CIRCADIAN DISRUPTIONS IN CHILDREN WITH DEVELOPMENTAL DISABILITIES WHO PRESENT FOR THE TREATMENT OF SLEEP PROBLEMS

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Chapter 1
Introduction and Literature

There has been an increasing body of literature that focuses on identifying sleep problems in children with developmental disabilities (DD), with the primary aim of determining the prevalence and describing the sleep problem (Glickman, 2010). Across a number of studies, the most commonly reported sleep problems in children with DD have been irregular sleep-wake patterns that include difficulty maintaining sleep, delayed sleep onset, early morning waking and/or impaired alertness during waking hours (Glickman, 2010). These suggest the involvement of the circadian system (i.e., related to where sleep occurs in the 24h cycle) (Glickman, 2010; Diomedi et al., 1999; Elia et al., 2000; Patzold et al., 1998; Richdale and Prior, 1995; Wiggs and Stores, 2004). For this reason, examining circadian disturbances in children with DD will be vital to a better understanding if it is affiliated with the presenting sleep problems and if circadian interventions will suffice to treat the sleep problems in children with DD (Glickman, 2010). However, the focus of the current study is on manipulating the circadian rhythm (e.g., bedtime fading with/without response cost) to see if it is a sufficiently contributing factor to their sleep problem.

Sleep problems in children with developmental disabilities

Research has shown that children with DD commonly face sleep disturbances (Cortesi, Giannotti, Ivaneko, & Johnson, 2010; Deliens, Leproult, Schmitz, Destrebecqz, & Peigneux, 2015; Moss, Gordon & O'Connell, 2014). These are some of the most frequently reported behaviour problems not only in children with DD, but also in typically developing children (Richdale & Wiggs, 2005). It was noted that 30% of preschool-aged children, who are typically developing, suffer from sleep-related problems (Richman, 1981; Owens, Spirito, McGuinn & Nobile 2000). In contrast, about 80% of children with DD suffer from sleep disorders, and the problem is usually more severe and persistent (Richdale & Wiggs, 2005). An example of this is seen in Hodge, Carollo, Lewin, Hoffman, and Sweeney (2014)'s study on children with autism where they were rated to have significantly more sleep difficulties, poorer sleep quality, and

lesser overall sleep as compared to typically developing children. The prevalence rates of their sleep problems have also been noted to range between 32- 71.5 % (Deliens et al., 2015). These figures are expected, as sleep has been identified as a multifaceted function of the central nervous system, and therefore it is an important component that should not be overlooked (Deliens et al., 2015). The focus of this research examines circadian disruptions (misalignment in one's sleep circadian rhythm) in three children with DD who present for the treatment of sleep problems and determine the efficacy of the use of a circadian intervention to treat the problem.

Children with DD appear to have sleep problems which may differ from the rest of typically developing children (Mann-Dosier, Vaughn, & Fan, 2017). Sleep disturbances in children with DD are often the result of biopsychosocial variables (i.e., differences in melatonin secretion) and behavioural variables (i.e., parental interactions, skills deficits, and antecedents) (Kronk, Bishop, Raspa, Bickel, Mandel, & Bailey, 2010). More research into children with DD has focused on biological aetiologies than behavioural factors. For example, some research has suggested that for individuals with DD, the sleep disturbances might be underpinned by differences in impaired melatonin secretion and sleep architecture which may be associated in causing an interference in the circadian rhythm; however, there is lack of information on that theory and it does not describe any causal links. In addition to that, some DD patients may have co-existing medical conditions which may/may not contribute to their sleep (i.e. delayed sleep onset), but more research would be required to determine this.

Effects of the sleep disturbances on their overall functioning

Many children with DD experience persistent sleep problems, especially with their long-term adverse impacts, which prospectively may further compromise their daytime functioning which is already impaired by their underlying condition (Allik, Larsson & Smedjie, 2006; Bougeron, 2007; Cortesi et al., 2010; Goldman et al., 2012; Kotagal & Broomall, 2012; Moon, Corkum & Smith, 2011; Richdale & Schreck, 2009; Richdale & Wiggs, 2005; Souders et al., 2009; Turner & Johnson, 2013; Vriend, Corkum, Moon & Smith, 2011). The effects of severe sleep disturbances may include cognitive deficits such as impaired memory or attention, behavioural problems, challenging daytime behaviours, impairments in academic functioning,

physical impairments such as stunted growth and immune function and even increased reports of irritability, misconduct, and depressed moods (Richdale & Wiggs, 2005). There has been experimental work conducted on children who were typically developing and who were considered so-called good sleepers, and the results noted were that only a subtle amount of sleep loss (i.e., about 30 minutes) was required, to induce impairments in their neurological functioning. On the other hand, increasing their sleep by about the same amount of time (i.e., about 30 minutes) could lead to improvements (Sadeh, Gruber & Raviv, 2003). The evidence that is presented above thus further substantiates that children require adequate good quality sleep which meets their individual sleep needs, so as to aid in their optimal daily functioning and their overall wellbeing.

Effects on other family members

Research has demonstrated that, in some cases, parents of children with DD and sleep problems may also be at risk in experiencing adverse impacts in their daytime functioning or sleep patterns, and that associations between childhood sleep disorders and parenting difficulties, marital discord, and general family stress has also been found (Kodak & Piazza, 2008; Kotagal & Broomall, 2012; Turner & Johnson, 2013; Vriend et al., 2011). There is some evidence that adverse impacts are reduced when sleep improves (Richdale, 2013; Richdale & Wiggs, 2005; Hoffman et al., 2005; Siversten et al., 2012; Tudor et al., 2012). Hodge et al., (2014) also similarly identified that children with DD needed their sleep problems treated, otherwise they would persist and become more severe with time. This reiterates the importance of addressing the problem effectively because it results in an improvement in the overall functioning of the child and their family (Baker & Richdale, 2017; Richdale & Wiggs, 2005). Thus, identification and intervention of sleep disturbances are recommended as they may aid in improving the daytime functioning of children with developmental disorders and their families. It is also helpful that the evaluation and management of sleep disorders in relation to paediatric developmental disorders is understood, because they may aid in improving the quality of life for this population of children and their families (Dosier, Vaughn & Fan, 2017).

Using behavioural interventions to address sleep problems

Behavioural interventions have been demonstrated to successfully treat sleep problems in typically developing children, including improving the duration and frequency of night wakings (NW) and sleep onset latency (SOL; amount of time taken for complete transition from full wakefulness to sleep) (Honaker & Meltzer, 2014; Meltzer & Mindell, 2014; Vriend et al., 2011). There is some evidence in studies, indicating that using behavioural treatments to address sleep problems in children is more efficacious compared to medication in the both long and short term (Moon, Corkum & Smith, 2011). According to Meltzer and Mindell (2014) there have been some studies conducted surrounding paediatric sleep problems where behavioural interventions were the chosen method of treatment. Some of these behavioural treatments include sleep hygiene, scheduled awakenings, extinction, and parent training. *Sleep hygiene* refers to a list of behaviours and environmental factors as well as other sleep-related conditions that may be adjusted as required for improving sleep (Stepanski & Wyatt, 2003). *Scheduled awakenings* usually involve waking the child approximately about 30 minutes prior to when they are most likely to experience a disruption in their sleep and is usually used to treat patients with parasomnias (Frank, Spirito, Stark & Owens-Stively, 1997). *Extinction* involved removing the reinforcement in order to reduce the behaviour (Weiskop, Richdale & Mathews, 2005). *Parent training* typically involved a therapist giving the parents coaching to become active agents of change to aid in addressing their child's sleep habits, problematic sleep patterns and behaviour or other sleep-related behaviours (Mindell, Kuhn, Lewin, Meltzer & Sadeh, 2006). The efficacy of these behavioural treatment methods on typically developing children was demonstrated within the research (Cortesi et al., 2010; Moss et al., 2014; Schreck, 2001). The extinction method, although being considered as an empirically supported treatment (because this method usually includes the behaviour problems increasing prior to improving) was, however, also demonstrated to be causing high levels of stress for the parents of these children (Mindell, 1999). Thus, this method of treatment was met with disapproval (France & Blampied, 2005).

Possible explanations for children with DD's sleep disturbances

To determine whether a child with DD had impaired sleep quantity or quality, it is important to learn more about their sleep needs and how it was affected or what caused it and this can be achieved via a Functional Behavioural Assessment (FBA). FBA is an assessment that aims to identify a specific target behaviour, the purpose behind the behaviour, and the factors contributing to, or maintaining, the behaviour (Scott, Anderson & Spaulding, 2008). These children could thus be assessed through their sleep-related behaviours, sleep timings, daytime functioning/sleepiness, sleep physiology and possibly manipulating their sleep times (Richdale & Wiggs, 2005). As mentioned, frequently reported sleep problems include increased sleep latencies, NWs, and difficulty waking in the mornings resulting in overall lesser sleep (Patzold et al., 1998; Richdale and Prior, 1995; Wiggs and Stores, 2004; Williams et al., 2004). Such sleep phenomena may suggest that many children with DD may be experiencing circadian disturbances (Glickman, 2010). This has been supported by research which has found abnormal melatonin levels and abnormal cortisol in children with DD. Furthermore, although there have been recent studies using melatonin treatment to correct sleep in these children, based on their impaired circadian rhythms, there is a lack of published reports that thoroughly inspect the alleged association between circadian rhythms and sleep disturbance in children with DD (Glickman, 2010). The association between sleep and melatonin in children with DD have also not been examined much within the framework of the typical development of circadian rhythms. (Glickman, 2010). This present study gathers existing information regarding circadian rhythms and sleep, as a means to aid in analysing the presenting sleep problems in the case studies of three children with DD. It also aims to identify the circadian-related components present among these children. A discussion is presented on the suggested treatment efficacy, by exploring the effects of circadian modifications within the treatment and if there is a need for suggestions for further alternative treatments.

Literature has indicated that sleep interferences do not simply diminish with increasing age and the adverse effects of sleep difficulties have warranted researchers to implement home-based functional assessment and treatment (Didden, Curfs, van Driel, de Moor, 2002). This present study will thus utilise FBA to form a suitable intervention to manipulate the circadian rhythms in children with DD, and sleep problems where problematic sleep scheduling is part of

the presentation. In addition, due to lack of information, more research on circadian-related sleep disorders is also recommended to determine if disruptions in their circadian rhythms might be a component of their sleep problems and if treating that is sufficient to treat sleep problems in children with DD.

Circadian sleep disorders

The misalignment between sleep-wake cycles and the circadian rhythms or the direct impairment of the circadian functioning may cause a disruption in the normal patterns of sleep and circadian rhythms. This is usually hallmarked by the inability to fall asleep during the desired times (Glickman, 2010). This includes complaints of maintaining or initiating sleep, impaired alertness during waking hours or early awakening and these are interestingly the commonly cited problems in children with DD (Patzold et al., 1998; Richdale & Prior, 1995; Wiggs & Stores, 2004; Williams et al., 2004). Thus, examining these circadian disorders may prove helpful in this study as it attempts to identify and treat the circadian components of sleep problems in these children and if necessary, moving on to treat other components.

Sleep and circadian rhythms in children with DD

Sleep is recognised not only as key for neurodevelopmental processes, but also for the maturation of vital brain functions and children with DD who are experiencing impairments in sleep therefore are susceptible to later psychiatric, emotional, behavioural, cognitive, and attentional problems too (Carmassi et al., 2019). An understanding of the relationship between circadian-related sleep disorders and children with DD would also be helpful in forming a suitable treatment to treat their circadian component. There is considerable speculation regarding circadian impairment based on two distinctly separate bodies of knowledge. One body of knowledge refer to the literature affirming the presence of sleep disturbances in children with DD with a small percentage characterising those problems (Richdale, 1999). However, within this existing research, literature surrounding the investigation of circadian regulation specifically

in children with DD is still relatively limited (Glickman, 2010). The following section will attempt to briefly outline and discuss the relevant literature surrounding circadian regulation.

There has been a reported increase in the amount of literature surrounding sleep problems in children with DD, resulting in an increase in the number of relevant studies (Turner & Johnson, 2013; Crowley, Acebo & Carskadon, 2007; Mclay, France, Knight, Blampied & Hastie, 2019; McLay, Roche, France, Lang, France & Busch, 2019; Wiggs & Stores 2004). A study conducted by Wiggs and Stores (2004) investigating the sleep patterns of 69 children with DD revealed some interesting findings. The children were monitored for 5 days and there were sleep disturbances noticed even if they were not reported by parents. Disturbances which were most frequently observed included advanced sleep phase syndrome (ASPS), delayed sleep phase syndrome (DSPS), long sleep latencies, and also NWs, consistent with previous questionnaire reports and sleep diaries (Wiggs & Stores, 2004). These sleep deficits reported to be commonly present in children with DD, correlated with some of the symptoms of the aforementioned circadian sleep disorders, which suggested a possible circadian component. The results showed that 8 children had circadian sleep-wake problems (i.e., disruptions in their sleep circadian) while 36 of them experienced behavioural sleep problems. This study was one of the first studies to carefully and distinctly characterise sleep problems under the standard diagnostic criteria. Prior to this, children whose sleep problems did not meet the standard classification fell under the *Other* category (American Psychiatric Association, 2013; Wiggs & Stores, 2004). Wiggs and Stores also mentioned that although problems such as sleeplessness, which is associated with the absence of certain factors (i.e., television/tablet viewing, parental presence), were mostly viewed as behavioural problems, the associations and conditions which trigger normal sleep should be serving as entraining cues. Thus, it was said that these children could be relying on other cues in their environment to retrieve time of day information and allowance of these behaviours (e.g. parental presence, television viewing) may have negative consequences on their sleep and circadian physiology (Wiggs & Stores, 2004). Furthermore, timings of when children are put to sleep may be too early, coinciding with the period where the circadian drive is the strongest, thus disturbing sleep initiation and important environmental factors such as ensuring their bedrooms are dark with the absences of arousing stimuli along with other good sleep hygiene habits may have been ignored or not addressed (Wiggs & Stores, 2004). In addition to that, it is assumed, that the body's homeostatic pressure and circadian system, is what represents the independent

drives on sleep-wake propensity, but they interact in a non-linear fashion, across the 24-hour light-dark cycle (Schmidt, Peigneux & Cajochen, 2012). Therefore, when the accumulated sleep pressure is particularly high during evening hours, the circadian-based wake propensity is stated to be at its highest (commonly occurs after 12 hours of wakefulness) (Schmidt et al., 2012). In contrast, when the circadian-based sleep propensity is at its maximum during the early hours of the morning, (~2h before the habitual wake time around 6am-7am), the homeostatic sleep pressure is low. Noting this close relationship between the two, it is understood that a misalignment between these two may therefore cause a disruption in an individual's sleep (Schmidt et al., 2012). Didden and Sigafoos (2001) reviewed the nature and treatment of sleep disorders in children with DD. One of the reviewed studies included 214 children with DD and it was reported that, about 86% of children with DD under 6 years old reported sleep problems and that the most commonly reported problems included delayed sleep onset (53%) and difficulty maintaining sleep (53%); these deficits again leaning towards possible disruptions in their circadian rhythms.

Carmassi, Palagini, Caruso, Masci, Nobili, Vita and Dell'Osso (2019) produced a detailed systematic review of sleep disturbances and the desynchronisation of circadian sleep in children with DD and found that children with DD frequently present circadian sleep disturbances such as delayed sleep phases. They also reported that the severity of their sleep problems can be attributed to circadian sleep alterations and sleep disturbances, and therefore there was most likely a definite relationship between circadian disturbances and children with DD. According to them, children with DD have also shown differences in their expression of genes related to delayed sleep phases and irregular sleep-wake cycles. Abnormalities in their expression of genes included genes responsible for melatonin secretion which may also contribute to the circadian disturbances. Because of this, evaluating and treating their circadian disturbances may prove useful in improving or treating sleep problems in children with DD.

Other studies that also studied sleep in children with DD, such as Malow et al.'s (2006) study, which characterised sleep in children with autism, found increased sleep latencies and decreased sleep efficiency as well to again be identified in these children, suggesting a possible circadian disruption (Malow et al., 2006; Limoges et al., 2005).

What are the circadian disorders that are likely to present in children with DD?

Children with DD present with a number of sleep disorders with circadian features. According to the International Classification of Sleep Disorders (ICSD) manual, sleep disorders with circadian features are classified under circadian rhythm sleep-wake disorders (CRSDs) (ICSD, 2014, p. 189). These disorders include delayed sleep-wake phase disorder (DSWPD), advanced sleep-wake phase disorder (ASWPD) and irregular sleep-wake rhythm disorder (IRSWRD). Other types of circadian disorders also exist but they are not described here because they are not relevant to this study. An example of an irrelevant circadian disorder, for the purpose of this study, is non-24h-sleep-wake disorder, also commonly known as free-running sleep-wake rhythm. It is diagnosed mostly in blind people and it rarely presents in children with autism.

The ICSD manual also states that the general criteria that apply across all CRSWDs include: (a) "the child displaying a recurring or chronic pattern of sleep-wake rhythm disruption that is occurring primarily due to misalignment between the required or desired sleep-wake schedule and their endogenous circadian rhythm or an alteration in the endogenous circadian timing system; (b) sleep-wake problems or disturbance such as excessive sleepiness or insomnia symptoms or both and; (c) affiliated impairments or distress in social, mental, physical, educational or other important areas of functioning" (ICSD, 2014, p. 189). Therefore, a child with DD who has the abovementioned impairments, are likely experiencing a circadian rhythm related disorder. The ICSD manual has also specified that it is necessary to consider the presence of dim-light melatonin onset as important in establishing a diagnosis for a circadian rhythm sleep-wake disorder (CRSWD) (ICSD, 2014, p. 189).

Delayed sleep-wake phase disorder (DSWPD)

DSWPD is characterised by a physiological shift in sleep onset to later bedtimes or time of the night and is noted as the most common form of sleep disturbance in children with DD (Moturi & Avis, 2010). More specifically, the manual states that the diagnostic criteria for DSWPD should include "a delay in the phase of the major sleep episode in relation to the desired

or required sleep time and wake-up time, symptoms should present for at least 3 months and the sleep disturbance is not better explained by another current sleep disorder or any other disorder" (ICSD, 2014, pp. 191-197). The biological clock in a circadian rhythm; typically consisting of approximately 24-hours; regulates sleep-wake timings in an individual (van Geijlswijk, Korzilius & Smits, 2010). Melatonin production starts to increase in dim light (also known as dim-light melatonin onset; DLMO), normally between 7pm and 9pm in children (van Geijlswijk, et al., 2010). DWSPD, however, occurs when the circadian clock, still entrained in the 24-hour rhythm, is at a delayed phase angle therefore causing a shift in their sleep-wake timings (van Geijswijk et al., 2010; Lack & Wright, 2007). Data on the general population report patients with DSWPD having difficulty falling asleep before 2am to 6 am and having late wake times typically between 10am to 1pm (Barion & Zee, 2007). Children with DD similarly display difficulties in falling asleep at their age-appropriate time or their desired night bedtimes and to wake spontaneously at desired waketimes, resulting in delayed sleep onset, delayed bedtimes, excessive daytime sleepiness, earlier sleep offset during daytime, chronic sleep deprivation, as well as an overall reduction in total sleep time (Moturi & Avis, 2010). DWSPD is also noted to be more commonly prevalent in children and adolescents, especially children with DD with a prevalence rate of 7% to 16% and precipitating factors that may contribute to the development and maintenance of these delayed sleep patterns may include social maladjustment, school avoidance, and family dysfunction (ICSD, 2014, pp. 191-197). The ICSD manual states that in addition to these mentioned features, in children with DD, their condition may be affiliated with "delays in other markers of the circadian phase, including typical pre-sleep surge in alertness" (ICSD, 2014, pp. 191-197). This may, therefore, present as bedtime resistance because their desired bedtimes are conflicting with their circadian-mediated readiness for sleep. Melatonin onset in children with DD has also been reported to have significant delays as compared to typically developing children as they reportedly have a number of alterations in "melatonin synthesis, levels, amplitude and patterns of secretion" (ICSD, 2014, pp. 191-197).

It is important to distinguish DSWPD from a so-called normal sleep pattern especially in adolescents, who display delayed schedules intermittently or regularly, without impaired functioning or distress (ICSD, 2014, pp. 191-197). Children with DSWPD usually demonstrate improvement in sleep initiation and maintenance when allowed to sleep on their preferred schedule however, if woken up before desired wake time, they may show evidence of excessive

daytime sleepiness or sleep inertia. Unfortunately, research on children with DD in particular is lacking so comparisons are not able to be made.

