sleep and creativity we are also doing some other research to see if giving parents advice about things they can do to try and help their child to sleep better is useful, both in terms of improving children's sleep patterns and also, if this in turn improves children's creativity. If your child sleep is problematic, we might contact you in the future and invite you to take part in this intervention trial.

What are the possible disadvantages and risks of taking part?
The study will not have any economic cost or risk for you or your child. It only requires a bit of your time and enthusiasm.

What are the possible benefits of taking part?
You would be given an evaluation of your child's sleep pattern and creative thinking which may be of interest. Overall we hope that the information we get from this study will further our understanding of the links between creativity and sleep.

Will what I say in this study be kept confidential?
The information collected from you and your child will be kept strictly confidential and your privacy and anonymity will be ensured, subject to legal limitations In accordance with Oxford Brookes University's policy the information will be kept securely, in paper or electronic form, for a period of five years after the completion of my research project after which time it will be destroyed.

What will happen to the results of the research study?
The results of this research will be submitted by Claudia Cipolla as her Doctorate of Philosophy thesis. The results may also be published in scientific journals and presented at relevant scientific meetings for professionals. In presenting the results no individual child or family will be identifiable. At the end of the research we will send a summary of the results to all participating families.

Who is organising and funding the research?
Claudia Cipolla is conducting this study as a research student at Oxford Brookes University, School of Social Science and Law, Department of Psychology with Dr Luci Wiggs as her Director of Studies. The Italian government is funding the research.

Who has reviewed the study?
The research has been approved by the University Research Ethics Committee, Oxford Brookes University.

Contact for Further Information
If you wish to have further information please contact either Claudia Cipolla [redacted] or Dr Luci Wiggs (01865 483710)

If you have any concerns about the way in which the study has been conducted, you can contact the Chair of the University Research Ethics Committee on ethics@brookes.ac.uk.

Many thanks for reading this information sheet.

Date

APPENDIX FIVE

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

Date

Dear Parent

You may remember that I wrote to you a few months ago to tell you about a study we are doing to look at the links between sleep patterns and creative thinking in children and adolescents. There has been some preliminary research which suggests that sleep loss reduces children's ability to think creatively. It is important to find out more about this because if this is the case, in the future we could explore whether improving children's sleep patterns could help them to think more creatively.

In my letter I asked if you might like to help with the study. Since I have not heard back from you I thought that I would write to you again, just in case the original letter or your reply has got lost somewhere along the line! As a reminder, if you did decide to take part in the study it would involve me assessing your child's sleep pattern and coming to see your child, at home, to do some activities with them which would measure their creativity.

If you are interested in finding out more about the study, or taking part, please fill in the enclosed reply slip and return it to your child's class teacher. On this slip you can tell us if we can send you a sleep questionnaire and/or contact you to have a chat about the study and to give you further details and to answer any questions you may have.

It is up to you to decide whether or not to take part. Whatever you decide will in no way affect the future care of you or your child in any way.

In the meantime, please call me on [redacted] if you have any queries or would like to talk about this work
We look forward to hearing from you soon. Thank you very much, in anticipation, for filling in the reply slip. Apologies for troubling you again but it is very helpful for us to hear back from as many families as possible.

Yours faithfully
Claudia Cipolla

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

REPLY SLIP FOR PARENTS

Please tick one of the boxes below:

☐ **We are/might be interested in taking part in the study. Please send us the sleep questionnaire and further details**

Mr/Mrs/Ms...

Our child's name is...

Our address is..

....

....

Our telephone number is ....

☐ **We might be interested in taking part in the study. Please telephone us so that we can have a chat with you**

Mr/Mrs/Ms...

Our telephone number is ....

Thank you very much for completing this reply slip

Please now return this sheet to your child's class teacher

APPENDIX EIGHT

SLEEP DIARY

Child's name ....

Diary completed by ....

If you have any questions please telephone Claudia Cipolla [redacted]
Department of Psychology, Oxford Brookes University, Gipsy Lane, Headington, Oxford OX3 0BP

Date						
Time woke/woken						
Time got up						
Time and length of any daytime naps.						
Time to bed						
Time to sleep						
Any problems going to bed/getting off to sleep.	YES NO	YES NO	YES NO	YES NO	YES NO	YES NO
Time and length of any wakes during the night.						
Anything else of importance (day or night)						

APPENDIX NINE

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

Date

Dear

Thank you for taking part in our study to look at links between creativity and sleep patterns. You mentioned that your child may have a problem with sleeplessness. We are writing to tell you about another study we are conducting to see if we can help children to sleep better and, if we can, whether this improves their creative thinking.

I enclose an information sheet which explains in detail about the study. If, after reading the information sheet, you think that you and your child might be interested in helping us to find out if this treatment is helpful not only to improve children's sleep patterns but also, in turn, their creative thinking, please could you and your child complete the enclosed consent forms and return them to me in the stamped-addressed envelope provided. I will get in touch with you once I have received the forms.

If you have any questions about anything or would like to have a chat with me before you complete the consent forms please do not hesitate to contact me on [redacted]

Thank you very much for taking the time to read through the information sheet. I hope to speak to you soon!

Yours sincerely

Claudia Cipolla

APPENDIX TEN

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

INFORMATION SHEET FOR PARENTS

TITLE OF PROJECT: SLEEP PATTERNS AND CREATIVITY IN CHILDREN WITH AND WITHOUT HIGH FUNCTIONING AUTISM: AN INTERVENTION TRIAL

Name of researchers: Claudia Cipolla and Luci Wiggs

You are being invited to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Do not hesitate to ask me if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for your attention.

What is the purpose of this study?
The purpose of this study is to find out if giving parents advice about things they can do to try and help their child sleep better is helpful, both in terms of improving children's sleep patterns and also, if this in turn, improves children's creativity. We aim to explore this both in children with high functioning autism and Asperger's syndrome and also in typically developing children.

There have been a few studies which suggest that, in both adults and children, loss of sleep reduces people's ability to think creatively. Creative thinking is important as it allows us to solve problems, has an impact on performance at school and affects our capacity to be independent of others. Children with autism and Asperger's syndrome often have particular difficulty with creative thinking. Many children have trouble sleeping and even more children with autism and Asperger's syndrome seem to have difficulty sleeping. We are therefore interested to see if improving children's sleep patterns can help them to think more creatively.

To do this we are asking parents of children who have problems with sleeplessness (both with and without autism/Asperger's syndrome) to help us with our research. We would like to see if one way of helping children to be more creative might be to improve their sleep.

There is a lot of evidence that the most common childhood sleeplessness problems are best treated by 'teaching' children how to sleep better. These types of treatments are called 'behavioural' treatments and do not involve using any medicines. The same behavioural treatments for sleeplessness have been shown to be helpful for children with autism and Asperger's syndrome as well as typically developing children. We have written a booklet for parents which explain these techniques. In research studies this booklet has been shown to be helpful for young children (under 5 years) with sleeplessness problems (including children with autism/Asperger's syndrome) but we don't know if this booklet is helpful for older children. We think the advice, given in a booklet form, will be helpful for reducing sleep problems in older children but we don't know for sure so we are conducting this study to find out. We also want to see if improving children's sleep has a positive effect on children's creative thinking.

Why have I been chosen?
You are taking part in a study we are conducting to examine links between sleep patterns and creativity in children and adolescents. You have told us that your child has a problem with sleeplessness. We think that your child's sleeplessness problem may be helped by behavioural treatment and are inviting you to take part in a study which will investigate this treatment and its effects on creativity. We hope to invite 40 families in total to help with this research.

If at any stage we think that your child's sleeplessness problem is of a type which would not be helped by the treatment which we are using then we would tell you.

Do I have to take part?
It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason.

What will happen to me if I take part?
We would ask you to fill in the enclosed consent form and to return it to Claudia when she calls next week. Claudia will give you more details about the project and answer any questions. If you have any queries or would like to chat to her before you fill in the consent form please telephone her on the number given on the front page of this sheet.