Advanced sleep-wake phase disorder (ASWPD)

ASWPD is characterised by the involuntary, yet habitual, sleep wake times which are, similar to DSWPD, out of the conventional hours or desired sleep-wake times. Unlike DSWPD, children with ASPD typically present with earlier sleep onset (i.e. 6- 9pm) and wake times (i.e. 2am - 5am) relative to the conventional, age-appropriate sleep times (Barion & Zee, 2007). As a result, these children may display early morning awakenings, early evening, or late afternoon sleepiness, or even the inability to maintain sleep (Barion & Zee, 2007). The symptoms should also persist for "at least three months" and when they sleep according to their preferred schedule, which follows their internal biological clock, improvement is noted in their sleep quality and duration, (following advanced timing of sleep schedules) (ICSD, 2014, pp. 198-203). The other important criteria for ASWPD is to also ensure the sleep disturbance cannot be better explained by another current sleep disorder or any other disorder (ICSD, 2014, pp. 198-203). In comparison to children with DSWPD, however, children with ASWPD tend to undergo less difficulty in adjusting to their earlier schedules. Children who present with ASWPD are, however, rare because most children, including children with DD, usually present with DSWPD (Barion & Zee, 2007). ASWPD is thought to be caused by a number of factors, including unrealistic caregiver expectations of an "appropriate wake time or inappropriately early bedtime for a young child which results in an extended time in bed instead of an advanced sleep onset and offset (ICSD, 2014, pp. 198-203). Careful circadian analysis of a typically developing child indicated that the likely cause of her advanced sleep phase, was due to her short circadian period length of 23.3h (Garcia, Rosen & Mahowald, 2001). It is, also assumed, that a possible reason of the rarely reported cases of young children with ASWPD may be due to the "rarity of observation of advanced sleep onset and offset times among young children due to societal expectations of earlier sleep and wake times for this age group therefore dismissing a possibility of circadian misalignment", or due to the strict ICSD criteria, leading to the "extreme rarity of this condition" (Schrader, Bovim & Sand, 1993; ICSD, 2014, pp. 198-203). ASWPD should, however, be differentiated from so-called normal sleep patterns, especially with very young

children, who usually maintain their advanced schedules without impairment in functioning or distress (ICSD, 2014, pp. 198-203).

Although it is rare, ASWPD has been reported in children with DD. More specifically, children with autism and Smith-Magenis syndrome have demonstrated quite severe alterations in their melatonin secretions profiles, which has resulted in advanced phases of very early morning awakenings, and therefore it cannot be excluded from this study (ICSD, 2014, pp. 198-203).

Irregular sleep-wake rhythm disorder (ISWRD)

ISWRD is mainly characterised by the inability to clearly identify a circadian pattern of sleep-wake times. This disorder has been documented in both typically developing children and children with DD (Barion & Zee, 2007). ISWRD can be distinguished from other circadian sleep wake disorders by the absence or failure to receive prolonged periods of consolidated sleep, irregular distribution of sleep throughout the 24-hour day, as well as the lack of predictability to create a sleep-wake pattern across day-to-day or week-to-week. It has been suggested that the central processes responsible for the generation of circadian rhythms and the decreased exposure to external cues such as light and social activities are both dysfunctional but still key to the development and maintenance of ISWRD. As a result, the sleep and wake times of children effected by ISWRD are irregular and sleep occurs in short bouts throughout the day and night. According to the ICSD manual, diagnostic criteria includes, as mentioned above, “a chronic pattern of irregular sleep and wake episodes throughout the 24-hour period, characterized by insomnia symptoms or excessive daytime sleepiness, or both”, that symptoms should persists for at least three months and that the “sleep disturbance is not better explained by another sleep disorder or another disorder” (2014, pp. 204-208). Children with DD, such as ASD, are themselves at a predisposed risk to developing IRSWRD due to their highly “irregular sleep wake patterns” and abnormalities in their circadian rhythms which are both usually more chronic and severe. Melatonin dysregulation is also known as a potentially causing factor for IRSWD. Along with these are other precipitating factors such as poor sleep hygiene or the lack of exposure to external cues such as light and social activities (ICSD, 2014, pp. 204-208). Apart from autism and Aspergers, children with Angelman syndrome, Williams syndrome, or Smith-

Magenis syndrome are also at risk due to reported decreased concentrations, levels, and amplitude of melatonin secretions in these populations (ICSD, 2014, pp. 204-208). It was also reported that individuals whose light perceptions were impaired (congenitally blind), had a higher incidence rate of developing IRSWD (Okawa, Nanami, Wada, Shimizu, Hishikawa, Sasaki & Takahashi, 1987).

There is, however, a lack of literature pertaining to ISWRD and the pediatric population and therefore a lack of knowledge on the "natural history of the clinical entity of ISWRD" (ICSD, 2014, pp. 204-208). It is also known that poor sleep hygiene should be differentiated from the irregular sleep-wake pattern and the other factors of sleep fragmentation such as other sleep disorders or daytime napping should be identified and treated (ICSD, 2014, pp. 204-208). According to Zee and Vitiello (2009), the significant changes identified in the regulation of the circadian rhythm that occurs with aging and with neurodegenerative diseases (e.g., Alzheimer's disease), are likely to contribute to the prevalence of ISWRD, however ISRWD has also been identified in patients with traumatic brain injury and mental retardation.

Prevalence

The prevalence of CRSDs has been reported by Barion and Zee (2007) in their research studying clinical approaches to CRSDs.

DSWPD. The prevalence of DSPD in the general population has been reported to be between 0.13% and 0.17% but it is more commonly noted in children and adolescents with prevalence data reported to be between 7% and 16%. DSPD represent about 7% of presenting patients in sleep clinics that report sleep problems.

ASWPD. The prevalence of ASWPD on the contrary is reported to be rare as there have only been a few cases reported in literature to date. This limited prevalence data also seems to suggest that prevalence increases with age, with an estimate of about 1% seen in older or middle-aged adults.

ISWRD. Prevalence data were not available because of the lack of literature; however, *ISWRD* was reported to have most common association with brain injury, mental retardation, or dementia patients.

The focus of FBA is on causes of behaviour that can be found within the context in which the behaviour occurs, thus it will be useful in helping develop a suitable intervention. This is investigated in the case studies to establish whether a change in circadian sleep distribution is sufficient to change the sleep problem as a whole. The treatment is analysed to see if it was able to correct their circadian rhythms via adjusting their sleep/wake schedules using circadian-related interventions (i.e., sleep restriction, faded bedtime technique etc).

In this next section there is an exploration within the target population of circadian disturbance, types of circadian-related disorders, and behavioural interventions (both circadian – such as bedtime fading and sleep restriction – and non-circadian, such as – extinction), as well as biological and environmental determinants, including melatonin. At the end of the section is a summary of the findings and also the rationale for the current study.

Determinants of CRSD in children with DD

Circadian rhythm disturbances have complex determinants involving both biological and environmental determinants. These are described below.

Biological determinants

To help understand more about circadian disorders present in children with DD, a brief overview of the biological explanation for the circadian rhythm is warranted. Within us, there is an *endogenous clock,* which refers to our intrinsic biological clock, determining our sleep-wake cycles. This endogenous clock controls the timing of the sleep by synchronising with the external environment – what is regarded the social norm for sleep-wake times and sleep homeostatic processes – the drive to sleep. This complex interaction of the sleep homeostatic processes and endogenous circadian clock is what generates human sleep and wake behaviours (Barion & Zee,

2007). This clock is generated by a central pacemaker, which is commonly known as the suprachiasmatic nucleus (SCN) and is usually synchronised to the external environment (Barion & Zee, 2007). The circadian clock functions to promote wakefulness during the day and aid in the facilitation of sleep consolidation at night. A typical human being with a normal sleep-wake cycle is known to generally demonstrate a biphasic circadian rhythm, which refers to the practice of sleeping during two time periods in a day (24h) therefore, further illustrating the importance of the circadian rhythm as playing a vital role in the distribution and duration of sleep and wakefulness (Barion & Zee, 2007). When the desired sleep time is synchronised with the endogenous circadian rhythm, optimal sleep quality is achieved but when the timing of the circadian rhythm is misaligned between the external environment and the endogenous circadian clock, this results in an impairment of sleep quality, which leads to the development of circadian rhythm sleep disorders (CRSDs) (Barion & Zee, 2007).

CRSDs are described as the inability to utilise cues, such as zeitgebers (time-givers), which signal sleep and have difficulties in adapting to the light-dark cycle (Wasdell, Jan, Bomben, Freeman, Rietveld, Tai, Hamilton & Weiss, 2008). According to Wasdell et al., (2008), children with DD have demonstrated difficulties in understanding or interpreting these abovementioned cues due to their limited developmental capacities, which results in their sleep patterns not following the social norm. According to Carmassi et al., (2019) in 50-80% of children with DD, there have been abnormalities noted in the "maturation of the circadian system" which "principally lead to alterations in their sleep-wake pattern". Their social timing deficits were hypothesised to be related to the variations in the function or structure of their biological clock/clock-related genes therefore putting them at a higher risk of developing circadian sleep dysregulation (Carmassi et al., 2019). This hypothesis has been backed by research indicating that circadian-related gene abnormalities have been detected in children with DD, and that the mutation/s in these genes are responsible for the variations in the circadian system regulation as well as fragmentation of sleep (Carmassi et al., 2019).

Several studies showed that children with DD who presented with circadian sleep disruptions, mostly experienced impairments mainly caused by two specific circadian-related problems: delayed sleep phase periods and irregular sleep-wake patterns of sleep (Carmassi et al., 2019). In these studies, mutations of circadian-related genes were detected in children with

DD who were struggling with sleep problems. Wasdell, Jan, Bomben, Freeman, Rietvald, Tai & Weiss's, 2008, study also backs this theory that children with DD most commonly face delayed sleep onset (Wasdell et al., 2008).

Causes of CRSD in children with DD

Carmassi et al. (2019) concluded that most children with DD have several variations or mutations in their clock genes as well as alterations in their circadian and sleep rhythms, which were reflected as delayed sleep phase or irregular sleep phase periods; however, despite this evidence, there is still a lack of studies that review CRSDs in children with DD (Jan, Bax, Owens, Ispiroglu & Wasdell, 2012).

Melatonin

Melatonin also plays an important role in sleep and circadian rhythm disorders in children with DD. Melatonin is a hormone produced by the pineal gland and is associated with the circadian rhythm. Individuals who are normally entrained to the environmental light-dark cycle have high melatonin secretion at night and low melatonin secretion during the daytime (Arendt, 1995; Tamarkin et al., 1980). Kotagal and Broomall (2012) highlighted in their review of sleep in children with DD, that sleep initiation and maintenance disorders have been shown to have a frequent association with children with DD. Kotagal and Broomall (2012) noted that children with DD are at increased risk of having a stronger association with sleep-onset association disorders and also anxiety falling asleep due to their lower amounts of melatonin levels. Other studies have also revealed that about 65% of the population of children with DD had less than half the average amounts of melatonin as compared to typically developing children (Kotagal & Broomall, 2012). This deficit has been shown to be contributed to by both diurnal and nocturnal melatonin levels, potentially suggesting that there is a global deficit in melatonin production in these children and not just an inverted day-night rhythm or a delayed phase (Bourgeron, 2007; Kulman et al., 2000; Nir et al., 1995; Tordjman et al., 2005, 2012, 2015).

Melatonin has been tested and reported as an efficient method of intervention for sleep disorders in children with DD; however, there has been a lack of randomised-controlled trials done on children with DD so it may still not be regarded as evidence-based (Guénolé et al., 2011). Exogenous melatonin (which helps induce sleep) may be effective in compensating for a lack of endogenous melatonin owing to damage in the SCN. These observations suggest that melatonin is important in regulating the circadian rhythm and sleep (Jan et al., 2012).

Environmental determinants

Behavioural theory of sleep disturbance

Understanding the principles of behaviour may be used to further unpack and inform how sleep disturbance is manifested, how sleep problems are maintained, and also what needs to be focused on to prevent the occurrence of sleep disturbance (Meltzer & McLaughlin Crabtree, 2015).

Sleep onset association can be classified as a form of classical conditioning, whereby falling asleep occurs with an associated event or stimulus that is consistently paired with sleep onset behaviour (Meltzer & McLaughlin Crabtree, 2015). By linking a behaviour with an antecedent stimulus, it may aid in triggering the desired behaviour. An example of this would be to establish a wind-down bedtime routine that will have a consistent end point which the child can learn to associate with sleep onset as well as establishing a consistent appropriate sleep environment (i.e. dark room, comfortable temperature etc.,) that the child is able to familiarise with over time and associate that environment with sleep onset (Wiggs, 2008).

Similarly, children may also begin to rely on the presence of a certain stimuli (e.g., co-sleeping with parents) which have been previously present and continuously paired with sleep, during initial sleep onset as well as during NWs (France, Henderson, & Hudson, 1996). This is where caution is needed because appropriate consistent cues that are present when the child fell asleep but are not present when the child wakes could result in difficulties for the child with

resuming and maintaining sleep (such as the onset of sleep refusal, delayed sleep onset) (France & Blampied, 1993).

Operant conditioning involves using consequences, which may appear in the form of positive or negative reinforcements, to change or modify the form and occurrence of behaviour (Wiggs, 2008). Punishment contingencies are part of the operant conditioning process and is also understood to reduce the likelihood of the behaviour occurring again. Because of this both negative and positive reinforcements may similarly influence the initiation and maintenance of sleep disturbance. Sleep acts as a reinforcer for all of the steps that leads up to sleep onset (France & Blampied, 1999). If, however, their circadian rhythms are disrupted, children and their parents may become susceptible to the coercive behaviour trap where undesired responses contributing to sleep disturbance are reinforced by undesired maintenance of the responses (France & Blampied, 1993). For example, a child who has trouble falling asleep quickly, due to a disruption in the circadian rhythms, might resort to co-sleeping with their parents, or other sleep interfering behaviour, such as screaming or crying, to avoid having to go to bed quickly. The parent/s may respond by maintaining the undesired sleep cue (i.e. co-sleeping with parents again) so as to reduce, avoid or diminish the child's distress and initiate sleep in the short term (France et al., 2003). They enter a vicious cycle, where both parties are negatively reinforced by avoiding the distress and positively reinforced by the onset of sleep. They are most likely to re-engage in the same behaviour pattern repeatedly to avoid distress and initiate sleep, whenever necessary (France et al., 1996). A child with DD may therefore exhibit the abovementioned behaviours if their circadian rhythm is disrupted, which demonstrates the importance of treating circadian sleep disturbances in children with DD.

The importance of sleep treatment in children with DD

Research suggests that circadian rhythm disturbances may have a negative effect on several parts of the brain, which may affect the child's daily functioning (Reghunandanan, V., & Reghunandanan, R., 2006). The severity of neurological impairment, lower levels of cognition as well comorbidities in children with DD has been reported to increase the prevalence of sleep

disturbances, including CRSDs (Jan et al., 2012). Sleep is known to be a complex neurological function and the brain is also reported to play a strong role in influencing or controlling sleep (Jan et al., 2012). Jan et al., (2012) stated, that the malfunctions of the brains of some children with DD, are a major modulator in the development of CRSD, which needs to be taken into consideration. On this basis, the development and prevalence of CRSDs in children with DD, can possibly be attributed to neurological impairment (including impaired perceptions of circadian-related changes in the environment) (Jan et al., 2012). A recent study that studied the patterns of sleep-wake impairment in a group of children with DD, also found these children to be presenting with CRSDs (Jan et al., 2012). The study similarly concluded that brain played a vital role in the regulation of their sleep-wake cycle (Jan et al., 2012).

Importance of sleep

Wang et al. (2011) also reiterated the importance of sleep for procedural memory involves the memory of complex sequences using cognitive and motor skills, as this may also be affected by impaired sleep. This strengthens the theory that 'sleep plays a vital role in the development of language' and also highlights the importance of treatment. Geoffray et al., (2016) similarly reported in their conclusion, a found correlation between sleep circadian disorders and children with DD, including CRSDs having a potential to further exacerbate other DD symptoms. This may therefore hinder the child's neurodevelopment, by preventing the child from learning through social interactions and the environment (Geoffray et al., 2016). Due to this, treating their sleep disturbances may very well treat or lessen impairments in other areas of functioning as well. Early detection is therefore recommended, because the impairment of sleep circadian rhythms can be present in any child, and early identification and treatment have been proven useful in addressing these sleep disturbances (Geoffray el al., 2016).

The above literature has demonstrated the need for an effective behavioural intervention to treat children with DD, who present with circadian components in their sleep disturbances. An intervention becomes necessary, when a desired stimulus (i.e., disrupted circadian rhythm) triggers undesired behaviours, resulting in sleep difficulties such as delayed sleep onset or night awakenings, among other problems. The contributing factors to the child's sleep disturbance (i.e., disrupted sleep circadian rhythm) therefore, should ideally be identified through

behavioural principles, in order to select the most appropriate intervention for the child and his/her family. This is recommended, as every child's chain of events and treatment methods used to reinforce and reinitiate sleep, may be unique for each family (Meltzer & McLaughlin, 2015).

The current literature aims to get a better insight in understanding the different types of circadian interventions that are available and that could be used to treat children with DD experiencing circadian-sleep problems.

Interventions for sleep problems in children with DD

Circadian interventions.

Bedtime fading with or without response cost (FBRC/FBRC-W).

FBRC/FBRC-W has been considered as an efficient circadian intervention due to two biological factors which aid in the function of the fading and response cost procedure. Bedtime fading involves putting the children to bed at a time where they were most likely to achieve rapid sleep onset, and then fading the bedtime gradually over time as the child continued to achieve quick sleep onset (i.e., within 15 minutes) until desired bedtime was achieved. The response cost component in FBRC, refers to removing the child from the bedroom environment if he/she had not initiated sleep within 15 minutes of bedtime, and keeping him awake for a period of time. The child would then be put back to bed, and this procedure would be repeated until the child was able to initiate sleep within 15 minutes of being put to bed (Piazza & Fisher, 1991). The purpose of this intervention is to increase biological sleep pressure, which in turn helps in bringing on more rapid sleep onset for the child (Ortiz & McCormick, 2007). Secondly, setting a consistent sleep and wake up routine may aid in the synchronisation and regularity of the child's circadian rhythm with the desired schedule (Ortiz & McCormick, 2007). This intervention has proved successful within the population of children with DD. There have been but few studies conducted using this intervention method, to treat other populations besides children with DD, although more research on the general population and other populations are still warranted. One study was identified included a population other than children with DD, who were monitored for

their efficacy in reducing sleep problems with the treatment, is Meltzer's study. In her study, Meltzer (2010) found that faded bedtime may be useful in helping treat younger typically developing children (<5 years old) who have significant phase delay or are diagnosed with Behavioural Insomnia of Childhood (BIC; which basically refers to NW and bedtime problems), but there is still a lack of studies in this area and more research is warranted. Another example is the study conducted by Morin, et al. (2006), who also reported that restricting sleep in adults with sleep problems or insomnia produced the best results among other treatments that was tested.

Bedtime fading interventions also known as FBRC, have mostly been the chosen method for treating sleep disturbances in children with DD, due to its demonstrated efficacy for treating sleep problems in this population. One of Piazza and Fisher (1991)'s studies included treating two children, who had profound intellectual disabilities and who were experiencing paediatric insomnia and had not been aided by other treatments. Both children were successfully treated using FBRC and this was demonstrated by an overall reduction in inappropriate daytime sleep and an increase in appropriate consolidated night-time sleep. Piazza and Fisher also used FBRC to treat four children with profound mental retardation including severe behaviour problems (1991b). Similarly, all four children demonstrated success post intervention by a reduction in inappropriate daytime sleep and an increase in appropriate consolidated night-time sleep.

In another study involving children with DD, Piazza, Fisher, and Moser (1991), tested out this intervention with 3 children who suffered both from Rett syndrome and also from severely dysfunctional sleep. Once again, the treatment was met with success and all three girls were able to successfully demonstrate a reduction in inappropriate daytime sleep and an increase in appropriate night-time sleep (Ortiz & McCormick, 2007).

Delemere and Dounavi (2018), also reported successful results from their study of implementing FBRC-W with six children who were diagnosed with autism and demonstrated sleep problems via the Sleep Assessment and Treatment Tool (SATT). They reported an overall decrease in sleep onset latency and an increase in total sleep duration after receiving the bedtime fading intervention.

Similarly, Weisskop, Mathews and Richdale (2001), treated a five-year old boy diagnosed with autism who had sleep problems and the child displayed clinically significant

results from the FBRC-W, including showing consistency during the 3-month and 12-month follow-ups.

Other studies demonstrating successful treatment effects from FBRC-W, included improvement in NWs, which were present in three of the children. This study was, however, replicated by researchers Ashbaugh and Peck (1998), with a typically developing two-year old girl, another example of being extended to a different population. Similarly, treatment effects were successful with this typically developing child as well, demonstrating improvements overall in the child's multiple sleep problems (Ortz & McCormick, 2007).

These studies give a fairly good understanding of the efficacy of this treatment for the population of children with DD. Although these studies do not specifically specify a circadian disturbance, the sleep patterns and presentations of these children indicate a possible circadian component. The results from these studies also further validates this as a possible treatment approach for circadian-related sleep problems in children with DD, as it has shown efficacy in treating these circadian-related sleep presentations.

Another advantage of this intervention is that, according to Ortiz and McCormick (2007), the intervention is fairly easy to implement and also, the child will be likely to feel sleepy and have rapid sleep onset when bedtime is enforced at a later time, therefore, causing little or no effort nor struggles between the parent and the child (2007). Also, the child is not forced to stay in bed if he/she is not yet ready to fall asleep.

Chronotherapy.

Chronotherapy is similar to bedtime fading because it aims to treats the delayed sleep phase in people, who show features of a circadian rhythm disruption as well. Chronotherapy, like bedtime fading, aims to reset the participants' or patients' biological clocks by delaying their sleep timings and it is a popular method which is used to treat people experiencing delayed sleep phases or disruptions in their circadian timings. That means the patient has to progressively stay awake later each night until their sleep period completes a cycle around the clock and achieves a desired nocturnal interval. The difference between these two treatments is that chronotherapy

delays the bedtime by 3-hour successions each day until the desired sleep and wake times are achieved as compared to faded bedtime that starts with setting a later bedtime close to the time the participant is likely to fall asleep quickly and works its way in 15-min successions earlier each day until the desired sleep and wake time is achieved.

Czeisler, Richardson, Coleman, Zimmerman, Moore-Ede, Dement and Weitzman (1981), recruited five adults who were diagnosed Delayed Sleep Phase (DSP) insomnia with the aim of treating those who experienced difficulties in getting to sleep at night and trouble waking up in the morning. They aimed to reset their biological clocks by delaying their sleep phases. All five participants in the study recorded successful results including during follow-ups and were also able to end their chronic dependence on medications.

Chronotherapy is more commonly used to treat adults with DSP insomnia or with severe insomnia who do not have intellectual disabilities, but one study tested out this treatment on a 10-year old girl with DD who was experiencing DSP (Piazza, Hagopian, Hughes & Fisher, 1997). Her bedtime was systematically delayed whilst maintaining a consistent schedule during her wake time. Results demonstrated a success in achieving an age-appropriate bedtime within 11 days of treatment, including a consistent improvement during a 4-month follow-up. This indicated that this treatment may be helpful for the population of children with DD, although more research with this population is still warranted, in that the use of chronotherapy to treat children with DD has hardly been reported in existing literature.

The existing literature, surrounding the use of chronotherapy to treat children or adolescents with DD, is still scarce, and therefore, it remains unclear whether it is an effective treatment for this group. Moreover, in a study conducted by Oren and Wehr (1992), a patient with DSP, who was treated with chronotherapy suffered from adverse effects from the treatment. He was unable to restore his sleep rhythm to a 24-hour sleep schedule and developed hypernyctohemeral syndrome, which is a rare non-24-hour sleep-wake disorder. Oren and Wehr (1992) explained that this is rare disorder is especially debilitating because it does not match with social and professional obligations and therefore causes major disruptions in one's schedule. Oren and Wehr (1992) also mentioned diagnosing two more men with this syndrome, both who developed it after receiving chronotherapy for their DSP. All three patients were noted to experience these adverse effects for at least five years. Oren and Wehr's research (1992)

suggested that this syndrome could be caused by the physiologic after effect of lengthening their sleep-wake cycle during the treatment of chronotherapy and that this lengthening may end up slowing the body's intrinsic rhythm to such an extent that the body is no longer able to reset to the normal 24-hour day entrainment (Oren & Wehr, 1992). In light of the possibility of potential adverse effects of using chronotherapy to treat DSP, including limited literature surrounding chronotherapy and children or adolescents with DD, this may not be an ideal option to treat delayed sleep phases in children with DD. It should, however, also be noted, that this is just one study stating such claims and there are barely any published studies that target children or adolescents, and therefore more research on the potential effects using chronotherapy for children with DD should also be warranted.

Bright light therapy.

One of the aetiologies of DSP disorder (DSPD) suggested is, the sensitivity to evening light or insensitivity to morning light (Gradisar & Crowley, 2013). Gradisar and Crowley (2013) stated in their research that the primary stimulus that synchronises with the circadian rhythm is the daily variations and exposure to the dark and light cycle; there is delay in circadian shift when light is presented in the evening (beginning of sleep) and an advance when light is introduced in the morning (end of sleep). For this reason, Gradisar and his colleagues used bright light therapy to attempt to target the delayed circadian rhythm in a group of youths with DSPD. Bright light therapy consisted of scheduling natural sunlight or bright light at the adolescents' usual wake up time, gradually increasing the exposure to light daily. Results showed that the bright light treatment was effective, and produced long-term benefits; however, more research and consistent results is definitely warranted to make a stronger conclusion on the positive effects of using bright light therapy for children with DSPD.

Cole, Smith, Alcala, Elliott and Kripke (2002) also attempted to treat DSPD using bright light therapy with a group of 600 volunteers between the ages of 18 to 40 years old. The results of their study also showed that bright light therapy had a positive effect on the participants, in reducing their symptoms of DSPD. They concluded that bright light therapy aids in advancing one's circadian rhythm and it might benefit people who are diagnosed with DSPD. Although the efficacy around using this treatment may have also been demonstrated with adults diagnosed

with DSPD, the study did not include the population of children with DSPD. More research using bright light therapy to treat children with DD is needed.

Like other researchers, Gooley (2008) similarly believed that patients with DSPD usually have their sleep-wake pattern misaligned/ out of synchronisation with their circadian rhythm as well as the external environment. He believed the exposure to bright light at the right timing can aid in resetting their sleep wake times which can in turn enhance their overall quality of sleep and daytime alertness. He emphasised the timing of introducing the bright light treatment as that needs to be synchronised with the circadian cycle as well, and, based on field study reports, concluded that the bright light treatment for patients with disrupted circadian rhythms should be administered according to the proper guideline because there is no standard guidelines that govern the application of bright light treatment for DSPD at the moment (Gooley, 2008). This noted, studies (Gooley, 2008) using bright light treatment to treat participants with circadian rhythm sleep disorders (CRSD) have reported successful results in reducing or treating the problem and therefore, Gooley (2008) concluded that it is an effective treatment for the population of people experiencing CRSD or DSPD. Despite bright light therapy being an empirically supported treatment for DSPD, there has been a lack of literature surrounding the use of bright light treatment for children with DD who are experiencing DSPD. Most of the reported studies that used bright light to treat participants with DSPD, involved adults, and therefore there is insufficient evidence to conclude that it may be a suitable treatment for children with DD at this stage and further research would definitely be needed.

Sleep restriction.

Sleep restriction is designed to interrupt or to break the associations between sleep problems and the bedroom by restricting the time the patient spends in bed while awake (Kloss, Nash, Horsey & Taylor, 2011). Sleep restriction therapy may be a possible choice for intervention because it is said to aid in strengthening or improving the circadian sleep drive that may have become dysregulated with irregular sleep-wake schedules in that it helps initiate a regular sleep and wake cycle (Kloss, et al., 2011). Sleep restriction has been cited as one of the most effective interventions for sleep problems within the population of children (Kloss et al., 2011).

Christodulu and Durand (2004) investigated the effect of sleep restriction on NW in children with DD. The aim was to eliminate these problems without causing increasing disruption before or during bedtime. Once the participants had demonstrated improvement, their bedtimes were gradually faded back to their age-appropriate bedtimes. The combined effects of sleep restriction and bedtime fading successfully eliminated their sleep problems. Christodulu and Durand (2004) also reported similar treatment efficacy in being able to eliminate these sleep problems in children with DD, suggesting that sleep restriction alone, or with bedtime fading, may be a potential treatment option for addressing circadian disturbances within the population of children with DD.

Spielman, Saskin, and Thorpy (1987) attempted to treat 35 patients' chronic insomnia with sleep restriction therapy. There has been recognition of different causes of insomnia, one of them being due to DSPD. The patients showed improvements in their total overall sleep times, sleep latency, sleep efficiency and also total wake time. They concluded sleep restriction therapy was effective to treat insomnia. The population here, however, consisted of adults with a history of sleep problems not children therefore further research should be warranted within the population of children with DD.

Gruber, Wiebe, Montecalvo, Brunetti, Amsel, and Carrier (2011) conducted a research studying the impact of sleep restriction on 43 children between 7 and 11 years old with attention deficit hyperactivity disorder (ADHD). They restricted the sleep of these children for an hour nightly, over the course of 6 nights and found that these children demonstrated an improvement in shorter sleep latency and higher sleep efficiency, including an overall improvement in the quality of sleep. Although this study does include children, the population of children with DD were intentionally excluded from this study, further research regarding using sleep restriction therapy would be beneficial to the population of children with DD.

Christodulu and Durand (2004) also conducted a study to investigate the effects behavioural interventions such as positive bedtime routine and sleep restriction on eliminating or decreasing sleep problems in four children with DD. The results of sleep restriction coupled with positive bedtime routine were successful in reducing or eliminating sleep problems and supported the use of these behavioural interventions in these children with DD. The intervention also resulted in an improvement in overall parental satisfaction in getting their children to sleep.

There was, however, a return to baseline for one of the participants after the intervention when the family went on a vacation, but they were able to address her sleep disturbances with the reintroduction of the intervention and were able to reduce her sleep disturbances once again. Sleep restriction, while being an effective method of behavioural intervention in reducing sleep disturbances in children with DD, may not be sustainable in some cases (e.g., due to difficulty for parents in some cases to restrict their child from sleeping) and also the sleep restriction therapy was coupled with positive bedtime routine which may have aided the intervention. There should be continued future research separately studying the efficacy of sleep restriction therapy in reducing sleep problems in children with DD as this may be a promising option for children with DD experiencing sleep disturbances as currently, there is a lack of relevant literature in this population.

Non-circadian interventions.

Standard extinction.

Standard extinction, is one of the behavioural interventions used to treat children with sleep disturbances by consistently removing the reinforcement for undesirable sleep behaviours (parents ignoring all bedtime disruptions, i.e., putting the child to bed after which there is no interaction until morning) (Weiskop, Matthews & Richdale, 2001). There has, however, been concern regarding the implementation of this intervention for parents, due to difficulties in ignoring their child for extended periods of time. There is also limited research surrounding using standard extinction to treat children with DD who have DSP or circadian sleep disturbances.

One of few the studies that treated children with DD experiencing sleep problems and using the standard extinction procedure is by Weiskop, Richdale, and Mathews (2005). They investigated the efficacy of using standard extinction to eliminate or decrease sleep problems in children with DD but there was no comment if they were experiencing circadian sleep disturbances. During the intervention, the researchers combined positive bedtime routines and reinforcements with the extinction procedure. Efficacy was achieved from the programme but

the limited amount of available literature surrounding extinction with children with DD with circadian sleep difficulties still requires more research and for this reason the efficacy of this treatment is still an open question. Furthermore, there were only ten children in the study, who underwent the intervention and therefore a larger number of participants in this population should be recruited to test for consistency in efficacy of the programme.

Didden, Curfs, Driel and Moor (2001) also treated children with DD, using the standard extinction procedure. They recruited four children who were experiencing disruptive night-time behaviours such as problems in settling to sleep, having NW episodes and early morning waking. Although none were diagnosed with DSP, they demonstrated components of circadian disruption (e.g., sleep onset delay, early awakenings,). The results post intervention indicated a success in effectively reducing these sleep problems faced by these children. In contrast, parents of these participants were reported to have found it challenging to implement this treatment due to fear of causing psychological trauma to their child or affecting the parent-child relationship. Moreover, although this study demonstrated effectiveness in addressing the sleep disturbances within the population of children with DD, the other question of whether the same efficacy in addressing children with DD facing circadian sleep disturbances still remains and therefore future research would still be warranted to determine this.

Standard extinction was also used to treat children with intellectual disabilities (ID) who were experiencing sleep difficulties and who were experiencing daytime behaviours as well (Thackeray & Richdale, 2002). Improvements in their overall sleep quality were demonstrated through the intervention however their daytime behaviours had little or no effect from the intervention. They reported standard extinction as being a quick and effective intervention to ameliorate sleep disturbances in children with ID. Standard extinction has been demonstrated in several studies to be effective in ameliorating sleep difficulties in children but there still needs to be more literature on ameliorating circadian disturbances in children.

Modified/Gradual extinction.

Gradual extinction is an alternative form of extinction and involves ignoring the child as in standard extinction, but for a limited amount of time before checking in and providing limited

attention and brief reassurance. The intervals between check will also increase either during the night or day by day (i.e., 3 minutes, then 5 minutes, then 10 minutes). As with standard extinction there is also a limited amount of research available investigating the treatment of circadian disturbances in children with DD using graduated extinction. Using graduated extinction for circadian disturbances in children with DD might be successful, due to the *planned ignoring* or *systematic ignoring* that still provides the child with limited parental attention but is increasing their independence to fall asleep. This, in turn, will likely decrease the frequency of sleep interfering behaviours (e.g., bid for parental attention) thereby helping the child to achieve quick sleep onset. The anticipated result of using extinction to address problem behaviours such as sleep onset delay, bedtime resistance, and NW, is that they will be eliminated (Vriend et al., 2011). Gradisar, Jackson, Spurrier, Gibson, Whitham, Williams, Dolby and Kennaway (2016), conducted a randomised controlled trial to evaluate the effects of graduated extinction as well as bedtime fading in infants who were experiencing sleep disturbances. The study results found improvements in sleep latency and sleep onset as well as a reduction in NWs in both intervention groups. They concluded that both behaviour interventions produced significant sleep benefits without affecting parent-child attachment or child emotions and behaviour (these were assessed as well) neither any adverse stress responses. Although this study was successful, the inclusion criteria for the study were determined by parental reports of the child having sleep problems and there was no indication of investigation whether there were circadian disturbances present in their sleep disturbances, and therefore, further research using graduated extinction would be warranted to determine its efficacy with children with DD experiencing circadian sleep disturbances.

Durand and Mindell (1990) investigated the efficacy of graduated extinction in a 14-month typically developing child who had difficulties falling asleep and woke frequently during the night. This involved her parents putting her to bed (she needed parental presence to fall asleep) and wait successively for longer bouts of time (2 minutes, 3 minutes, 4 minutes), before going in just to provide brief reassurance if she was displaying sleep interfering behaviours (tantrums, yelling, etc). Her NWs were addressed in the same manner, with her parents entering the room for brief period (15-30 seconds) when she woke to provide reassurance before leaving again. They would then wait progressively for longer periods of time (five-minute increments) before reentering her room to provide brief and neutral reassurances again. The idea was for the

child to learn to fall asleep independently by reducing the amount of parental attention she received. In about 50 weeks, they were able to successfully treat her sleep problems despite a slight regression during week 48-49 (when the child stayed with her grandmother). Because the study only involved one family, that too a typically developing infant, there is insufficient criteria met to conclude this as an efficient treatment for children with DD.

Another study involving the use of graduated extinction to address sleep disturbances in children with DD, is a study conducted by Knight and Johnson (2014). In this study, graduated extinction was used along with other treatment options, such as positive bedtime routines, white noise, and chronotherapy, and a behavioural treatment package to treat three children with DD who were experiencing sleep disturbances. All three participants indicated improvements in decreasing sleep latency as well as the number of NWs. The researchers noted that the disturbances were not completely eliminated. Also, the sample size of the study was relatively small and other behavioural problems were not addressed. The study, while targeting sleep latency as one of its target behaviours, did not really address other aspects of DSP and therefore, without further research, this treatment package may not yet be identified as a possible ideal treatment to ameliorate circadian sleep disturbances in children with DD.

Based on previous literature, it may be assumed that behavioural interventions were deemed as the preferred choice of treatment for children or adolescents experiencing sleep disturbances. Although there is limited literature there has been some success in using bedtime fading to treat sleep disturbances such as delayed sleep phases which might have a circadian component. Due to the nature and focus of the intervention, bedtime fading does appear to be the ideal choice for the purpose of the current study; however, that can only be ascertained once the FBA has been completed for each child. The current study focuses on identifying if any circadian components are present in the sleep disturbances faced by children with DD and whether treating their circadian rhythm is sufficient to treat the problem. Relevant circadian interventions based on the FBA were selected for the children and then the interventions assessed for effectiveness as a treatment of their sleep disturbances. If they were not, alternative circadian interventions would be considered and analysed to determine if they might/could have produced different treatment effects.

Although using bedtime fading to treat sleep disturbances such as delayed sleep phases in children appears to be the ideal intervention, literature surrounding this is still lacking. In addition, barely any of these studies attribute or explore the underlying cause of relevant sleep difficulties to be a circadian disturbance. Even if identifying sleep disturbances as having circadian components, they are limited with the population of children with DD. The efficacy of using bedtime fading to treat circadian components does seem to have a promising future due to the successful results previously achieved in treating delayed sleep phases. For this reason, the current study could further augment the efficacy of using bedtime fading to treat circadian sleep disturbances in children with DD.

In the unfortunate case that all preferred choices of circadian interventions do not work, the next choice would be to proceed on to other interventions such as extinction. Standard and graduated/modified extinction has also demonstrated success in treating sleep disturbances such as delayed sleep onset, early awakenings or night awakenings including eliminating sleep interfering behaviour such as co-sleeping, therefore it may be considered if all circadian interventions fail to treat the problem.

Rationale

The current literature review has shed more light on how circadian rhythm plays a role in sleep problems, in the population of children with DD. The present study aims to identify circadian components from sleep disturbances present in children with DD, address them with the relevant behavioural intervention to see if addressing circadian components within these sleep disturbances are sufficient to treat the problem. If circadian treatments prove unsuccessful, other possible interventions would be suggested.

Sleep disturbance can be a rather serious problem and has been evaluated to have long term adverse effects in multiple areas of one's life if left untreated (Goldman et al., 2012; Richdale & Schreck, 2009). It requires the identification of the causal factors so as to be able to effectively develop appropriate intervention plans (Glickman, 2010). Because there are many variables to consider within the complexities of children with DD and sleep, it will be rather

challenging to interpret and utilise existing data as well as design treatments that will yield meaningful knowledge (Glickman, 2010).

Glickman (2010) hypothesised the probable contribution of circadian mechanisms towards specific sleep problems that occur in children with DD and that impairment in these circadian rhythms may be a potential cause of their sleep problems. Several other studies have also shown that the course and severity of DD may be further affected due to their disrupted sleep circadian rhythms (Cortesi et al., 2010; Goldman et al., 2012; Moon et al., 2011). According to Turner and Johnson (2013), sleep plays a multi-beneficial role within many developmental processes such as the development of executive functioning and maturation of the brain. For this reason, the efficacy of early detection and intervention for disrupted sleep circadian in children with DD is advised to be implemented as it is vital for optimising memory, behavioural, cognition, behavioural regulation, learning, and overall functioning in these children (Bougeron, 2007; Kotagal & Broomall, 2012; Turner & Johnson, 2013).

According to Kotagal and Broomall (2012), researchers have reported that practitioners are noticing an increasing number of patients with DD presenting for treatment and, further, that the cause may relate to disrupted sleep circadian components present in their sleep problems As already discussed, researchers have noted that this occurs when the normal patterns of sleep and circadian rhythms are disrupted, as a consequence of a misalignment between the sleep-wake cycle and the circadian system, due to direct impairment of the circadian functioning or due to the lack of physiological sleep pressure. According to Talay-Ongan and Wood (2000), children with DD are more susceptible to various variables that potentially impact the normal signs of entrainment, including the possible misalignment between the light-dark cycles and the circadian phase, thus there is a possibility that correcting their sleep circadian rhythms may treat the problem. This intensifies the need to develop an effective sleep intervention programme that involves treating the circadian component for these children, because the severity and magnitude of this rising issue demands increased research and treatment. Richdale and Wiggs (2005) have also reinforced that these sleep difficulties could be ameliorated if circadian rhythms are corrected, so as to improve overall functioning not only for the child but the child's family members as well. Thus, an adjustment to the sleep/wake schedules of children with DD may be a

treatment that could be tested to see if it is able to ameliorate sleep disturbances in children with DD and if not, to explore other types of treatment which may yield better success. This, however, may differ from family to family and can be ascertained by FBA.

Although the reviewed studies have generally found positive treatment effects in treating circadian components, existing literature surrounding the subject is still lacking. These will have considerable consequences for each child and their families (directly or indirectly) experiencing sleep difficulties, as there is a substantial prevalence within the population of children with DD. This justifies the need for further investigation into the efficacy of treating circadian components in sleep disturbances in children with DD.

The present study will thus add to the existing literature by investigating the efficacy of FBA-based behavioural interventions in addressing circadian-sleep disturbances within the population of children with DD. The aim is to improve the overall quality of sleep and daily functioning of children with DD and their families via an efficient behavioural treatment programme with its primary focus, on using a suitable intervention with circadian components. This will be determined via functional assessment of the case studies of three children with DD, to identify circadian components and to evaluate the circadian components of the intervention they were given and discuss it in the light of the literature review.

Research Questions.

In line with the aims of the study, the following questions will aid its structure and direction:

1. To what extent are circadian features evident in the presentation of children with DD and sleep problems?
2. Do FBA-based interventions have a positive effect on children with DD who suffer from disruptions in their sleep circadian rhythms?
3. If so, are circadian treatments sufficient to treat the child's sleep disturbances or are alternative treatments required?
4. How does the present treatment affect or improve the wellbeing of the child and their family members?

Chapter 2

General Methods

Research team.