The study we are conducting use a multiple baseline design. This means that because we do not know for sure if the treatment will be a helpful way of treating sleep problems in children of this age group and if this will affect their creative thinking, we need to use a varying time schedule that allows to determinate if change in sleep behaviour corresponded with the introduction of the booklet. Everyone who takes part in the study will be offered the booklet treatment but half of the families will be offered after one week and half will be offered after three weeks. There is a one in two chance of being put into the 'one week group' or 'three weeks group'. Which group your family is in will be decided by a computer, at random, which has no information about the individual – i.e. by chance.

If you decided to take part, with your permission we would write to your child's GP to let him/her know that you are taking part in the study

After six weeks Claudia would come and visit families in both groups in your home, at times which are convenient for you. She would give you a questionnaire which asks for your opinions about the behavioural treatment (this takes about 10 minutes to fill-in) and another which asks about your child's sleep and behaviour and asks about how your mood and health has been in general (this takes about 30 minutes to fill-in) and ask you to keep a diary of your child's sleep pattern for one week or three weeks. In the diary you can note things like the time that you child went to sleep, woke up, if he/she woke up during the night etc. Your child can help you to fill in the diary and, if they would like to, we have a questionnaire for them to complete which asks them to tell us about their sleep.

She would also give you a little monitor to use for about 5 nights which records your child's body movements when he/she is asleep. From looking at the pattern of movement we can tell when your child is sleeping and when he/she is awake. The monitor looks and is worn like a watch. These monitors have been used a lot with children including children with autism and Asperger's syndrome.

During this visit she would spend some time with your child to assess your child's creative abilities. The creativity assessment involves a series of fun activities including asking your children to draw pictures, tell stories and make objects with different shapes. This creativity assessment would last about 1½ hours and would be audio-recorded.

One week or three weeks later, she would visit you again to collect the diary, body movement monitor and questionnaire(s).

To see if the treatment is long-lasting, 2 months later Claudia would send you a brief questionnaire about your child's current sleep pattern and a week-long sleep diary for you to fill in and post back to her. At the end of this families who were allocated to the control group will be given the booklet.

What are the possible disadvantages and risks of taking part?
The study will not have any economic cost or risk for you or your child. It only requires a bit of your time and enthusiasm. Behavioural treatments for sleep problems sometimes involve a brief worsening of any sleep problem before improvements are seen.

What are the possible benefits of taking part?

We hope that the advice in the booklet will help your child's sleep pattern. However, this cannot be guaranteed. We also hope that improvements in your child's sleep pattern will lead to improvements in their creative thinking but again, we are not sure about this and that is why we are doing this study.
Overall we hope that the information we get from this study will further our understanding of the links between creativity and sleep and also help us to know if we can improve creativity and sleep patterns by the methods tested in this study.

Will what I say in this study be kept confidential?
The information collected you and your child will be kept strictly confidential and your privacy and anonymity will be ensured, subject to legal limitations. In accordance with Oxford Brookes University's policy the information will be kept securely, in paper or electronic form, for a period of five years after the completion of my research project after which time it will be destroyed.

What will happen to the results of the research study?
The results of this research will be submitted by Claudia Cipolla as her Doctorate of Philosophy thesis. The results may also be published in scientific journals and presented at relevant scientific meetings for professionals. In presenting the results no individual child or family will be identifiable. At the end of the research we will send a summary of the results to all participating families.

Who is organising and funding the research?
Claudia Cipolla is conducting this study as a research student at Oxford Brookes University, School of Social Science and Law, Department of Psychology with Dr Luci Wiggs as her Director of Studies. The Italian government is funding the research.

Who has reviewed the study?
The research has been approved by the University Research Ethics Committee, Oxford Brookes University.

Contact for Further Information
If you wish to have further information please contact either Claudia Cipolla [redacted] or Dr Luci Wiggs (01865 483710)

If you have any concerns about the way in which the study has been conducted, you can contact the Chair of the University Research Ethics Committee on <u>ethics@brookes.ac.uk</u>.

Many thanks for reading this information sheet.

Date

APPENDIX ELEVEN

OXFORD
BROOKES
UNIVERSITY

DEPARTMENT OF PSYCHOLOGY
OXFORD BROOKES UNIVERSITY
GIPSY LANE
HEADINGTON
OXFORD OX3 0BP

CONTACT NUMBER:
[redacted]

Dear

Re: Name
Address

The parents of the child, named above, have expressed an interest in taking part in a research study investigating the links between sleep patterns and creativity in children with and without high functioning autism/Asperger's syndrome. The study is being carried out by myself and Dr Luci Wiggs, in the Psychology Department at Oxford Brookes University.