The research on the efficacy of treating circadian-related components in children with DD presenting with sleep difficulties is part of a wider research project that is led by two senior academics at the University of Canterbury, who are a registered clinical psychologist and an ABA practitioner. The wider research project aims to investigate the efficacy of FBA-based interventions for sleep difficulties in children with DD. The clinical assessment and interventions with the families were conducted under their supervision, by a registered psychologist or registered intern psychologist.

Ethics and participant consent.

The study was approved by University of Canterbury Human Ethics Committee and both parents and child signed written consent to participate in the study. Additional parental and child consent were obtained for audio-visual recording of their child's sleep, to be taken during the programme.

Data analysis.

Tables were drawn up to constitute data collated for each child, from baseline, intervention, and during maintenance and follow-up, for each target dependent variable. These included sleep onset latency (SOL) (mins), frequencies of curtain calls (CCs), duration of NWs (mins), frequencies of night awakenings (mins). The visual analysis allowed for monitoring of changes in behaviour throughout the intervention and determine if these changes could be attributed to the intervention.

Referral.

Participants were recruited within New Zealand. Participants responded to flyers that provided information regarding the study to relevant organisations. The study invited participants for self-referral or referral from relevant organisations.

The cases discussed in the current study were existing cases, and because of this, this study by itself was unable to shape the interventions. Access was granted to a small number of completed cases that fit the criteria of the current study. The cases have therefore been included to reflect on the presence and importance of circadian features and their implications for treatment.

Screening and consent.

The potential families were initially screened by a telephone interview to ensure they fit the inclusion criteria. The nature of the programme was then described to the families and their initial consent to participate was obtained. A child consent form also had to be signed; however, parents could sign on their behalf if the child was unable to consent themselves due to their developmental level. Both families signed the consent form on their children's behalf.

Inclusion/exclusion criteria.

Participant cases were eligible for inclusion the study if they had, 1) a formal diagnosis of a developmental disorder; 2) presented with sleep difficulties which had possible circadian components such as delayed sleep onset latency, frequent and/or long-lasting early morning awakenings or night awakenings, and daytime sleepiness which were identified via parent-reported sleep diaries and video footage; and 3) were aged between 3 and 19 years old. Children who had any medical or physical complexities that could interfere with efficacy of the treatment or their safety, was excluded. Children whose parents were unable to or unwilling to adhere to treatment procedures, established from initial screening, had to also be excluded from the study.

Participant justification.

The study age range was selected with the aim of focusing on children with developmental disabilities. Participants' personal information such as actual names and ethnicity were omitted to ensure confidentiality and anonymity of participants and pseudonyms were used to replace actual names. The participant number of cases to be included were variable and according to timeframe feasible for a master's thesis, and the data available.

Setting.

The research team was based in Christchurch but participants being based in Christchurch was not an inclusion criterion for the research project. Clinical intake interviews, which included initial screening for eligibility of participants and including the intervention of participants, were conducted throughout New Zealand. Baseline, intervention, follow-up, and maintenance occurred within the participants' homes and contact was kept whenever needed via telephone or Skype. Relevant equipment or resources were also delivered via email or standard post. The intervention was carried out in the participant's home and the intervention was carried out by their primary caregivers. Communication was facilitated between the team and the families via telephone, email, and text, as required.

Materials.

Parent-reported sleep diaries.

Parents were instructed to record sleep diaries daily by observing and measuring their child's sleep behaviours. These sleep diaries documented times of where and when the child was put to bed, the time he/she actually fell asleep, CCs (if any) before achieving sleep onset, frequency, and durations of NWs, total sleep duration, daytime naps, bedtime non-compliance, co-sleeping and the time he/she was up for the day (France et al., 1996). Parents were to record detailed events of each night including if the child was unwell or any other factors that may have impacted the child's sleep (such as temperature and weather). The diaries were split into 7-days

per page with the dates and days included. They were used to record changes in participants' sleep patterns and behaviour from baseline, throughout intervention and at follow-ups.

Video footage.

Video recording equipment, such as time-lapse, night-time, and infrared cameras were used to record visual footage of participant sleep patterns and behaviour at night. They were placed in a convenient location within the participant's bedroom and recording took place from after the participant had been put to bed until he/she had risen for the day. This was to help the research team and the participant's parents be able to observe his/her behaviour while sleeping and measure it for accuracies with the sleep diaries. Video footages were used during baseline and throughout intervention, but not at follow-up.

Sleep assessment and treatment tool (SATT, Hanley, 2005).

The sleep assessment and treatment tool (SATT) is a functional assessment questionnaire that aims to highlight the correlation between environmental variables and sleep problems of participants. It includes sections such as the child's sleep history, identification of problems, antecedent conditions, consequent actions, goals, the child's sleep schedule as well as any sleep-interfering behaviour.

Children's sleep habits questionnaire (CSHQ, Owens et al. 2000).

The children's sleep habits questionnaire (CSHQ) is a 45-item parent-completed questionnaire that attempts to evaluate the child's sleep problems based on their current sleep behaviours. The questions are set according to a 5-point Likert scale from *never* to *always* and a score above 41 indicates a presenting sleep problem. The questionnaire includes domains such as bedtime behaviours, daytime behaviours, sleep duration and morning waking. It is typically administered during baseline and at post-intervention.

Questions About Behavioural Function (QABF, Vollmer & Matson, 1995).

Questions about behavioural function (QABF) is a widely used parent completed 25-item brief functional assessment questionnaire to assess the child's maladaptive behaviours on a 3-point Likert-type scale of *never occurs* to *occurs often*. This is completed during baseline.

Child Behaviour Checklist (CBCL; Achenbach & Rescorla 2001).

Child behavior checklist (CBCL) refers to a checklist that parents are to complete to help detect behavioural and emotional problems in children as well as adolescents. It uses a 3-point Likert scale from *absent* to *occurs often* to measure syndromes such as somatic complaints, anxiety/depressed, attention problems, aggressive problems, social problems and the like. This was used to assess for any internalising or externalising behaviours that might interfere with the treatment and to address it accordingly if it was.

Treatment Acceptability Rating Form-Revised (TARF; Reimers & Wacker, 1988).

Treatment acceptability rating form-revised (TARF) is a 20-item questionnaire used to measure parent's perceptions towards the overall acceptability of the treatment on a 7-point Likert scale of strongly disagree to strongly agree. It contains questions such as "Is this treatment fair?" "Would you recommend it to others?" and such, to measure for social validity. This was completed during post-intervention at follow-ups.

Dependent Variables.

Dependent variables were measured by the type of sleep difficulties and the parents' intervention goals. Data for these dependent variables were collated from the start of the bedtime routine until the time when the child woke for the day.

Awake. Awake was defined as the child having his/her eyes open and demonstrating some form of sleep-interfering behaviour (defined below), excessive body movements or vocalising that would indicate wakefulness.

Asleep. Asleep would be defined as the child demonstrating his eyes closed, without vocalisation or voluntary movements.

Co-sleeping. Co-sleeping was defined as the child lying in bed with someone (caregiver) for an amount of time at any point during the night including initial sleep onset or due to a NW. This behaviour could have been initiated by either the child or the parent.

Sleep-interfering behaviour. Sleep-interfering behaviour was defined as any behaviours that was exhibited after putting the child to bed that interfered with the child's ability to initiate sleep. This included stereotypic behaviours (i.e., repetitive movements), vocalisation (such as, screaming, talking, crying, singing) and physical movements (such as, standing, walking, sitting, leaving the room, playing with objects).

Sleep onset latency. Sleep onset latency (SOL) was defined as the duration that the child takes to achieve initial sleep onset, after being put to bed.

Night waking. NWs were defined as any awakening that occurred between, after initial sleep onset and before set wake time, and lasting for more than 2 minutes.

Curtain calls. CCs were defined as any behaviours demonstrated to gain parental attention (i.e., leaving their bedroom to get to their parents, calling out to parents to enter their bedroom without leaving their bed), exhibited during initial sleep onset.

Procedure.

The study phases included assessment, baseline, intervention, maintenance, and short-term follow-ups.

Assessment.

This phase consisted of a standard open-ended clinical interview (via Skype or telephone) taken by the registered inter psychologist at the University of Canterbury. Key information was gained around information containing previous records or attempts to address sleep difficulties, the child's official diagnosis, any physical or medical conditions that may be contributing factors and, relevant developmental and family history of the child's life and family, including the opportunity for parents to clear any doubts they had. Pre-intervention measures (i.e., sleep outcome measures, child's general behaviour, daytime functioning) were also completed. Collated data was then utilised to formulate the FBA-based interventions.

Baseline.

Parents collected data for baseline for one to two weeks via sleep diaries and video footages and were instructed to respond as normal to their child's sleep difficulties so as to accurately obtain their natural existing behaviours.

Intervention.

After collating data from baseline, the research team and the participant and their family decided the most appropriate approach to treatment. This was dependent on the results from the individual FBA and are different for each participant. Based on the evidence accumulated, an intervention which was based on antecedents (i.e., irregular sleep-wake times, lack of bedtime routine) and consequences (i.e., established set sleep-wake times, consistent bedtime routine) was created. This included targeted child interventions and reward programmes. Support was available to the parents throughout the intervention if and whenever the need should arise.

The intervention continued until the child's presenting sleep problems had significantly decreased or had been eliminated. The time period for the intervention approximately lasted between two to three weeks per participant.

Common treatment components.

Establishing consistent sleep-wake times. Parents were asked to establish a consistent sleep-wake schedule by putting their child to sleep at specific sleep times and wake them at specific wake times. Daytime naps were also eliminated to aid in increasing their physiological sleep pressure towards bedtime. Children were kept awake by various methods individual to each family (e.g., watching television, playing with toys, or other activities).

Faded bedtime. Faded bedtime involved pushing the child's bedtime to a later time. This was meant to increase the child's pressure for sleep, which would in turn decrease their sleep onset latency (i.e., child achieves quick initial sleep onset). Their bedtime was then gradually shifted forwards in 15-minute increments as the child showed progress at each stage.

Discriminative stimuli for sleep and wake times. To learn how to differentiate the difference between sleep and wake times and expected behaviour associated with each respectively, visual aids were introduced to the children. This included prompts such as social stories and Gro-clocks. The social story consisted were individually designed for each child and typically consisted of steps for their bedtime routine, sleep-behaviour goals and reinforcement contingencies, which were typically depicted using pictures and brief developmentally appropriate, text. This was typically read to the child every night before bedtime to familiarise them with the new routine. The Gro-clocks were used to help the children differentiate between morning (sun) and night (moon). Information about this was also added in the social story; they had to stay in bed while the moon was showing and could leave their bed when the sun showed. This was also useful during any sleep interfering behaviours, where the child could self-direct or the parents could redirect the child towards the clock reminding them that they could get up when the sun showed.

Rewards. Reinforcement contingents were provided by parents to their child, in the form of tangible rewards (i.e., access to tablet, food, television time) or verbal praise ("well done staying in bed!" etc.), when the child demonstrated the target behaviours (i.e., staying in bed all night).

Maintenance.

After the intervention, the parents were asked to begin the maintenance phase. During this period, the parents were to continue the new behaviour plan to become assimilated in their daily lives. They were still able to contact the research team if the need arose.

Follow-up.

Short and long-term follow-ups were conducted again at 5 to 6 weeks and 11 to 12 weeks respectively. This was to measure the maintenance of behaviour and record any changes that occurred. Parent-reported sleep diaries were therefore completed during follow-ups.

Description of Planned Data Analysis.

Multiple case study designs allow good visual analysis for the gathered data (Kazdin, 1981). Visual representations (tables, graphs) would be used to present and measure the efficacy of FBA informed interventions in the current study. Data that been collated from the case studies would be demonstrated via graphs of relevant dependent variables.

Treatment efficacy was demonstrated via a decrease in problem behaviours during intervention and follow-up phases as compared to baseline. If stable behaviour was demonstrated during baseline, it would likely suggest that any change that co-occurred with the intervention was due to the intervention itself. If changes were reflected during baseline, it was likely there were other factors that were likely contributing to or causing the change and not by the intervention itself. Some literature surrounding sleep has demonstrated that sleep difficulties may worsen over time, therefore improvements resulting from treatment efficacy would be received well, as it would be considered unexpected.

Treatment efficacy was also measured by analysing pre and post outcome data on CSHQ which indicated treatment efficacy in reducing or eliminating sleep difficulties. TARF-R was completed by participants and their families to assess overall treatment acceptability.

Chapter 3

Case Study 1

Dexter (pseudonym) was an 8-year -old boy with a diagnosis of ASD who lived with his mother and sister (10 years old). His mother responded to an advertisement by the clinical research team looking for participants for their sleep study, to help improve his sleep.

Presenting complaints.

Dexter's mother's main concern was that he was not receiving optimal sleep due to his sleep onset delay, curtain calls (CCs; i.e., sleep-interfering behaviour that was displayed after being put to bed such as bids for parent attention, leaving the bedroom, requesting food/drink, etc.), and early awakenings, and how this would impact on his overall development. Dexter's mother also raised concerns about his consumption of caffeinated drink (Coca Cola) and requested for it to be addressed within the programme provided. She also provided information that Dexter had not yet established toilet training and that it may a factor that was interfering with his sleep. She aimed for Dexter to achieve a consistent bedtime of 9:30pm and wake time of 7:30am.

Dexter's mother showed keen interest in getting him to participate in the study due to their specific concerns regarding the length of time it took for him to fall asleep (between 4 to 6hr after going to bed) and the amount of sleep he was receiving every night as he usually fell asleep around midnight or later, and was awake for the day by 7.30 to 7.45am.

Family context and developmental history.

During assessment, it was identified that at a young age, Dexter had been observed to have delayed development and limited verbal language. He was initially able to sleep through the night as an infant but started to experience sleep disturbances when he began to attend school. At

the time the study commenced Dexter was not toilet trained and was reported to be a selective eater. He had also not received any intervention for his sleep prior to commencing the study.

Case study method.

A clinical interview was conducted to attain key information regarding Dexter's sleep, developmental history, and behaviour, as well as to rule out other medical or physical factors that may be contributing to his sleep problems or that might affect treatment. It was decided that it was appropriate to include this family in the sleep programme.

An FBA was conducted to attain all information regarding antecedent and consequence variables impacting his sleep and the function of any sleep interfering behaviours (via clinical interview, analysis of sleep diaries and video content, data from assessment tools (e.g., SATT)). Dexter's mother identified that his Coca Cola consumption was one of the concerns that needed to be addressed within the sleep programme. Dexter's mother, therefore, recorded diaries for his daily Coca Cola consumption to establish how much Coca Cola was being consumed to baseline and to develop an approach to reducing this. It was anticipated that by decreasing his consumption of caffeine (Coca Cola) in his daily diet, it would have a positive effect on his sleep.

The time between when Dexter was put to bed and time until he finally fell asleep was determined as sleep onset delay (SOD). CCs was considered as any behaviours demonstrated for parental attention during this time. CCs were defined as, all instances of Dexter getting out of bed to seek his mother's attention (such as, to wish her goodnight) or requests made for attention or for tangible items (ice pack, drinks, hugs) after he was put to bed.

Dexter's mother was asked to record sleep diaries for two weeks during baseline. These sleep diaries recorded a) sleep setting, b) time put to bed, c) the frequency of CCs, d) parental responses to each CC, f) the time of sleep onset and g) the time he woke up for the day. She was advised to respond to Dexter's sleep and his behaviour how she normally would. Once the baseline diaries had been collected and FBA completed, the information was used to formulate an individualised treatment plan.

Definition of key terms.

Interfering behaviours. This referred to any behaviour that was adversely affecting Dexter's sleep onset delay including parental responses to those behaviours.

Curtain Calls. This referred to instances of Dexter getting upset, requesting items (i.e., toys), getting up from bed and using his iPad, including any other negotiating behaviour (ie., pleading with his mother).

Parental attention. This referred to Dexter's mother's responses to his sleep interfering behaviour and included, verbally and/or physically returning Dexter to bed, changing his nappy before putting him back to bed, repeatedly wishing him "night-night" or spending time with him on his iPad.

Results of the FBA.

The SATT contained information regarding the antecedent and consequence variables that were influencing Dexter's sleep. The outcome of the SATT showed that Dexter did not have a set bedtime routine or practice proper sleep hygiene. He went to bed at variable times almost every night and eventually fell asleep at variable times. He could be asleep as early as 9pm to as late as 2am and in between these times, there was a high frequency of CCs. The average amount of time he took to fall asleep was around 270 minutes. After he had been put to bed, he would come out to wish "nigh-night", to ask for a drink or a hug, an ice pack or some other distraction. The length of time spent engaged in CCs or time taken to fall asleep was also variable every night. The number of CCs Dexter demonstrated, ranged between 1 and 13, with the average number of CCs being around 7 to 8. There were no reported NWs across study phases. His wake time was also highly variable and ranged between 3am and 10:30am.

The outcome of the FBA suggested that there were a number of antecedent and consequence variables that were influencing Dexter's sleep problems. Antecedent variables detected that were affecting Dexter's sleep problems were the lack of proper sleep hygiene practices and consistent bedtime routine (lack of regular pre sleep-initiating practices such as no access to toys and electronic gadgets at bedtime). The timings of Dexter's sleep and nature of his

sleep pattern suggested a disturbance in his circadian rhythm as the most likely primary cause of his sleep problems. Dexter's sleep disturbance was also likely influenced by the amount of caffeine in his system (i.e., from the consumption of Coca Cola), which was possibly disrupting his sleep circadian rhythm, The consequences that were detected to be reinforcing SOD seemed to be the parental responses to CCs and easy access to tangible items such as digital devices, as well as food and drinks.

Table 1 provides a summary of each identified problem behaviour, including the source of information for each behaviour, treatment goals, factors identified during the FBA process as maintaining or precipitating the target behaviour/s and the treatment that was used to target the problem behaviour.

The goals of Dexter's parents were for him to a) fall asleep by 9.30pm and wake by 7.30am and b) reduce/eliminate the consumption of Coca Cola in his diet.

Table 1 Parent reported sleep problem/s, factors precipitating and/or maintaining the sleep problem, hypothesised function, and intervention

	Parent reported sleep problem/ s	Factors precipitating and/or maintaining behaviour	Hypothesised function	Intervention
Dexter	Frequency of CCs	Social attention	Attention	Positive sleep hygiene; Caffeine restriction; Positive reinforcement; Elimination of device use/story reading in bedroom
	SOD/ Delayed Sleep Phase	Social attention; caffeine consumption, need for nappy changes, access to tangible items, diminished sleep pressure	Attention Tangible Escape from bed	Positive sleep hygiene (including scheduled toileting); Graduated bedtime fading; Caffeine restriction; Elimination of device use/story reading in bedroom; Positive reinforcement; Consistent sleep-wake times
	Early Morning Wakings	Social attention; access to preferred activities	Attention Tangible Escape from bed	Positive sleep hygiene; Delayed bedtime; Positive reinforcement; Elimination of device use/story reading in bedroom; Consistent wake-time

Note. CCs; Curtain Calls, SOD; Sleep Onset Delay

Treatment procedures.

The interventions used included a) manipulating antecedents: establishing a consistent bedtime routine (i.e., positive sleep hygiene+ set sleep-wake times), reducing Coca Cola consumption to reduce the intake of caffeine, establishment of regular toileting, restricting access to tangible items before bedtime (i.e., electronic devices, food/drinks), b) consequence manipulations: regular bedtime schedules, use of tangible items as positive reinforcements.

These strategies were important to help overcome his sleep difficulties as they directly targeted the sleep interfering behaviours. Each of these strategies is described in detail below.

Faded bedtime. Faded bedtime was identified for use with Dexter as he was experiencing severe SOD (up to 420 minutes) and a high frequency of CCs. The goal was therefore to decrease his sleep onset time and the frequency of CCs. This was to be carried out by setting Dexter's sleep time closer to his natural sleep time (8pm) initially, and gradually fade it back as each faded bedtime was met with success, until the goal bedtime (9.30pm) was achieved. The purpose of using a faded bedtime procedure was to set a later initial bedtime (10.10pm) and gradually bringing it forward until goal bedtime (9.30pm), would help reset Dexter's circadian rhythm by increasing his biological sleep pressure, thereby reducing his SOD.

Reducing caffeine consumption. Dexter's daily consumption was gradually reduced while gradually increasing his intake of water and also water was strategically placed within reaching distance from Dexter, wherever he was playing.

Consistent bedtime routine. A consistent bedtime routine was established for Dexter. His bedtime routine included first brushing his teeth then washing his face, having a nappy change and putting on his PJs, doing calming activities before bed such as playing on his devices in the living room, then placing his devices back in the finish box and having a final nappy change before going into his bedroom and wishing good night. He then got access to his devices again when he woke up in the morning at 7.30am, because his device use was identified as problematic during the assessment. It was gathered during assessment that Dexter's mother was not very

particular about his wakeup time, just that it did not cross 8am. She was asked to follow this routine consistently each night.

Dexter was to be put to bed at 10.10pm after having some device time in the living room. After being put to bed, Dexter's mother would bid him goodnight, turn of the lights and close the door. She would usually be out of the room immediately after that, as Dexter is able to fall asleep without her being in the room. It was agreed that if he did have a curtain call; she would redirect him back to bed.

Sleep hygiene. The temperature also seemed to be a minor factor as Dexter would request for an ice pack on hot nights or just wander off to the freezer. This was addressed by the mother giving him ice packs to take to bed with him.

Reinforcement. Reinforcement was delivered contingent upon him following his new sleep routine; using his finish box (a box used to put his iPad back before going to sleep) and an absence of CCs. Reinforcement was delivered immediately upon waking up for the day. This included access to his tablet in the morning.

Procedural modifications.

Dexter's mother continued putting him to bed at his usual bedtime. This was attributed to the changes already achieved in Dexter's sleep from his caffeine intake prior to intervention (maximum of 2 Coca Cola drinks/day before 4pm). There were improvements noted in Dexter's SOD and the number of CCs from baseline (recorded for two weeks before intervention) and throughout the first part of the intervention. They therefore did not modify his bedtime as was intended. Elimination of device use in bedroom was also accepted easily by Dexter as he had already started adapting to the new routine.

Results.

Treatment outcomes following the sleep intervention are presented in Figure 1 and 2. This includes data relating to sleep onset delay and CCs. Data from the CSHQ gathered at pre- and post-intervention has also been reported along with post-intervention data gathered from the TARF-R and follow-up.

After 18 days of the intervention, there were 0 curtain calls and sleep onsets were achieved within 15 minutes of being put to bed. From day 18 onwards to end of the intervention, Dexter had met his goal bedtime and there were no CCs noted.

Dexter's sleep intervention lasted for a total of 18 days (excluding baseline which was 7 days). There was no data available from day 14 to day 17 due to Dexter being away.

The frequency of CCs and duration of SOD have been presented in Figures 1 and 2 respectively.

Curtain calls.

Figure 1 demonstrates data for the frequency of CCs during baseline, intervention and follow-up for Dexter. It was noted that, there were some frequencies of CCs exhibited during baseline. They were variable, with an average of 3 CCs per night; however, they were noted to be almost eliminated following that, across all study phases (including at follow-ups) with the exception of night 27 (three CCs) and night 32 (one CC). CC episode noted on night 32 was extremely brief (only 1 to 2 minutes). Dexter had far less frequencies of CCs during baseline (zero CCs on 5 nights) as compared to during assessment (CCs noted every night; up to 13 CCs per night).

Sleep onset delays.

Several nights with SOD were demonstrated during baseline but there was a decrease to almost, but not zero nights, immediately after treatment initiation for Dexter. There was an average SOD of >30 minutes throughout all sleep phases (except baseline, which had an average

of about 4.5 hours) including at follow-up, with the exception of few nights (see Figure 2). It was noted that Dexter's mother did not entirely adhere to the set bedtime of 10.10pm during intervention phase and instead put, or allowed Dexter to put himself to sleep, between 8pm to 9.30 each night, with the exception on those nights where they were unable to record sleep diaries. The supposed start time (10.10pm) during the intervention phase was therefore deviated most likely, due to improvements already noted from the decrease in caffeine (Coca Cola) from Dexter's diet during baseline.

There were some nights where Dexter spent a period away from home (day 14-17) therefore data was not available some nights (day 124; long-term follow-up) where Dexter was unwell which were considered as exceptions.

Caffeine intake.

Figure 3 represents the plan for decreasing Dexter's Coca Cola-consumption whilst increasing his intake of water. He was started off with 6 glasses of Coca Cola at home and 2 glass of water each at home and at school. This was gradually reduced daily by about 1 to 1.5 glasses whilst increasing his intake of water by about 2 mouthfuls to 2 glasses, which was a success.

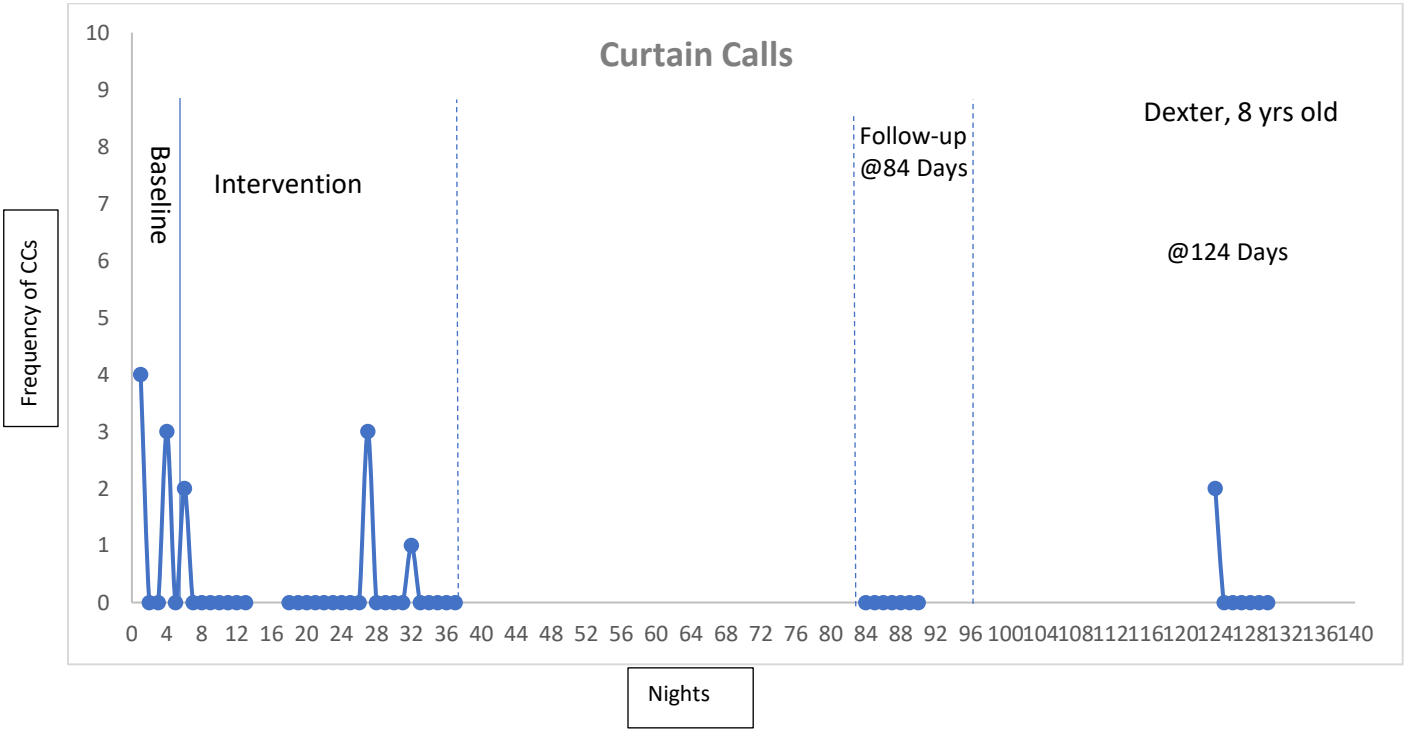

Figure 1:The frequency of curtain calls during baseline, intervention, and short- and long-term follow-up

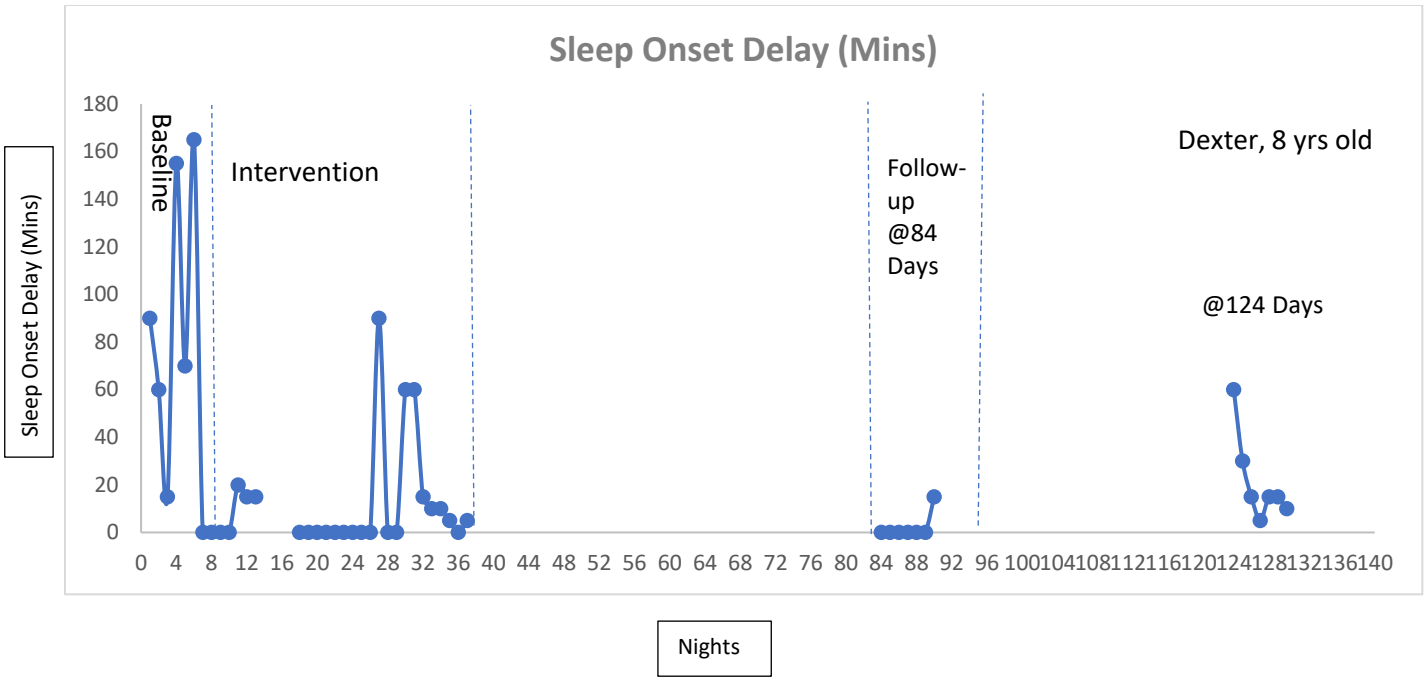

Figure 2:Duration of sleep onset delay in minutes for baseline, intervention, and short- and long-term follow-up

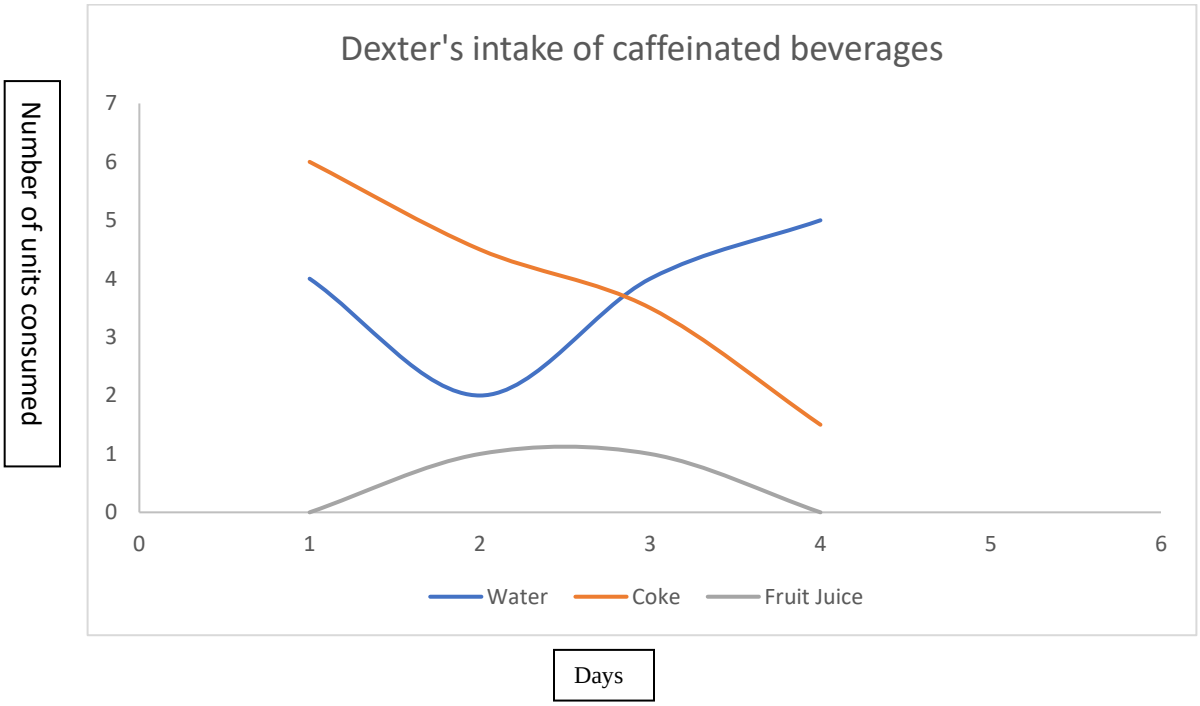

Figure 3: Dexter's intake of caffeinated beverages

The results from the CSHQ Abbreviated Form (Owens et al., 2000) are collated in Table 2. The results indicate that there was a decrease in bedtime resistance scores from 8/24 pre-intervention to 6/24 post-intervention, and sleep onset delay from 3/4 to 1/4, which indicated a decrease in frequencies of nights where Dexter had SOD, between pre- and post-intervention. There was an overall improvement in the total scores between the pre- (score= 47) and post-intervention (score= 38) which indicated that Dexter's sleep disturbances had reduced by the end of intervention.

Table 2: Comparison of Pre-and Postintervention Scores on the CSHQ Completed by Dexter's Mother.

Variable Scores	Pre-intervention	Post-intervention	Maximum score
Bedtime resistance	8	6	24
Sleep onset delay	3	1	4

Note. CSHQ=Children's Sleep Habits Questionnaire

Results of the TARF-R are collated in Table 3. The overall score showed a very good level of treatment acceptability with a score of 108 out of 119. The results demonstrate that Dexter's mother rated the treatment as reasonable (21/21) and effective (21/21). She also rated 3/21 on the reverse 21-point scale demonstrating that there were no likely side-effects to result from the treatment neither did Dexter experience any discomfort during the treatment process. There was also high understanding of the treatment process with a score of (7/7). She also reported relatively high on willingness to complete the treatment (15/21) and that the treatment was very affordable (14/14). She reported Dexter's sleep problems to be quite severe (11/14) and that sufficient time was needed to carry out the treatment (16/21). From the results, it was indicative that Dexter's mother was overall satisfied and happy with the treatment procedure and outcomes.

Table 3: Postintervention Treatment Acceptability Ratings on the TARF-R

Variables Score	Mother	Maximum
Total Accessibility	108	119
Reasonableness	21	21
Willingness	15	21
Cost	14	14
Side-Effects	3 (reverse= 21)	21
Effectiveness	21	21
Disruption/Time	16	21
Problem severity[a]	11	14
Understanding of Treatment[a]	7	7

Note. TARF-R= Treatment Acceptability Rating Form-Revised.

[a]Not Included in total treatment acceptability score.

Post-treatment interview.

Dexter's mother reported the intervention to be clear and easy to understand, acceptable, effective, but also perceived it to be "tedious"; requiring quite a bit of effort and time to implement; including the "couple of months needed for diaries". Dexter was reported to have shown overall improvements after undergoing the sleep programme. Dexter's overall mood had also shown improvement and he was reported to be more humorous and sociable. She reported also having learnt valuable strategies which enabled her to continue to manage his sleep even after the programme ended. In addition to all the benefits on Dexter's sleep and behaviour, she also reported getting more sleep herself and noticed an improvement in her own moods and overall wellbeing.

Case Study 2

Tom (pseudonym) was a 6 years old boy living with his parents. By the age of 2 he had been diagnosed as having ASD. As had been the situation for Case Study 1, Tom's parents responded to an advertisement posted by the clinical research team, recruiting participants for a sleep programme, because they wanted to help their son with his ongoing sleep disturbances.

Presenting complaints.

Tom's parents had concerns around the frequency and duration of Tom's NWs and how this might affect his daily functioning. Tom typically had two NWs per night where the first episode would occur approximately 4 hours after initial sleep onset (between 6.30 to 7pm). During these NWs, he would sometimes express aggressive behaviour (such as shouting or hitting) or have toileting accidents which would occasionally result in Tom's mom co-sleeping with him. Tom's parents also reported that he was experiencing daytime sleepiness at school. He also started taking melatonin for his sleep but that had no significant impact on his NWs. He was not taking melatonin at the time of the study. Tom's parent's goals were for his NWs to be reduced or eliminated.

Family context and developmental history.

Tom's parents reported that he was able to have uninterrupted sleep for about 10 to 12 hours as an infant. Tom also had a consistent sleep routine and slept in a conducive sleep environment. Tom's parents also mentioned that he had yet to establish toilet training and had a preference for having routines (consistency and predictability in what he was expected to do or how to behave). He also tended to have stereotyped interests (such as aeroplanes and dinosaurs). Tom had limited verbal capacity and typically communicated using physical gestures (visual and sign language).

Case study methods.

Tom's parents' specific concerns regarding his sleep led to him being involved in the sleep programme. A clinical interview was conducted to gather relevant information regarding Tom's sleep, family setting and developmental history, his behaviours mostly pertaining to sleep including information to rule our factors that may have or could be contributing to his sleep problems or that had the potential to interfere with the treatment (medical or physical).

Tom's NWs was identified as a primary concern by his parents and needing to be addressed within the intervention.

NWs were deemed as any waking episodes that had occurred after initial sleep onset and the duration of these awakenings were noted, as the time between the NWs until sleep onset was resumed. Behaviours that had interfered with Tom's ability to stay asleep at night or that contributed to his NWs, were considered as sleep interfering behaviours.

Definition of key terms.

Sleep-interfering behaviours. Interfering behaviours that occurred during NWs usually consisted of behaviours such as talking, crying, banging or kicking, going into parent's room, or toileting needs.

Parental responses. Parental responses to these behaviours included responding to toileting needs, cuddling or resettling back to bed or co-sleeping.

Research procedures.

SATT (Hanley, 2005) was completed by Tom's mother during the assessment stage.

Tom's parents were instructed to continue with their typical daily routine without making any changes while recording the sleep diaries during baseline. This was to ensure accuracy of data collated.

Results of the FBA.

From the diaries, it was indicated that Tom did not face issues with sleep onset. Tom typically fell asleep between 6.30 to 7pm and woke between 6am to 6:30am for the day but he experienced episodes of NWs with the first one being about 300 minutes to 480 minutes after sleep onset (approximately between 10:30pm to 2:30am) and the second NW typically being between 3am to 6:30am. The amount of time of each NW episode was variable, between 30 minutes to 120 minutes with the average being about 90 minutes. The sleep diaries for him also showed that the toileting accidents that at occurred at night, coincided with his NWs.

From the FBA, it was detected that the antecedent variables contributing to Tom's sleep difficulties included daytime naps, attention-seeking behaviours (such as, aggressiveness by biting or kicking and throwing tantrums) and nighttime toilet accidents. The consequences appeared to be parental responses, such as attending to his toileting needs and resettling him or co-sleeping, after each NW. The primary concern that seem to maintain the NWs seemed to be a circadian disruption in his sleep schedule with early bedtimes (6-7pm), and parental attention. There seemed to be a lack of physiological sleep pressure for Tom due to his daytime naps and early bedtimes resulting in his circadian body clock becoming out of synchronicity with the external environment (going to sleep when it is not yet dark). His sleep patterns therefore suggested a circadian disturbance to be the most likely factor for his sleep difficulties with parental responses to his toileting needs or other bids for attention being a secondary factor.

Table 4 provides an overview of each identified problem behaviour, source of information for each behaviour, intervention goals, the factors identified during the FBA process as precipitating or maintaining the target behaviours and the intervention implemented to address the target behaviours. The goals of Tom's parents consisted of a) Tom to independently achieve 8 hours of consolidated sleep through the night without NWs, and b) for the parents to be able to sleep in their own bed without having to co-sleep with Tom after his NWs.

Table 4: Parent reported sleep problem/s, factors precipitating and/or maintaining the sleep problem, hypothesised function, and intervention

	Parent reported sleep problem/s	Factors partcipating and/or maintaining behaviour	Hypothesised function	Intervention
Tom	Frequent NWs	Social attention; access to tangible items, diminished sleep pressure from daytime naps	-Attention -Tangible -Escape from bed	Positive sleep hygiene (including scheduled toileting); Graduated bedtime fading; Social story; Elimination of device use/story reading in bedroom; Positive reinforcement; Consistent sleep-wake times
	Prolonged NWs/ night tantrums	Social attention; access to tangible items, diminished sleep pressure from daytime naps	-Attention -Tangible -Escape from bed	Positive sleep hygiene (including scheduled toileting); Graduated bedtime fading; Social story Elimination of device use/story reading in bedroom; Positive reinforcement; Consistent sleep-wake times

Note. NW; Night Waking

Treatment procedures.