The study is a randomised controlled trial of booklet-delivered behavioural treatment for sleeplessness problems in children. Such problems are acknowledged to be common in typically developing children and to occur even more frequently in children with high functioning autism or Asperger's syndrome. There have been a few studies which suggest that, in both adults and children, loss of sleep reduces people's ability to think creatively. We are therefore interested to see if improving children's sleep patterns can help them to think more creatively.

Although behavioural interventions have been seen to be effective for typically and atypically developing children, delivering these interventions by means of a parent-information booklet has only been formally evaluated in younger pre-school aged children. This study hopes to address this omission by including children aged 4-10 years. Assessment of the child's creativity and sleep will be made. This will be done by asking parents (and children where appropriate) to complete questionnaires and asking parents to complete a diary of their child's sleep pattern. The child will wear a small wrist-watch-like device for recording their body movements. From this information we can tell if the child is awake or asleep and we will therefore have objective information, as well as parent report. Only children with sleeplessness and no other sleep-related complaints will be included in the trial.

I enclose a sheet which gives information about the study, including our contact details and also a copy of the intervention booklet which might be of interest to you. We would be happy to discuss the study if you would like further information and, with the parents permission can make any sleep-related assessments available to you if this would be helpful.

Yours sincerely
Claudia Cipolla

APPENDIX EIGHTEEN

Examples of stories:

Story invented by a 10 years old typical child (Danielle)

Real card chosen: C1

D: There were lots of big lions, mummy lion, daddy lion, brother lion, and baby lion. They all lived in Africa. It was very cold and very cloudy and the mummy and daddy went off and brother and baby they ran away.

Then brother and baby found something but the only problem is their mummy and daddy couldn't find them and they thought that they had run away for good. So they followed the grey path and the clouds of white and grey and it was REALLY cold. The mummy and daddy were shivering; it was about to snow. And the grass was turning green and all they could see was frost and snow. And the green grass was covered in snow.

I: and then?

D: Then the baby and brother found a circus and the mummy and daddy they saw them on a high high rollercoaster. They all lived happily ever after.

Story invented by a 10 years old child with autism (Alex)

Real card chosen: C1

A: That is what I think was lions lying on the grass and there's a path that goes between them and between the grass.

I: And? Can you invent a story? What's happened to these lions?

A: They're asleep.

I: And?

A: I don't know. And there are two on each side of the path.

I: Do you want to add something else?

A: No.

I: Okay thank you very much.

Story invented by a 7 years old typical child (Jacob)

Imaginary card chosen: G2

J: There was a man who had an orange head and a small face. He always wore a suit and he was always crying, he was crying because other people didn't like him because he had an orange head. So they were all nasty to him and they started laughing at him and calling him names and he only had one friend. He always went to school but he didn't want to sit near people. One day he went on a trip to the mountains and took a picture, and fell better a bit. The end.

Story invented by a 7 years old child with autism (Jonathan)

Imaginary card chosen: G2

J: Once upon a time there was a castle where some people were thinking of what they could do in the surroundings because at the moment they were surrounded by water which means that no one could get to them but a few people were good at building boats so they all decided that they should bring out guards. So one day they built the finest and most greatest flying saucers that could guard the ship and keep out people. And that's what happened; everyone got knocked under the sea by the saucers. And some of them got so dizzy that they died.

But one didn't. He just...in my opinion...he was already there. And he kept on saying "I CAN do it" and he kept on going in his boat until he reached the door. And he had to be very quiet so that none of the people would know he was there. But one of them did know because even though he wore a potion to them he wasn't *(inaudible)* at the castle.

He had eyes at the back of his head and could see everything and he could see what was going on behind him so he knew that he was there and started up his gang to find out where he was and bring him to him. So he sent out all his messengers and all his spies and they went out all around the castle until they finally found him trying to get the room that kept the castle alive. And all the water… (Jonathan linked this story with the next based on the real card)