Tom's treatment consisted of a) antecedent manipulations: restricting daytime naps, limiting activities other than sleep, that he engaged in while in his bed, establishing consistent sleep-wake times, a social story, and a Gro-clock (details below); b) consequence manipulations: positive reinforcement of sleeping through the night without NWs. More detailed information regarding each intervention component is provided below.

These strategies were formulated to help deal with Tom's sleep interfering behaviours and promote sleep-conducive behaviours. Identifying his sleep interfering behaviours were helpful as they helped in developing these relevant strategies to help Tom overcome his sleep problems. Each strategy has been described in detail below.

Faded bedtime. Based on parent-reported sleep diaries, the data containing Tom's sleep time helped the research team to establish what time to start the graduated bedtime fading. Tom's circadian rhythm was aimed to be reset by setting a consistent sleep and wakeup time. It was therefore decided that his bedtime would be delayed by an hour to help consolidate his sleep and help him sleep until later in the mornings. The time would gradually be adjusted as his sleep improved. Tom's parents were told to push back his bedtime by an hour when intervention started. Tom was therefore put to bed at 8pm (his normal bedtime is usually 7pm) each night after treatment initiation and woken at 6.30am the morning after.

Fading his bedtime was also discussed in preparation for daylight saving. Tom also had his bedtime shifted gradually to 7.45pm and then to 7.30pm when it was time to prepare for daylight saving. His wake time was also pushed earlier (ie. 5.45am) and gradually moved back forward, once Tom had been consistently reinforced for staying in his room until the sun showed on the clock. This is meant to help Tom increase his chances of success and thereby increase his understanding or learning of what is expected of him.

Sleep hygiene. The sleep hygiene included using a battery operated Gro -clock, completing all activities before bed (e.g. reading before bedtime) out of the bedroom area and using the bedroom only for sleeping, and also using social stories. Consequence modifications included rewards for demonstrating appropriate behaviour.

Consistent bedtime routine. For Tom to learn to associate his bed with sleeping, any other activities other than sleeping were eliminated from his bedroom. He would complete his typical bedtime routine before sleeping, which consisted of brushing his teeth, toileting, putting on his pyjamas and reading a book *outside* of his bedroom. He was read to in the lounge and put to bed while he was still awake. The parent could then either leave the room or sit in a chair next to him if needed, until he achieved sleep onset (faded out gradually). Tom's parents were to implement this routine every night to help set a consistency of the new routine for Tom.

Social story. By day 26 of the intervention procedure, Tom's bedtime had been established but his wakeup times were still variable and earlier than the set wake-up time. Tom was also leaving his bedroom after waking up therefore Tom's parents tried to keep Tom in his room until his set wake time even if he was not sleeping. A social story was therefore created for Tom, informing him of his new bedtime routine and model desired sleep behaviour. This was meant to

be read to him every night at least once right before going to bed. It included photos of Tom and text describing the bedtime routine. It also included pictures of his Gro-clock. There were also pictures of his rewards with text prompts, which he would earn for getting through the night without leaving his room.

Gro-clock. Tom was introduced to the gro-clock and had it placed by his bedside every night. The gro-clock depicted stars for nighttime with text prompting him to go back to bed and only wake up when he saw the sun on the clock signaling morning. He was made aware that he was able to check the clock if/whenever he had a NW, to see if the sun or moon was up and behave accordingly.

Reinforcement. Reinforcement contingent for staying in his bed all night would be immediately delivered upon waking for the day. This included tangible and social rewards. Tangible rewards included device time and intangible rewards included social praise such as providing him with a lot of attention and specific praises (such as, "well done for staying in bed all night!", cuddles, kisses, thumbs up, and hugs). Once Tom had learnt how to stay in his own bed all night, the daily rewards would be gradually faded out. This was not recorded, when/if his rewards were faded out.

Modifications to the time was attributed to daylight saving. It was implemented just prior to start of daylight saving, to prepare Tom for the change and to avoid as much disruption to his new routine as possible.

Results.

Intervention outcomes following the treatment programme for Tom, have been presented in Figure 4. This included relevant data relating to the frequencies and durations of his NWs. Because the study was completed recently, only short-term follow-up was collected, and no other post-treatment data are reported.

Tom's treatment programme lasted for a total of 59 days (baseline: 7 days). There was no data available on day 28 and day 55. No explanations were provided.

The frequency and duration of NWs for baseline, intervention and follow-up are present in Figure 4.

Night awakenings.

During baseline, Tom exhibited a high frequency of NWs (two NWs every night). The duration lasted from 10 minutes to 60 minutes. There was an average of 30 minutes throughout baseline. There was however an immediate decrease in the frequency of NWs (observed only on 5 nights, one NW episode per night) after intervention initiation, which lasted until day 26. The duration lasted between 1 minute (he resumed sleep right after waking) and 15 minutes. An average of 5 minutes was noted for the duration of NWs during this period, demonstrating an improvement from baseline. The substantial decrease in frequency of NWs were further noted during from day 26 until lasted until day 59, with only three NWs lasting between 15 to 20 minutes in duration. An average of only 10 minutes was noted throughout this period. It was therefore noted, that NWs were almost eliminated in T2, after the introduction of the social story and gro-clock, with the exception of nights where Tom was noted to be unwell or had the occasional toileting accident.

Follow-up.

Short-term data were collected via the sleep diary. Long-term data are yet to be collated because the study was recently finished. Tom's family was asked to maintain the second part of the treatment (day 26 to 59) treatment protocol but without recording the sleep diaries.

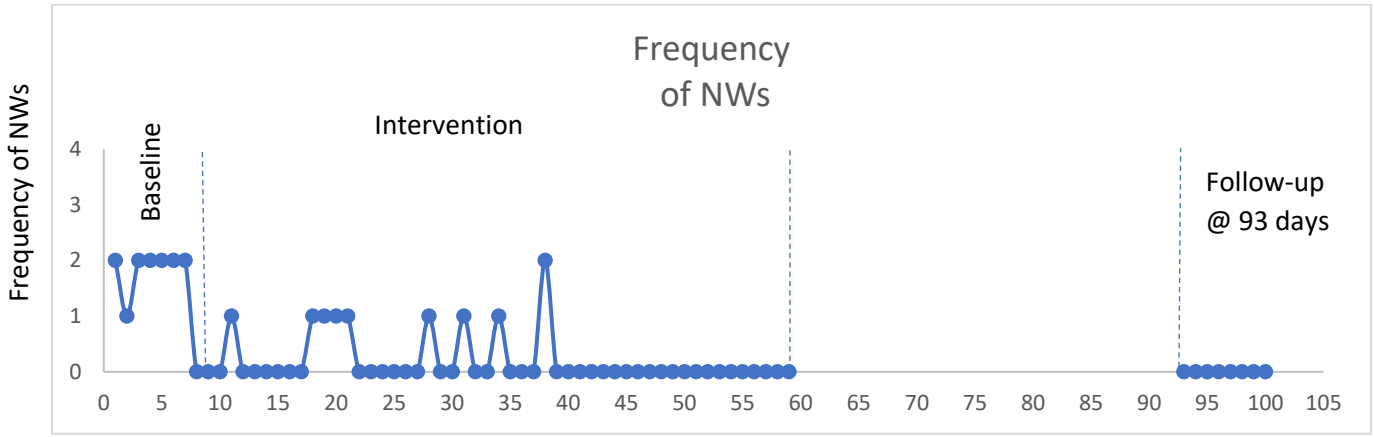

Figure 4: The frequency of NWs during baseline, intervention and short term follow-up

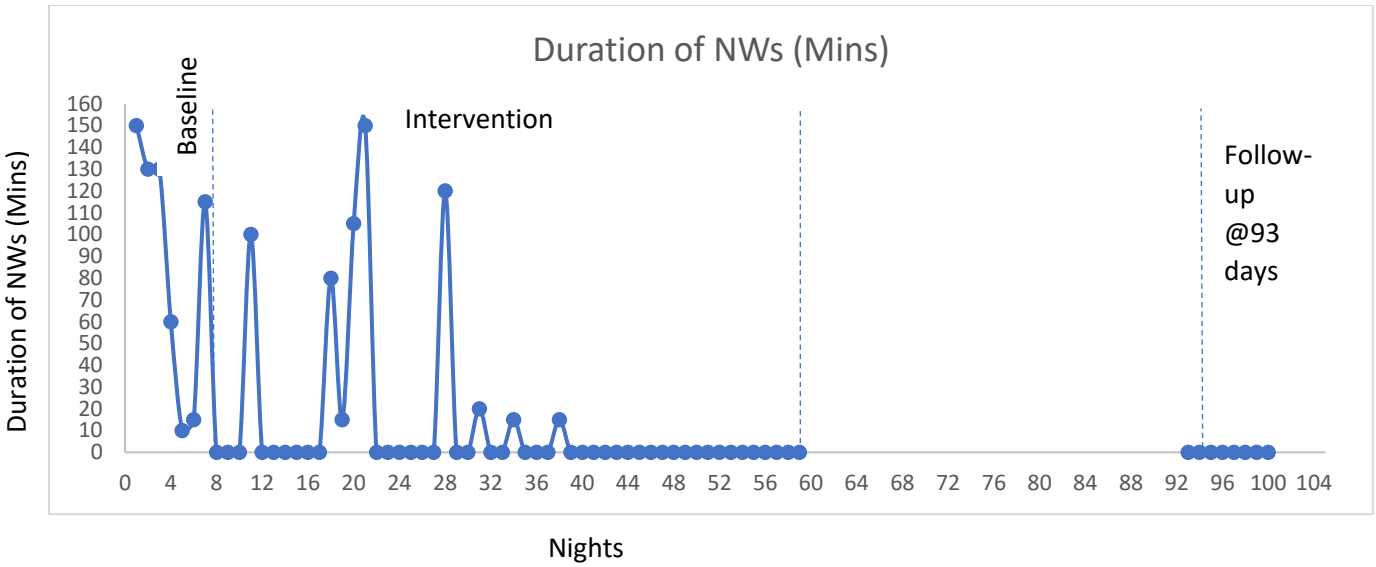

Figure 5: The duration of NWs during baseline, intervention and short term follow up.

As demonstrated in Figure 5, NWs were eliminated at short-term follow-up. His sleep (8pm) and wake times (6.30am) had also achieved consistency. This illustrates an improvement from baseline, where at least two NWs were present every night. Data were not, however, available for long-term follow-up.

Post-treatment interview.

Tom's mother reported that there were dramatic changes in Tom's sleep. Tom's mother mentioned that there was still daytime sleepiness noted, however she attributed it to the busy schedules during school time. He was also still using his social story and gro-clock to help him maintain his routine which indicated that he had adapted well to the changes in his sleep schedules.

Case Study 3

Mike (pseudonym) was an 11- year old boy diagnosed with Down syndrome and ASD. He lived with his parents and two siblings. He was referred to the sleep programme by his paediatrician, to help improve his sleep.

Presenting complaints.

The primary concerns reported by Mike's mother around his sleep, were his night awakenings, sleep-competing behaviour, which involves stereotypy (i.e., vocalising by imitating sounds or noises) and also daytime naps.

Mike presented with NWs, at least once a night, and these were long in duration. His NWs tended to present between 2am to 4am most nights and once awake he did not go back to sleep. He would then sometimes fall back asleep much later, around 8am, after which he would be challenging to wake. Mike was also reported to have frequent daytime naps at school (several times a week). Mike's mother mentioned that his sleep problems seem to deteriorate after being at his grandparents' place (every second weekend), he was also taking melatonin for his sleep, but it had not been very useful in improving his sleep. Mike's mother's goals were for him to go to bed at 9pm and wake up at 7am (which is the time she wakes up).

Family setting and developmental history.

Mike had delayed development, was non-verbal and communicated by physical gestures. Mike was reported to have problems with sleep onset as an infant, which persisted until he was about five years old. Mike's parents made efforts to address his sleep onset (by consistently returning him to his bed) during that period, which improved, however, he had since then started presenting with NWs which has been persisting until date (7 years). He had also not received any interventions for his sleep disturbances.

Case study method.

The clinical interview conducted to gather information related to Mike's sleep, developmental history, family setting, presenting complaints, including medical or physical factors that could contribute to his sleep disturbances or had the potential to interfere with the treatment in any way.

The FBA conducted with the family gathered information regarding antecedent and consequence variables that were interfering with his sleep including the function of these sleep interfering behaviours to help form the intervention. This included information from the clinical interviews, sleep diaries, video content and also data gathered from assessment questionnaires (i.e., SATT, QABF). Information concluded from the FBA helped in formulating a comprehensive treatment plan for Mike. Mike's NWs and his sleep competing behaviours (i.e., stereotypy), were identified to be a primary concern by his mother. She was asked to record sleep diaries during assessment, to aid in establishing the baseline to help monitor if Mike sleeps better through the night after treatment, by the decreasing or eliminating Mike's NW episodes during treatment. It was foreseen that addressing his NWs and eliminating sleep-competing behaviours would improve Mike's overall sleep. Mike's sleep-competing behaviours were referred to any behaviour that interrupted his sleep at night and contributed to his NW.

Mike's mother was asked to record sleep diaries and to respond to them as honestly and as accurately as she could, regarding Mike's behaviours, to be able to obtain accurate data, that would aid the formulation of his individualised intervention plan. These parent-reported sleep diaries included the time he was put to sleep, location of where he was put to sleep, the time and duration of each NW including detailed description surrounding each NW, parental responses to each NW episode, the time at which he resumed sleep and also the time he at which he was awake for the day.

Definition of key terms.

Sleep-competing behaviours. Sleep-competing behaviours included stereotypy such as, repeated vocalising of sounds or noises, accessing tangible items upon waking (i.e., iPad, toys) and also requests for food.

Parental responses. Parental responses to these behaviours include Mike's parents providing him with access to the iPad, co-sleeping with him (either on his own bed or on parent's bed) or providing him with food.

Results of the FBA.

The diaries indicated that Mike did not have any sleep onset issues. It was noted that Mike typically went to bed between 9 and 10pm and was asleep within 15 minutes. His wake times were variable, many of them depending on his NW episodes. He usually woke for the day after his NWs, or fell asleep much later i.e., 8am. His NW timings were also variable although most of them fell between 2am and 4am. His sleep diaries indicated that during his NWs, he would wander into the lounge where he accessed his preferred videos on his iPad or toys. Mike's excitement level would increase from watching videos and he would start vocalising or jumping, which would attract the attention of his parents. Parental responses included trying to regulate his excitement and attempting to get Mike to resume sleep in his own bed or in their bed (co-sleeping). During this time Mike sometimes asked for food. If he fell asleep again around 8am, Mike would stay asleep until mid-morning at the earliest or midday. Mike also had daytime naps most days during the week, with an average of 2 naps per day of 90 minutes to 180 minutes and mainly occurred if he did not resume sleep following a NW.

From the FBA, it was apparent that the antecedent variables that were contributing to Mike's sleep appeared to be a circadian disturbance (his circadian rhythm seemed to be out of sync with the environment and was demonstrated by inconsistent sleep/wake times). His daytime naps were also contributing to his lack of physiological sleep pressure at night. The consequences appeared to the reinforcement received from sleep-competing behaviours such as access to preferred items (such as, playing on his iPad or toys without any restrictions) and

access to his preferred food (via parental responses). The primary factor of concern was that, his NWs seemed to be maintained by the lack of synchronisation between his circadian rhythm and the external environment. The circadian disruption present in Mike's sleep/wake schedules was the most likely cause of his sleep disturbances, with his access to preferred activities or food of his choice following NWs being a secondary factor.

Table 5 represents the identified parent-reported sleep behaviours, the hypothesised functions of disruptive behaviours, factors identified during FBA (as precipitating and/or maintaining the target behaviours and the formulated individualised intervention that was carried out to address the identified problem behaviours. Mike's parent's goals included a) Mike going to sleep at 9pm and sleeping through the night, and if he did wake, to be able to independently put himself back to sleep without leaving the bedroom and, b) to wake at 7am for the day.

Table 5: Parent reported sleep problem/s, factors precipitating and/or maintaining the sleep problem, hypothesised function, and intervention

	Parent reported sleep problem/s	**Factors participating and/or maintaining behaviour**	**Hypothesised function**	**Intervention**
Mike	Frequent NWs	Social attention; access to tangible items, diminished sleep pressure from daytime naps	Attention Tangible Escape from bed	Positive sleep hygiene; Graduated bedtime fading; Social story; Elimination of device use/story reading in bedroom; Positive reinforcement; Consistent sleep-wake times
	Prolonged NWs/ night tantrums	Social attention; access to tangible items, diminished sleep pressure from daytime naps	Attention Tangible Escape from bed	Positive sleep hygiene (including scheduled toileting); Graduated bedtime fading; Social story Elimination of device use/story reading in bedroom; Positive reinforcement; Consistent sleep-wake times

Daytime Napping	Social attention; access to tangible items, diminished sleep pressure from daytime naps	Attention Tangible Escape from bed	Positive sleep hygiene (including scheduled toileting); Graduated bedtime fading; Social story Elimination of device use/story reading in bedroom; Positive reinforcement; Consistent sleep-wake times

Note: NW = Night Wakings

Treatment procedures.

Treatment consisted of a combination of antecedent and consequence-based modifications that were designed to eliminate Mike's NWs. Antecedent modifications included; fading back his bedtime to a later time; restricting daytime naps; establishing set sleep/wake times; helping Mike learn to establish sleep if he wakes, modification to parental responses. Consequence manipulations include positive reinforcement of treatment goals and removing access to preferred items upon waking and changing the parental responses to NWs. More detailed information about each treatment component is provided below. These were put in place to promote more sleep-inducing behaviours and eliminate Mike's sleep-interfering behaviours. Details of each strategy have been described below.

Sleep restriction and delayed bedtime. In order to increase the physiological sleep pressure for Mike to sleep throughout the night and prevent daytime napping, sleep restriction was implemented. The time that Mike was allowed to be asleep was firmly restricted, allowing it to occur only between appropriate sleep hours. In addition to a set, delayed bedtime (11pm), this meant Mike was strictly woken up at the set wake time (7am) in the morning, despite how his night went, and firmly restricted from napping throughout the day. An alarm clock was also introduced to help Mike with a clear indication of the wake-up times. Upon wake-up time, Mike was exposed to bright light, by bringing him out of his bedroom into a brightly lit area to help in making a contrast between night and day.

To reset Mike's circadian rhythm in order to synchronise his circadian rhythm with the environment, a consistent sleep wake time was set. His bedtime had to be delayed by about an hour. The times would gradually be brought forward when he started to fall asleep within 15 minutes of being put to bed. Mike's parents were therefore asked to put Mike to sleep an hour later (11pm), than his usual bedtime (10pm); as it was close to his natural sleep time and more likely for him to initiate sleep quickly (within 15 minutes); and woken at 7am for the day. This was completed in Phase 2 of the intervention once sleep restriction was established.

Positive sleep hygiene. His sleep hygiene routines included restricting access to preferred tangible items (iPad, toys) to only daytime, to accumulate sleep pressure.

Consistent bedtime routine. Mike would complete his usual bedtime routine of brushing his teeth, changing into his pyjamas, and toileting. He would be put to bed while still awake. The intentions were for this to be faded out gradually to encourage Mike learning to manage his NWs on his own. This new routine was to be implemented every night so create a consistency and understanding of what was expected, for Mike.

Social story. Mike was introduced to a social story during the treatment to help inform and regulate the new bedtime routine including model desired behaviour. Mike's parents were asked to read it to him every night before going to bed. The social story, which consisted of texts and photos, consisted of steps for his bedtime routine, including what to do during NWs and the reinforcements received the morning after, for staying in bed throughout the night.

Reinforcement. Reinforcement was delivered immediately upon waking for the day for staying in bed all night. This included tangible rewards and social rewards. Tangible rewards included preferred items (i.e., iPad) and social reinforcement included praise, cuddles, kisses, hugs etc).

Results.

Data pertaining to the frequencies and durations of Mike's NWs and daytime naps across baseline and intervention phases are presented in Figure 6 and 7. CSHQ data gathered from pre and post-intervention, and post-intervention data has also been reported in Table 6.

Sleep diaries.

Mike's treatment lasted for 63 days (excluding short-term and long-term follow-ups). Baseline was recorded for 7 days. There was no data available for short and long-term follow-ups. The duration of NWs and daytime sleep during intervention have been presented in Figure 1.

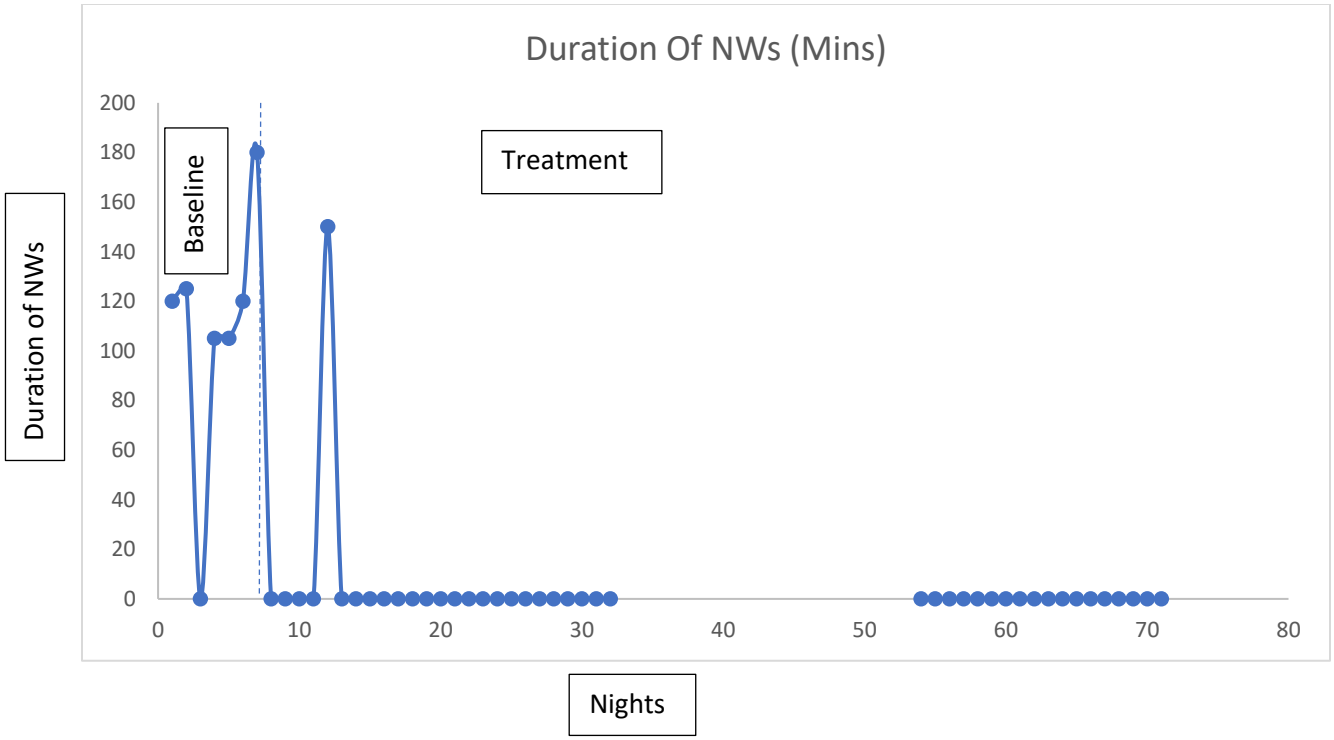

Figure 6: Duration (minutes) of night-awakenings throughout baseline and treatment phases

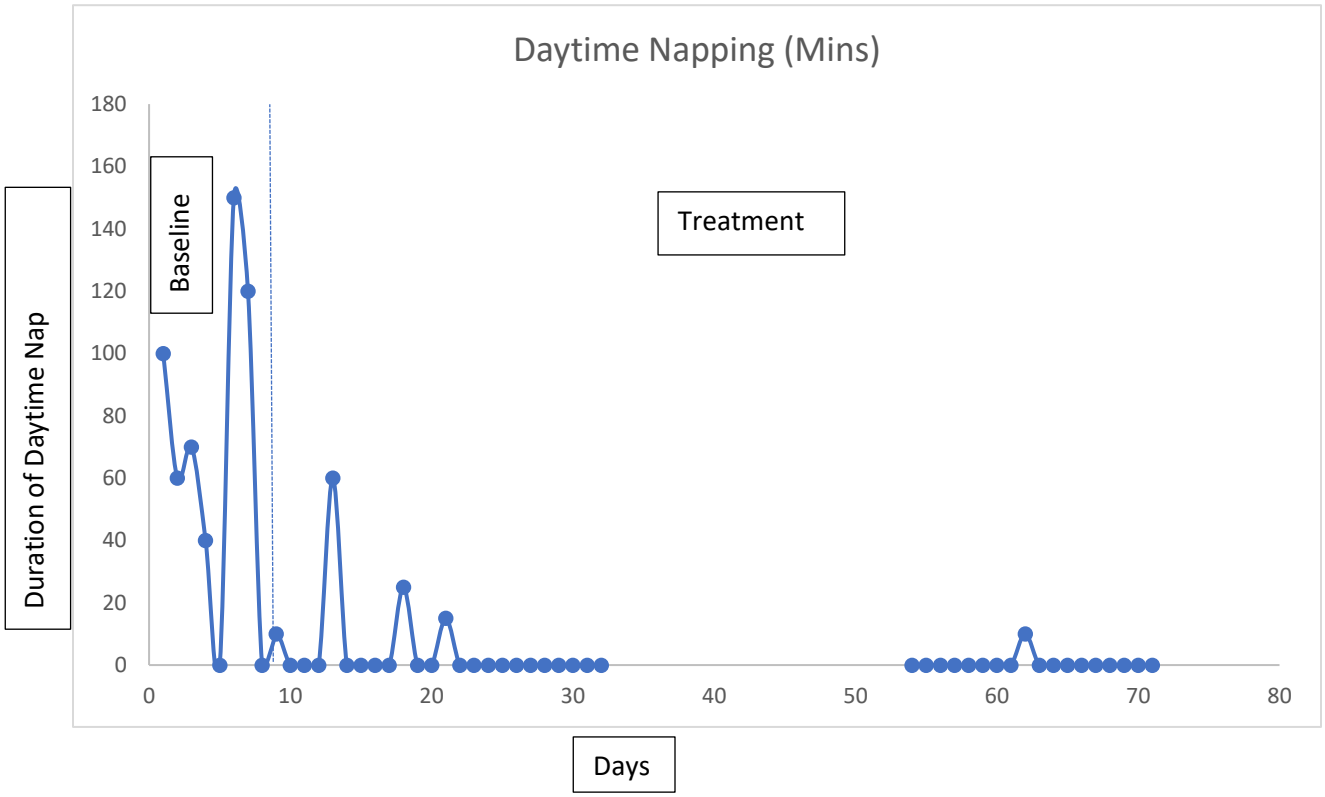

Figure 7: Duration (minutes) of daytime sleep throughout baseline and treatment phases

Night Awakenings.

An immediate difference was noted in his NWs. There was only one episode of NW observed, until day 35 where his bedtime was faded back to 10.45pm. Mike was adjusting well to the new time of 10.45pm and bedtime was brought forward again on day 47, to 10.30pm and again on day 57 to 10.15pm. There were almost no NWs noted throughout the treatment phase. It can be noted in Figure 6, that during baseline, there was a highly variable duration of NWs exhibited, the duration lasting from between 50 minutes to 4.5 hours. There was an average of about 83 minutes throughout baseline. NWs were eliminated upon treatment initiation (except for two nights, on day 35 and 41), and this substantial decrease was consistent throughout the treatment procedures until day 63 (final day of treatment). The first NW (day 35) lasted for about 210 minutes. The duration of the NW on day 41 was however far briefer at only 15 minutes. It was therefore concluded that NWs were almost eliminated by the end of treatment. Frequencies of NW were also noted to occur only once on nights they occurred, therefore not graphed.

Daytime Sleep.

During baseline, Mike demonstrated highly variable duration of daytime naps. The duration of daytime napping episodes lasted from between 60 minutes to 350 minutes with an average duration of 100 minutes, but since treatment started there was a reduction in daytime naps; they were almost eliminated, with the exception of four occasions. At 15 minutes three of these were very brief and were at the start of the treatment, and there were no daytime naps during the final 20 days of treatment.

The results from CSHQ are indicated in Table 6. There was a noted decrease in NWs from 6/9 to 3/9 from pre-intervention to post-intervention, and daytime sleepiness from 13/24 to 10/24, which demonstrated the decrease in frequencies of daytime sleepiness. The difference between the overall total scores of pre- (score=54) and post-intervention (score-40), reflects an overall decrease in Mike's sleep disturbances.

Table 6: Comparison of Pre-and Postintervention Scores on the CSHQ Completed by Mike's Mother.

Variable Scores	Preintervention	Postintervention	Maximum score
Daytime sleepiness	13	10	24
Night awakenings	6	3	9
Total scores	54	40	

Note. CSHQ=Children's Sleep Habits Questionnaire

Results from the TARF-R are presented in Table 7. The TARF-R was completed by Mike's mother post-intervention. Results indicate an impressive overall total score for level of treatment acceptability with 109 out of 119. Mike's mother rated reasonableness, effectiveness, and willingness to participate highly across these variables (21/21) and reported a good understanding of the treatment process (7/7). She also reported it to be very cost-effective (14/14) but that felt the treatment had side effects (18/21). She reported Mike's sleep disturbances to have medium severity (8/14). It was evident from the scores that Mike's mother was satisfied with the treatment procedures and outcomes.

Table 7:Postintervention Treatment Acceptability Ratings on the TARF-R

Variables Score	Mother	Maximum
Total Accessibility	109	119
Reasonableness	21	21
Willingness	21	21
Cost	14	14
Side-Effects	18 (reverse= 21)	21
Effectiveness	21	21
Disruption/Time	14	21
Problem severity[a]	8	14
Understanding of Treatment[a]	7	7

Note. TARF-R= Treatment Acceptability Rating Form-Revised.

[a]Not Included in total treatment acceptability score.

Post-treatment interview.

Mike's mother noted an overall improvement and consistency in elimination of NWs and daytime sleepiness, and an overall improvement in his behaviour, mood and sleep schedules. She reported that he had adjusted well to the new routine and she was confident in continuing with the new routine. She was overall very pleased with the treatment outcomes.

Chapter 4

Discussion

The aim of the study was to improve the sleep of children with DD, who presented with circadian-like components and to investigate the effectiveness of interventions that included circadian components. The interventions were informed by the outcomes of FBA and included multiple components, the common component being circadian. The current study provides evidence to support the use FBA-based interventions to treat sleep disturbances in children with DD. However, the question to be solved is, whether the changes that came about in the children's sleep can be attributed to their individualised intervention plans?

The intervention involved designing individualised behavioural interventions for each child based on the results of the FBA. Identifying factors maintaining the children's circadian disruptions (e.g., daytime napping) helped determine the components of the intervention. Common components of intervention included fading bedtimes, positive sleep hygiene, and a consistent bedtime routine. These interventions resulted in an elimination of SOD for Dexter and elimination of NWs for both Tom and Mike.

Data from the sleep diaries indicated improvements across the variables (i.e., reduction in SOD, CCs, NWs, day time naps) related to the participants' sleep disturbances from pre- to post-intervention. Parent-reported CSHQ results also showed similar improvements following treatment.

Dexter.

There were several factors to be taken into consideration with Dexter's intervention. The initial observation was that Dexter's SOD was being reinforced by the lack of accumulated sleep pressure and caffeine consumption. The sleep problems were still occurring during baseline, however, was eliminated as a result of intervention. It appeared, that addressing Dexter's caffeine intake prior to intervention had created a huge change in Dexter's overall sleep throughout the intervention. This can be ascertained by the change reflected from baseline to the intervention including the consistency of this change that was maintained during short and long-term follow-

up. Dexter started showing a decrease in SOD in baseline, after his Coca Cola consumption had decreased and therefore, there was lack of evidence of clear treatment effects, other than caffeine reduction with regard to the consistency of the changes demonstrated in Dexter's SOD. Although Dexter did show changes, as demonstrated by his post-treatment scores at follow-up or long-term, changes observed before the introduction of treatment suggested that improvements were not likely attributable to the treatment effects, because there was a clear response to reduced caffeine, which occurred baseline, which was still maintained to be very low. Dexter's CSHQ scores following treatment, which correlated to the data in his sleep diaries regarding his SOD, indicated that the decrease demonstrated in baseline was maintained throughout until post-treatment.

Tom.

Based on the data, it appears that treatment resulted in substantial reduction in the duration and frequencies of NWs. Tom's NWs had been almost eliminated after introduction of the treatment, with the exception of some nights in which he needed a nappy change. On occasions, his sleep resumed quickly. Tom's intervention plan included establishing a consistent bedtime routine with set sleep and wake up times including pushing back his bedtime to a later time. The intervention included removing variables (such as, gadgets, power cables, story reading) that were potentially influencing his sleep and increasing physiological sleep pressure at night by keeping him awake in the day. Making these changes to his lifestyle aided in improving Tom's sleep; by reducing his NWs. This further reiterates that factors around the child's lifestyle should be taken into consideration when identifying influencing variables for sleep disruption in children with DD.

Mike.

The data demonstrated that there was a substantial decrease in the duration and frequencies of NW and daytime naps after treatment. There were clear treatment effects after treatment initiation, in eliminating his NWs (excluding the night he was unwell). This reduction in NWs was observed even on nights Mike did not sleep at home (i.e., at his grandparents). Mike's intervention plan consisted of setting a consistent bedtime routine and positive sleep hygiene. These strategies were implemented by restricting the number of hours Mike could sleep to be only between appropriate hours and eliminating any daytime naps. This accumulated sleep

pressure by night-time. There was also removal of variables (i.e., gadgets, toys) that were accessible and interfered with his sleep.

All three children demonstrated significant improvements from their identified problem behaviours which were consistent at maintenance and follow-ups. Graduated bedtime fading is a well-established effective treatment for addressing sleep problems such as SOD or NWs, in children with DD, due to its nature of accumulating sleep pressure for the child. The procedure of resetting their circadian rhythm, increases the pressure for consolidated sleep and also increases the probability of quick sleep onset. Graduated bedtime fading was therefore utilised within Dexter and Tom's treatment plan, and both, bedtime fading and sleep restriction was utilised in Mike's treatment plan, to attempt to effectively treat their sleep disturbances, which presented with circadian-like components.

Efficacy of using FBA.

Using FBA to formulate individualised treatment plans can be credited with the elimination or at least the reduction in sleep disturbances for all three participants, because it allowed for the identification of specific sleep interfering behaviours for each child. These precipitating factors that contributed to and maintained the behaviour were then addressed according to the individual child and family's goals, needs, and preferences, and this led to overall improvements in their sleep. For Dexter, FBA aided in identifying caffeine consumption as a possible contributing factor that was maintaining his SOD, which was then addressed accordingly. Mike's sleep interfering behaviour was identified via FBA, to be due to a lack of accumulated sleep pressure and reinforcements that were maintaining his NWs (i.e., access to gadgets, toys, food). It should, however, be important to establish if their sleep disturbances were a result of a disturbance in their circadian component or due to other factors. The child's sleep problems may present with circadian components, but once treated, using a circadian intervention, they might be left with other symptoms, that may need a non-circadian explanation. There are a number of external factors that can result in disturbances to the sleep/wake cycles (such as, caffeine consumption). This means it is necessary to very carefully assess and treat the various factors that can contribute to this. For example, Dexter's sleep disturbances presented as a circadian disturbance, but was instead addressed successfully during baseline with the decrease of caffeine in his diet and no new sleep

interfering behaviours were demonstrated. Therefore, it was established that Dexter's circadian disruption was likely contributed to by his caffeine consumption. There is a limitation, however, in terms of the conclusion that can be drawn, regarding the effects of circadian interventions alone and further research is warranted.

Another benefit of using FBA, its flexibility also allows for instances of external factors (i.e., illness) interfering with the treatment, to be taken into consideration. The current study demonstrates how treatment components can be implemented in a sequential manner, according to behaviour function, so as to aid families in carrying out the intervention more effectively without the overwhelming need to engage in multiple strategies at once. An example can be seen from Mike's treatment plan, where it was initiated with sleep restriction first (to aid in consolidation of sleep), before implementing faded bedtime (to reach goal bedtime) and removal of access to tangible and non-tangible items including parental presence. This allowed for Mike and his parents to get used to the first phase in the routine and prepare for undesired responses before proceeding to add another change. While this proved successful in eliminating Mike's NWs, implementing a multi-component intervention may have produced even quicker results (saving time). The current study demonstrates the importance of using FBA to assess sleep disturbances in children with DD rather than succumbing to default interventions (Singh & Zimmerman, 2015).

The unique element about FBA is, despite children showing similar presenting problems, FBA- based treatments individually address antecedent and consequence variables with the goal of removing reinforcements for sleep interfering behaviours and increasing adaptive alternatives of sleep. Efficacy demonstrated from these treatments advocates the importance of utilising FBA to assess sleep difficulties in children with DD instead of relying on default treatments and also, supports that the theory that factors precipitating and/or maintaining sleep difficulties are common in children with DD (Spruyt & Curfs, 2015). FBA's effectiveness of addressing challenging behaviours is well-established, and the results from this study further advocates it, while adding on to the lack of literature surrounding the use of FBA to formulate treatments for sleep difficulties in children with DD.

It was noted as well, that parents reported positive experiences from the intervention programme, except the minor considerations (understandable and expected) of the time and effort required. It has been reported that behavioural interventions that alter parental perceptions of control and efficacy with their children, and provide them more confidence and autonomy, may enable them to regulate their child's sleep behavior, including helping their child develop self-regulation (Johnson & McMahon, 2008).

In addition to behavioural treatments, interventions should also consider modifications to sleep/wake schedules as this has been proven as vital in addressing sleep disturbances with circadian components (Piazza & Fisher, 1991). This is important, as modifying the child's sleep/wake schedules may contribute to the accumulated increase in physiological sleep pressure that takes place when the child is put to bed at their later than normal time of sleep onset. This accumulated sleep pressure has been demonstrated to facilitate quick sleep onset and decrease the chances of CCs or NWs. Another element to this intervention is the establishment of consistent sleep-wake times which allows the body's circadian rhythm to get synchronised with the set sleep-wake schedule. Having a regular sleep-wake cycle in sync with the body's circadian rhythm may further facilitate the graduation of bedtimes as lack of sleep pressure is no longer a factor.

Efficacy of circadian interventions.

The results of this study replicate those of Ashbaugh and Peck (1998) where they support the efficacy of circadian interventions to improve sleep disturbances in children with DD. The results of the present study also extend those of Piazza and Fisher (1991), by indicating that procedural modifications could be made without affecting the efficacy of the treatment. Lack and Wright (2007) also promoted the use of circadian intervention to address sleep problems, especially those that present with SOD. Durand and Christodulu (2004) similarly advocated the use of circadian interventions for children with DD. They were able to eliminate bedtime resistance and NWs in two children with DD. They also reported overall parental satisfaction and easy use of the intervention (to be able to use it on a regular basis).

The following section aims to establish if the aims of the current were helpful in guiding the research. The first research question, as presented earlier, explores to what extent circadian features are evident in the presentation of children with DD and sleep problems. In all three cases, participants presented with circadian components in their sleep disturbances but after the initiation of treatment, it was observed that Dexter's sleep disturbances were mainly attributed to the caffeine consumption in his diet, which in turn presented as circadian components. Once his caffeine consumption had been addressed, his circadian-like symptoms saw an improvement as well. Tom and Mike's sleep were, however, demonstrated to be eliminated due to treatment effects, indicating their sleep problems to be due to a disruption in their circadian rhythms. But, for both participants, it was unclear if their sleep disturbances were directly the result of a circadian component, because there were other concurring treatment strategies implemented. More research would be warranted to establish the extent to which circadian features play a role in the presentation of children with DD and their sleep problems.

With regards to the second question, the results of the current study support the efficacy of using FBA–based interventions on children with DD who presented with circadian-related components within their sleep disturbances. Moreover, the results add to the literature surrounding circadian-sleep problems in children with DD. The FBA helped identify features of their sleep disturbances and used a specific combination of procedures tailored to meet the needs of the child such as, set sleep-wake times, social stories and rewards systems and the like. Effects from the individualised plan (directly – treatment, or indirectly –Coca Cola consumption) were noted in both children and their identified problems were addressed. This therefore demonstrates that FBA has the ability to develop treatment plans for children with DD and using FBA might increase the probability of efficacy of the intervention, due to only addressing and targeting specific problem behaviour. The current study adds to the existing pool of literature that FBA-based interventions are an effective method of identifying and treating the presenting circadian-related sleep interfering factor/s in a child with DD.

The third research question, was it sufficient to treat the child's sleep disturbances? All three children completed their FBA–based interventions successfully. The improvement noted in Dexter's sleep was attributed to his decrease in Coca Cola consumption despite presenting with

circadian components. There were similar mechanisms behind his sleep problems to, but not exactly, a circadian disturbance but he still benefitted from the treatment because of his individualised FBA–based intervention. Tom however, demonstrated that he had directly benefitted from his FBA–based intervention plan. Pushing back his bedtime to a later time (to realign his circadian rhythm) resulted in the successful elimination of his NWs, as demonstrated in all his sleep data. It was therefore established, that Tom's early sleep timings were likely causing a disruption between his circadian rhythm and the environment thereby resulting in his NWs. Similarly, Mike demonstrated direct treatment effects from his FBA–based intervention. Restricting his night-time sleep and daytime napping, coupled with a faded bedtime strategy (to meet his goal bedtime and wake-time), proved efficient in eliminating his NWs. It was therefore clear that a lack of physiological sleep pressure was causing a disruption in his circadian rhythm with the environment as was evidenced by his NWs and therefore, addressing the root cause of his sleep disturbance proved successful in eliminating his NWs.

The final research question used to guide the current study was, how the current study was able to improve the overall wellbeing of the child and their families. The utilisation of FBA tailored the intervention according to the developmental history and home environment of the child therefore methods of implementation were formulated with consideration given to the child, their families and the feasibility with minimal disruption. Tom's mother reported having more sleep and regaining some intimate time with her partner after the elimination of his NWs. Dexter's mother also reported feeling less stressed, having more personal time and receiving more sleep following intervention due to the improvements noted in Dexter's sleep onset and lack of CCs. These findings further intensify the success around using FBA-based interventions to treat the population of children with DD for their circadian sleep disturbances.

Limitations and future directions.

Limitations or complicating factors to this study may have had an impact on the treatment procedure. In Dexter's case he did not have toilet training at the time of the start of the study and therefore he had to have nappy changes on some nights which may have possibly interfered with the treatment. A regular toileting strategy was however established in response to this. Effort was

therefore made to eliminate or limit contingencies by maintaining a regular toileting schedule before bed (i.e., integrated within his bedtime routine).

Although the use of individualised treatment packages has been demonstrated to be beneficial in addressing specific sleep disturbances faced by a child with DD, it also makes it difficult to generalise the study to other children with DD even if they presented with similar symptoms, due to the treatment being specifically tailored to meet the needs of that particular child. It is also not certain, that the sleep problems experienced by the children in the current study are representative of the population of children with DD.

Another limitation is how the current study explores the efficacy of using circadian interventions to treat sleep disturbances in children with DD, who present with circadian-related components, but other treatment components were also used. Because this is a pilot study, more subsequent research needs to systematically evaluate circadian interventions on their own. Future research encompassing the extent to which circadian interventions may influence or play a role in treating sleep disturbances in children with DD is therefore warranted. The current study also includes a small number of participants with DD experiencing circadian sleep disturbances, and this may limit generalisability to other children with DD, who experience sleep disturbances, and this too requires future research.

This study could also be received better if it had a direct and systematic replication to extend the generality of this study's findings to a wider range of ages, capabilities and developmental disabilities. This study has successfully established that FBA is a key foundation to formulating individualised sleep interventions for children with DD. It might also be useful for future research to consider measuring the power of the environment and its ability to affect physiological effects in children with DD, who present with a sleep circadian disruption within future research. Finally, the current study can be added to the existing pool of research surrounding the use of FBA to inform the efficacy of behavioural interventions concerning sleep difficulties with presenting circadian components in children with DD.

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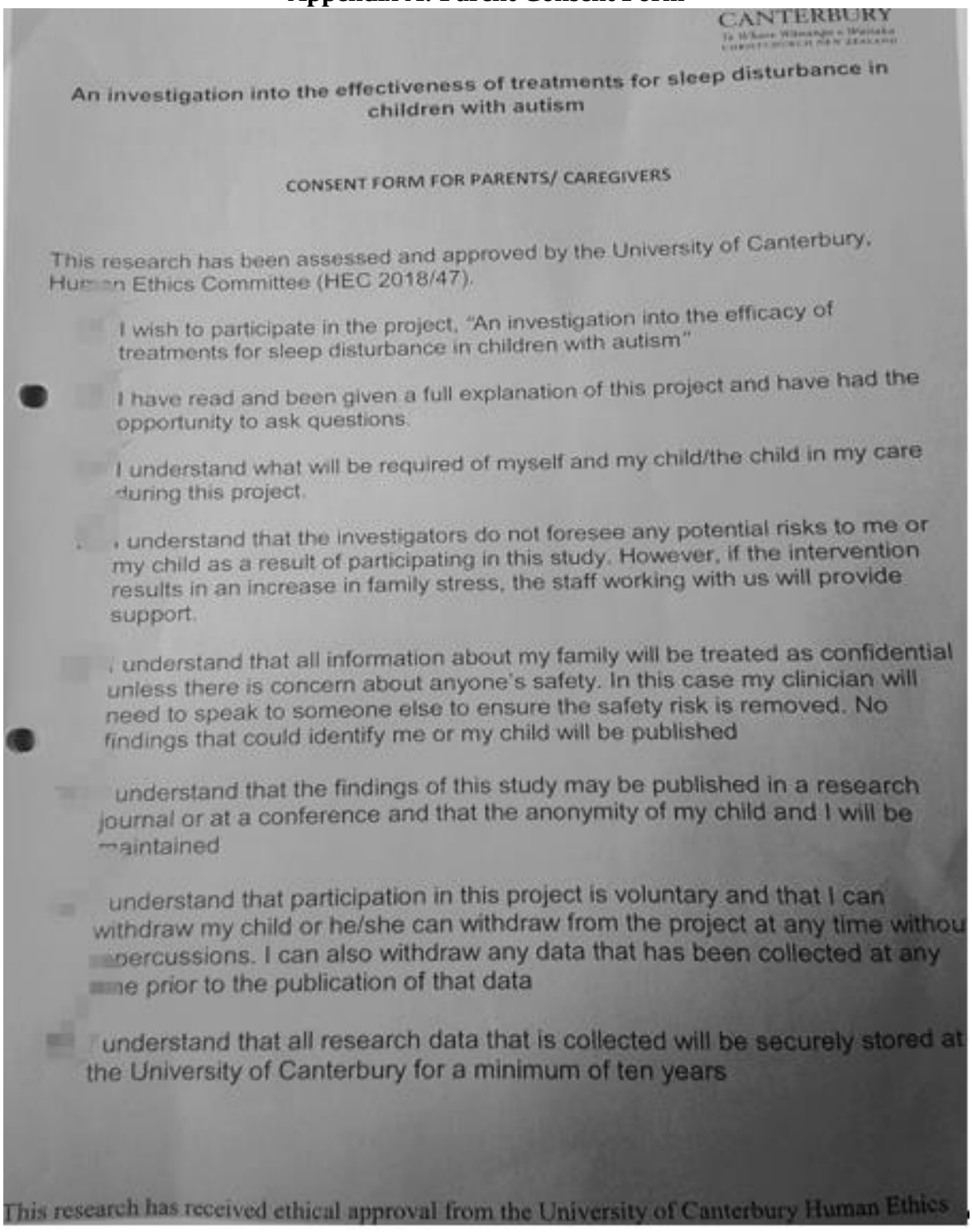
Appendices

Appendix A: Parent Consent Form



CANTERBURY

An investigation into the effectiveness of treatments for sleep disturbance in children with autism

CONSENT FORM FOR PARENTS/ CAREGIVERS

This research has been assessed and approved by the University of Canterbury, Human Ethics Committee (HEC 2018/47).

I wish to participate in the project, "An investigation into the efficacy of treatments for sleep disturbance in children with autism"

I have read and been given a full explanation of this project and have had the opportunity to ask questions.

I understand what will be required of myself and my child/the child in my care during this project.

I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study. However, if the intervention results in an increase in family stress, the staff working with us will provide support.

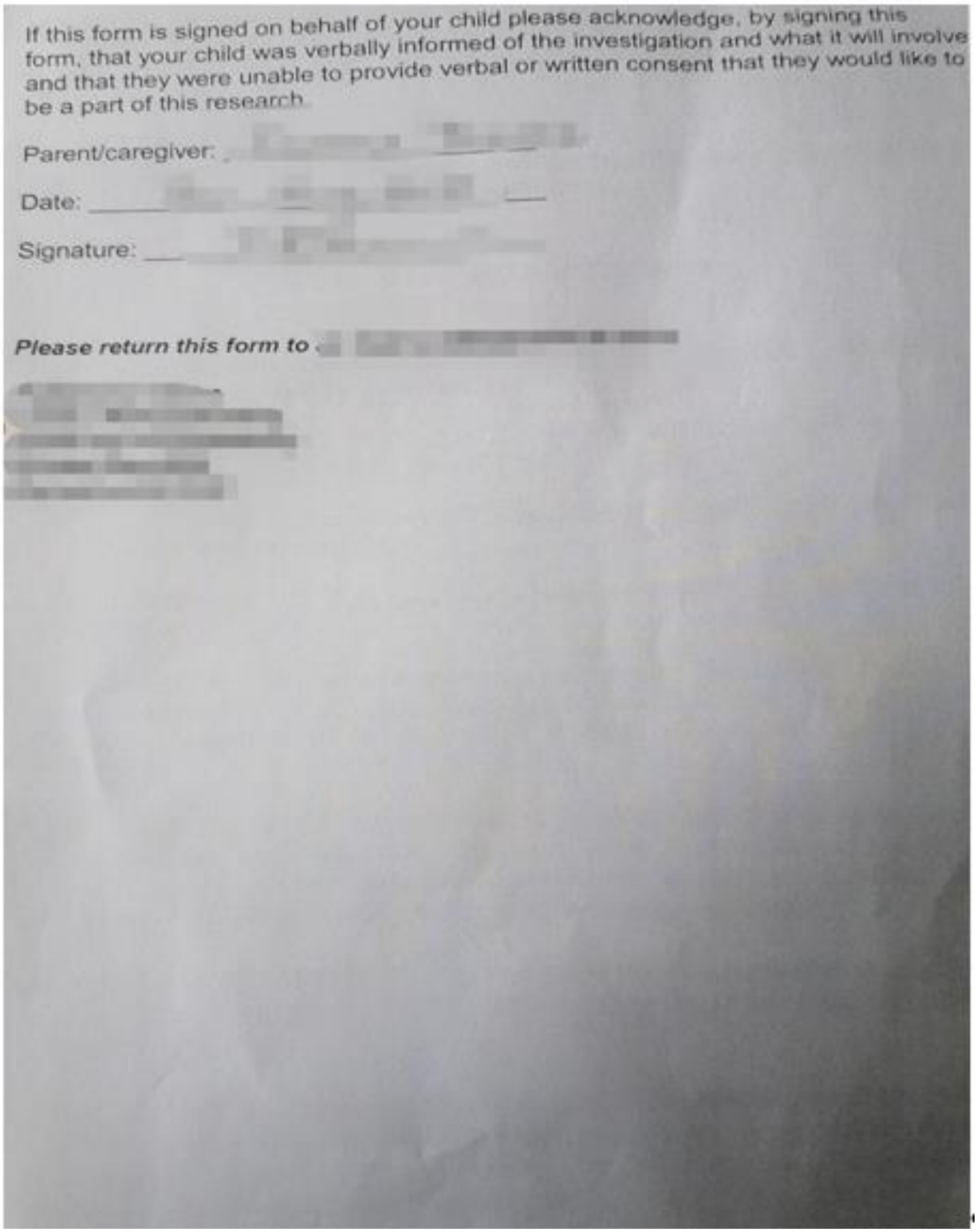
I understand that all information about my family will be treated as confidential unless there is concern about anyone's safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published

I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of my child and I will be maintained

I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data

I understand that all research data that is collected will be securely stored at the University of Canterbury for a minimum of ten years

This research has received ethical approval from the University of Canterbury Human Ethics



If this form is signed on behalf of your child please acknowledge, by signing this form, that your child was verbally informed of the investigation and what it will involve and that they were unable to provide verbal or written consent that they would like to be a part of this research

Parent/caregiver: _____

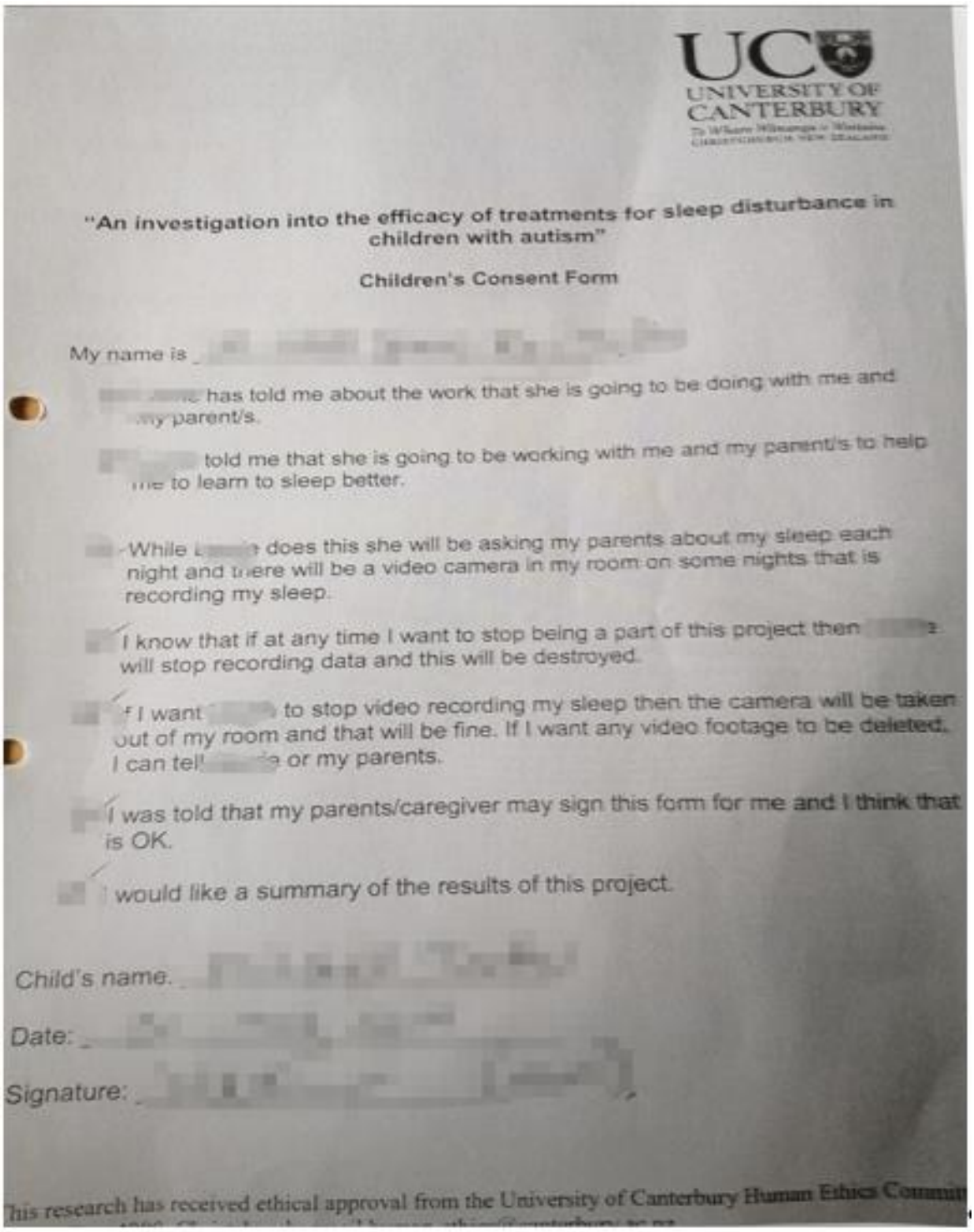
Date: _____

Signature: _____

Please return this form to [redacted]

[redacted]

Appendix B: Child Consent Form



UNIVERSITY OF CANTERBURY

"An investigation into the efficacy of treatments for sleep disturbance in children with autism"

Children's Consent Form

My name is _____

has told me about the work that she is going to be doing with me and my parent/s.

told me that she is going to be working with me and my parent/s to help me to learn to sleep better.

While she does this she will be asking my parents about my sleep each night and there will be a video camera in my room on some nights that is recording my sleep.

I know that if at any time I want to stop being a part of this project then she will stop recording data and this will be destroyed.

If I want to stop video recording my sleep then the camera will be taken out of my room and that will be fine. If I want any video footage to be deleted, I can tell her or my parents.

I was told that my parents/caregiver may sign this form for me and I think that is OK.

I would like a summary of the results of this project.

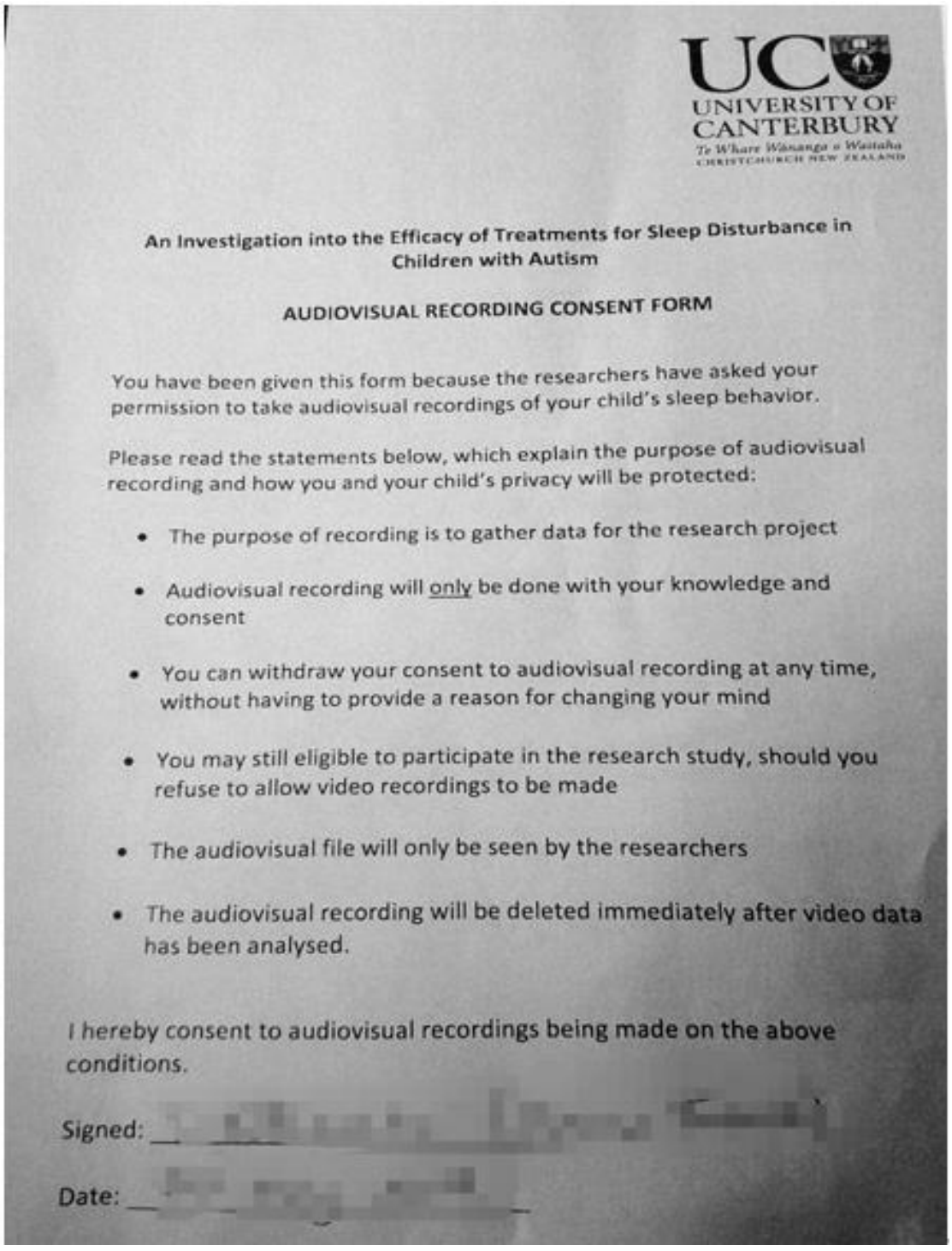
Child's name. _____

Date: _____

Signature: _____

This research has received ethical approval from the University of Canterbury Human Ethics Committee

Appendix C: Parent Audiovisual Recording Consent Form



UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An Investigation into the Efficacy of Treatments for Sleep Disturbance in Children with Autism

AUDIOVISUAL RECORDING CONSENT FORM

You have been given this form because the researchers have asked your permission to take audiovisual recordings of your child's sleep behavior.

Please read the statements below, which explain the purpose of audiovisual recording and how you and your child's privacy will be protected:

- The purpose of recording is to gather data for the research project
- Audiovisual recording will only be done with your knowledge and consent
- You can withdraw your consent to audiovisual recording at any time, without having to provide a reason for changing your mind
- You may still eligible to participate in the research study, should you refuse to allow video recordings to be made
- The audiovisual file will only be seen by the researchers
- The audiovisual recording will be deleted immediately after video data has been analysed.

I hereby consent to audiovisual recordings being made on the above conditions.

Signed: [redacted]

Date: [redacted]

Appendix D: Sleep Diary Template

	Date:	Monday:	Tuesday:	Wednesday:	Thursday:	Friday:	Saturday:	Sunday:
Daytime sleep	Setting (where fell asleep)							
	Time asleep							
	Time awake							
Night-time sleep	Setting (where fell asleep)							
	Time put to bed							
	Frequency of Curtain calls*							
	Curtain calls after put to bed (Describe each)							
	Your responses to each curtain call (Describe each)							
	Best estimate of time asleep							

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
1st Night time awakening	Time & Duration of awakening	____ mins	____ mins	____ mins	____ mins	____ mins	____ mins	____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
2nd Night time awakening	Time & Duration of awakening	____ mins	____ mins	____ mins	____ mins	____ mins	____ mins	____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
Time awake in the morning								

*Curtain calls: Any bids for parent attention (e.g. calling parents into the room, leaving the room to ask a question) between the time of being put to bed and falling asleep

Notes: