

AN INVESTIGATION INTO THE EFFECTIVENESS OF FUNCTIONAL
BEHAVIOURAL ASSESSMENT BASED INTERVENTIONS FOR SLEEP
DISTURBANCE IN CHILDREN WITH RARE GENETIC NEURODEVELOPMENTAL
DISORDERS (RGND)

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Abstract

Sleep disturbance is recognized in the literature as a highly prevalent feature of Rare Genetic Neurodevelopmental Disorders (RGND) in children. Despite the considerable number of studies that have reported the presence of sleep difficulties within this cohort, limited attention has been paid to the treatment of these sleep problems, and even less so investigating the effectiveness of FBA-based behavioural interventions, and the outcomes of such treatment methods on general child behaviour and parental wellbeing. The purpose of this study was to determine whether FBA-based behavioural sleep interventions are effective with children with RGND, to examine the impact that intervention may have on secondary outcome variables pertaining to general child behaviour and parental wellbeing, sleep, and relationship satisfaction, and to ultimately establish the acceptability of this form of treatment amongst parents. Three children aged between six and 12 took part in the study. The study followed a multiple-baseline-within-participants design and included measures taken at pre- and post-intervention time-points. The FBA-based behavioural interventions were found to be effective in the treatment of the children's sleep difficulties overall, with improvements observed across sleep onset latency, night wakings, and co-sleeping. General child behaviour, as measured by the VABS-II and the CBCL at pre- and post-treatment, had variable patterns of change across participants and across specific variables. Similarly, parental wellbeing, sleep, and relationship satisfaction, as measured at pre- and post-treatment by the DASS-21, PSQI, and the RQI, were mixed in their results. Despite the variability seen in these findings, the parents of all three children rated their acceptability of treatment highly on the TARF-R, and their post-treatment interviews reiterated their satisfaction with their child's intervention, most aspects of the study process, and the secondary outcomes that they noticed. The present findings contribute to the limited existing literature concerning the utilization of FBA-based behavioural sleep intervention with children with RGND, and the secondary effects of treatment on general child behaviour and parental wellbeing following such interventions. However, future

research should endeavour to address and rectify the limitations encountered in the current study, in order to further the evidence-base to an even greater extent.

Chapter 1

Rare Genetic Neurodevelopmental Disorders and Sleep

Sleep disturbance is recognized in the literature as a highly prevalent feature of Rare Genetic Neurodevelopmental Disorders (RGND) in children. A number of studies have reported the presence of sleep difficulties within this cohort, with between 30 to 90% of children with Angelman syndrome, Prader-Willi syndrome, Fragile X, Rett syndrome, and Williams syndrome experiencing some form of sleep disturbance (Annaz et al., 2011; Kronk et al., 2010; Richdale et al., 1999; Summers et al., 1995). Insomnias, including difficulties with sleep onset latency, night wakings and early morning wakings appear to be relatively consistent manifestations of sleep disturbance across the aforementioned syndromes (Annaz et al., 2011; Bruni et al., 2004; Cassidy et al., 1990; Kronk et al., 2010), however presentations do vary considerably and some sleep problems (e.g., excessive daytime sleepiness) occur more frequently among those with specific disorders (Hoban, 2000).

Behavioural interventions including antecedent-based and consequence-based procedures such as sleep hygiene and modification of bedtime routines, faded bedtime schedules, and extinction have considerable empirical support in the treatment of sleep problems among typically developing children (Johnson & Mindell, 2011; Owens, Palermo, & Rosen, 2002) and children with autism spectrum disorder (Turner & Johnson, 2012; Vriend et al., 2011). However, treatment studies investigating the effectiveness of these approaches with children with RGND are scarce. Research wherein behavioural strategies have been used, report positive treatment effects and henceforth provide strong support for further research in this area.

The current study will extend the current corpus of literature by assessing the effectiveness of FBA-based, behaviourally-based interventions for sleep problems in children with RGND.

In this chapter, a description of RGND is provided, along with the other challenging behaviours that are typically present in these disorders. The prevalence, type, and cause of sleep problems in children with RGND are also detailed in this section. Finally, the chapter concludes with a summary of the interventions frequently used in the treatment of sleep disturbance, and the functional behaviour assessment (FBA) framework that can be used to guide the selection of such interventions. Where appropriate, for the purpose of brevity, particular attention will be paid to Fragile X syndrome and Prader-Willi syndrome, as these are the diagnoses of two of the three children that took part in the present study. The third child presented with a unique chromosomal abnormality that is not associated with any labeled RGND.

Rare Genetic Neurodevelopmental Disorders

Definition. Genetic disorders are caused by “dysfunctional gene behaviour” or a mutation in the genome (World Health Organization, 2019). The structure and number of gene abnormalities determines the phenotype or observable characteristics of a disorder. Abnormalities may be typified by a single gene mutation, or may involve the addition or deletion of whole chromosomes (World Health Organization, 2019). Given the great multitude of possible abnormalities, there is suitably a multitude of genetic disorders. Rare genetic neurodevelopmental disorders (RGND) are a group of conditions that fall within this categorization, resulting from some form of genetic mutation which in turn disrupts typical human growth and development, particularly as it pertains to the nervous system

(Kindsvatter & Geroski, 2014). These conditions usually have an early onset, and as such the characteristics of these disabilities begin to become apparent during childhood and tend to persist (American Psychiatric Association, 2013). The European Union recognizes a disorder as being rare if it affects less than 5 in 10,000 of the general population (European Commission, 2008), and these syndromes have been determined as “rare” by the National Organization for Rare Disorders (EUROCAT, 2012). While there are no formal statistics to represent the prevalence of RGND as a group, it is known that 1-2% of the general population are diagnosed with neurodevelopmental disorders (Grigg-Damberger & Ralls, 2013), suggesting an even lower occurrence for those deemed “rare”. The various genetic mutations of RGNDs are expressed phenotypically in the form of physiological, intellectual, and developmental impairments (Davis et al., 2018). Additionally, the acquisition of motor, social, language, and cognitive skills is typically inhibited to varying extents due to abnormal brain function (Jeste, 2015). While physical and behavioural presentation can differ markedly depending on the individual chromosomal profile, each syndrome does include certain traits that are recognized as being characteristic of their RGND.

Syndromes diagnosed in study participants.

Fragile X syndrome. Fragile X syndrome (FXS) is caused by a mutation on the FMR-1 gene on the X chromosome, (Wheeler et al., 2015) resulting in a reduction in FMR1 protein (FMRP) levels (del Hoyo Soriano et al., 2018). Given that the X chromosome is implicated in this condition, females are not as frequently affected as males, and in the cases in which females are diagnosed, the symptoms are often more mild (del Hoyo Soriano et al., 2018). It has been found that between 1 in 3717 and 1 in 8918 people of European descent have Fragile X syndrome (Crawford et al., 2001). Physical features that typically present in individuals with Fragile X syndrome include an elongated face, prominent jaw, large ears, macroorchidism,

macrocephaly, flat feet, a narrow and highly-arched palate, and hyperextensible joints (Kidd et al., 2014; Schwarte, 2008).

People with Fragile X syndrome often present with a mild to severe intellectual disability (Bailey, Raspa, Olmsted, & Holiday, 2008; Roberts, Mirrett, P., & Burchinal, 2001; Roberts, McCary, Shinkareva, & Bailey, 2016), executive functioning, attention, and visual-spatial recognition difficulties, hyperactivity, and impulsivity, and seizure disorders (Kidd et al., 2014; Sullivan et al., 2007; Wheeler et al., 2015). There are also some overlapping features that are shared between FXS and autism spectrum disorder (ASD), such as difficulties with language and communication, poor eye contact, abnormal responses to sensory stimuli, and stereotypic behaviours (Kaufman et al., 2004; Kidd et al., 2014; Klusek, Martin, & Losh, 2014a). Accordingly, approximately 60% of males and 30% of females that receive a diagnosis for FXS also meet criteria for ASD (Garcia-Nonell et al., 2008; Klusek, Martin, & Losh, 2014a).

Prader-Willi syndrome. Prader-Willi syndrome (PWS) is a neurodevelopmental disorder that is caused by the non-functioning or absence of the paternally inherited genes at 15q11.2-q13 (OMIM #176270) (Goldstone et al., 2008). People with PWS often present with a mild to moderate intellectual disability, behavioural abnormalities, and a distinctive physical phenotype (Tunnicliffe, Woodcock, Bull, Oliver, & Penhallow, 2014). The physical characteristics typically found in individuals with PWS are almond-shaped eyes, narrow forehead and face, small triangular mouth, thin upper lip, underdeveloped genitalia, small stature, small hands and feet, and tapered fingers (Cassidy, Schwartz, Miller, & Driscoll, 2012; Goldstone et al., 2008; Kundert, 2008; Tunnicliffe et al., 2014). Hypopigmentation may also be found in individuals with PWS and is caused by deletion of 15q11.2-q13 on the paternally derived chromosome (Goldstone et al., 2008). PWS has an estimated incidence of 1 in 10,000 to 1 in 25,000 live births (Khavari et al., 2017).

While the phenotypical presentation of Prader-Willi syndrome changes over the course of development (Boer & Clarke, 1999; Dosier, Vaughn, & Fan, 2017), it is characterised by low muscle tone (a condition known as hypotonia), deficiency in the production of sex hormones by the sex glands, affecting growth and sexual development (known as hypogonadism or cryptorchidism), general developmental delay (Beauloye et al., 2015; Holland et al., 1993; 1995; Khayat et al., 2017; Kundert, 2008; Pavone et al., 2015), mild to moderate cognitive impairment, autistic characteristics, and aggression (Dosier, Vaughn, & Fan, 2017), hyperphagia (overeating) and consequently result in a number of health comorbidities including obesity, hypothyroidism, Type 2 diabetes, and respiratory issues (Laurier et al., 2015). Additional features that emerge include the inefficient secretion of growth hormones, adrenotonsillar hypertrophy, and sleep disordered breathing (Beauloye et al., 2015). These clinical conditions have been attributed to hypothalamic-pituitary dysfunction (Beauloye et al., 2015). Symptoms that are more psychological in nature tend to emerge in adolescence, particularly symptoms of depression and anxiety, particularly low energy, lethargy, withdrawal, confusion, temper tantrums and obsessive-compulsive behaviours (Goldstone et al., 2008; Kundert, 2008; Pavone et al., 2015).

Challenging Behaviours in People with Rare Genetic Neurodevelopment Disorders

Children with rare genetic neurodevelopmental disorders (RGND) are commonly reported to exhibit a number of challenging behaviours that negatively impact their own functioning and wellbeing, as well as that of their family members (Hodap & Dykens, 2007; Holm et al., 1993; Neo & Tonnsen, 2019; Reilly et al., 2015; Waite et al., 2017).

There are a number of concerning behaviours that are syndrome-specific, wherein they may be a distinctive feature of that particular genetic mutation. An example of such a syndrome-specific behavior is insatiable appetite and overeating

("hyperphagia"), as is seen in Prader-Willi syndrome (Boer & Clarke, 1999; Cassidy et al., 2012; Kundert, 2008; Tunncliffe et al., 2014). In this case, the challenging behavior in question may be attributed to dysfunction of the hypothalamus, a brain region that is recognized as being impaired in PWS (Beuloye et al., 2015). Other challenging behaviour typically featured in those with PWS include compulsive behaviours and temper tantrums (Dimitropoulos et al., 2006; Maas et al., 2010; Tunncliffe et al. 2014). Angelman syndrome is similarly associated with a selection of distinct challenging behaviours including hyperactivity, and aggression, often in the form of property destruction (Didden et al., 2009; Strachan et al, 2009; Zhdanova et al., 1999). For children with Williams Syndrome, anxious and depressive symptomology is commonly exhibited (Leyfer et al., 2006). Unfortunately, prevalence statistics have not been reported in the literature as of yet for these syndrome-specific challenging behaviours and thus future research will be needed to establish such incidence rates.

Additionally, some problematic behaviours are a shared clinical feature occurring across RGND (e.g., self-injury; Hall, Arron, Sloneem, & Oliver, 2008). Sleep is another area of challenging behaviour for many children with RGND.

Sleep in children with Rare Genetic Neurodevelopmental Disorders

The function of sleep. Although the exact function of sleep has not yet been established, it has been determined that it plays a critical role in human development, as evidenced by the fact that children spend more time sleeping than any other activity (Wiggs, 2007). In addition to this anecdotal inference, the association between sleep and developmental processes such as physical growth, brain growth, memory consolidation, energy conservation, behavioral and emotional regulation, learning and attention, and social interaction is well recognised (Brown, Kuo, Phillips, Berry, & Tan, 2013; Deliens et al., 2015; Richdale, 2013; Staples, 2013; Stores & Wiggs, 1998; Turner & Johnson, 2012). Researchers and health

professionals in the field of child development heed the numerous health ramifications of poor sleep (Fallone, Owens & Deane, 2002). Cardiovascular, metabolic, and autoimmune disorders are among some of the physical health conditions with which poor sleep has been found to be associated (Brown et al., 2013; Colten et al., 2006; McDougall, Kerr, & Espie, 2005). Sleep deprivation has also been linked to increases in stress responsivity, somatic pain, increased incidence of mood disorders, impairments related to memory, cognition, and performance, poor psychosocial health, academic difficulties, increase in risk taking behaviours including drunk driving, smoking, and delinquency, decreased quality of life, and behavioural problems (Ahrberg, Dresler, Niedermaier, Steiger, & Genzel, 2012; Bagley & El-Sheikh, 2013; Lushington et al., 2013; Medic, Wille, & Hemels, 2017; Owens, 2014; Reale, Guarnera, & Mazzone, 2013; Schmidt & Van der Linden, 2015). Thus the significant impact that sleep problems can have on children with RGND and the wider family ecology provides ample justification for the investigation into effective intervention approaches.

Categories of sleep disturbance. There are three main classifications for sleep disorders in children, adolescents, and adults, either typically developing or otherwise. These are: (1) dyssomnias, (2) parasomnias, and (3) sleep disorders which are diagnostic features of an illness or disorder (e.g., sleep apnoea/sleep-disordered breathing) (American Academy of Sleep Medicine, 2014; American Psychiatric Association, 2013; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008).

Dyssomnias. Dyssomnia is a term which comprises insomnia and hypersomnia, those being difficulty initiating or maintaining sleep, and excessive sleepiness, respectively (American Academy of Sleep Medicine, 2005; Esbensen & Schwichtenberg, 2017). Insomnias may be further divided into two categories: behavioural insomnias, where presentations may include irregular sleep-wake patterns, night-time wakings, early morning waking, settling difficulties, delayed sleep

onset latency, and bedtime resistance (Esbensen & Schwichtenberg, 2017; Nunes & Bruni, 2015; Spruyt & Curfs, 2015), and insomnias caused by physiological factors, such as pain, abnormal melatonin levels, heart problems, and acid reflux (Esbensen & Schwichtenberg, 2017). An alternative conceptualization of dyssomnias organizes the sleep difficulties into intrinsic (the cause being from within the body), extrinsic (cause relating to pathological environment or conditions), and circadian rhythm disorders (Thorpy, 2012).

Parasomnias. Parasomnias are atypical arousals that take place during sleep and include sleep walking, sleep talking, excessive nightmares, and sleep terrors (Esbensen & Schwichtenberg, 2017; Markov, Jaffe, & Doghramji, 2006; Thorpy, 2012). These sleep-events involve behaviours, movements, perceptions, emotions, and autonomic nervous system functioning (Thorpy, 2012). Parasomnias are commonly distinguished as arousal disorders, sleep-wake transition disorders, parasomnias of Rapid Eye Movement (REM) sleep, and nonspecific parasomnias (Davis, Parker, & Montgomery, 2004; Thorpy, 2012).

Sleep apnoea/sleep-disordered breathing. Sleep-disordered breathing is an umbrella term encompassing such conditions as Obstructive Sleep Apnoea (OSA), Central Sleep Apnoea (CSA), and hypopnea (Murata et al., 2017). Sleep apnoea refers to the temporary lapse in breathing or shallow breathing that can occur during sleep in these conditions. In this way, sleep disordered breathing is considered a dyssomnia, due to its disruption to sleep. Although the focus of this thesis is on remedy of behavioural sleep problems, it is important to consider the exacerbating effect of such conditions on sleep disturbance in typically developing children, as well as children with RGND (Camfferman et al., 2006; Hamlin et al., 2011; Pavonne et al., 2015; Verbraecken et al., 2002).

Prevalence of sleep problems in RGND. Sleep difficulties are a commonly reported clinical problem among the parents of children with RGND. Contrasted with typically-developing preschool-aged children, of whom approximately 25% are reported by their parents to experience sleep problems, children with neurodevelopmental disorders have a significantly higher prevalence of sleep disturbance of some form (Blackmer & Feinstein, 2016; Robinson-Shelton & Malow, 2016). Studies indicate that between 30% to 90% of children with RGND present with some type of sleep disturbance (Annaz et al., 2011; Dykens & Kasari, 1997; Kidd et al., 2014; Kronk et al., 2010; Richdale, Cotton, & Hibbit, 1999; Summers, Allison, Lynch, & Sandley, 1995; Wong, Leonard, Jacoby, Ellaway & Downs, 2015; Young, Nagarajan, de Klerk, Jacoby, Ellaway, & Leonard, 2007). This compares to just 10 to 35 percent of typically developing children (Armstrong, Quinn, & Dadds, 1994; Bryars, Yolton, Rausch, Lanphear, & Beebe, 2012; Courturier et al., 2005; Davis et al., 2004; Gaylor, Goodlin-Jones, & Anders, 2001; Krakowiak et al., 2008; Lam, Hiscock, & Wake, 2003; Owens & Moore, 2017; Richdale & Schreck, 2009a; Scher et al., 1995; Singh & Zimmerman, 2015; Wake et al., 2006). The wide range in prevalence estimates reported in the literature may be attributed in part to such factors as variability in study aims and design, the type of neurodevelopmental disorder examined, and the type of sleep problems studied (Didden et al., 2002; Elia et al., 2000; Honomichl et al., 2002; Walz, Beebe, & Briars, 2005). In addition, estimates of sleep disturbance experienced by children may be skewed if those difficulties are not disruptive to the rest of the family or disregarded as “normal”, and therefore go unreported. Lack of parental understanding with regard to the appropriateness of certain sleep-related behaviours at different developmental stages may cause over-reporting, and henceforth overestimated prevalence statistics (Kronk et al., 2010).

Course of sleep problems in RGND. Sleep disturbance can persist in this population into adolescence and adulthood, especially if left untreated (Grigg-

Damberger & Ralls, 2013; Ipsiroglu et al., 2011). Sleep problems in typically developing children on the other hand have been shown to decline with age (e.g., 72% to 37% from age 2-5 to 10-17 years old) (Allik, Larsson, & Smedje, 2008; Clarkson, Williams, & Silva, 1986; Fisher, Pauley, & McGuire, 1989; Gregory & O'Connor, 2002; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Richdale & Prior, 1995; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012). Such sustained sleep difficulties are likely to negatively impact learning, behaviour, and cognitive develop in both the short- and long-term (Colten & Altevogt, 2006; Kronk, Bishop Raspa, Bickel, Mandel, & Bailey, 2010; Wiggs & Stores, 1996). Furthermore, child sleep problems may have a concurrently negative impact on the well-being, quality of life, physical and mental health of the broader family system (Chu & Richdale, 2009; Cotton & Richdale, 2010; McDougall et al., 2005). In a qualitative investigation of the experience of parents of children with Rett syndrome (RTT), McDougall et al., (2005) found mood, energy levels, and performance to be compromised as a result of their child's sleep problems. Similarly, the researchers uncovered a theme across participants that was characterized by strained relations between parents and general social nonfulfillment (McDougall et al., 2005).

Causes of sleep problems in children with RGND. In order to determine which are the most appropriate interventions, it is advantageous to first understand the etiology of the sleep problem. Within normally-developing populations, sleep disturbance is often rooted in behavioural causes (Owens & Mindell, 2011), however the etiological basis of sleep problems in children with RGND is multifactorial, consisting of biological (including comorbid health conditions), environmental, psychological, behavioural, and social factors (Robinson-Shelton & Malow, 2016). Also, given the unique nature of each RGND, research has uncovered heterogeneous, syndrome-specific variables contributing to the development and maintenance of sleep problems present in children with RGND (Blackmer & Feinstein, 2016; Clayton-Smith, 1993; Dan & Boyd, 2003; Didden, Korzilius, Smits,

Curfs, & Dykens, 2004; Goldman, Bichell, et al., 2012; Miano et al., 2004; Miano et al., 2008).

Biological factors. The literature that explores the cause of sleep problems in children with RGND is dominated by investigation into biological abnormalities, for example, circadian rhythm disturbances, abnormalities in sleep architecture or sleep/wake schedules, issues with the production and synthesis of melatonin, comorbid health conditions such as seizure disorders and conditions that impair one's ability to breathe, such as Central Sleep Apnoea (CSA), Obstructive Sleep Apnea (OSA), and hypopnea (Lin et al., 2007; Murata et al., 2017; Pavone et al., 2015; Urquhart et al., 2013), and side effects of medication (Braam et al., 2008; Forrest et al., 2009; Hiroe, 2000; Kronk et al., 2010; Dosier et al., 2017; Potocki et al., 2000; Spryt, Braam, Smits, & Curfs, 2016; Takaesu et al., 2012).

Sleep and wake states in humans are controlled by their internal circadian rhythm, which is in turn regulated by the neurohormone melatonin (Arendt, 2005; Didden & Sigafos, 2001). The typical course of sleep/wake schedule development begins with the polyphasic sleep pattern (meaning sleep for multiple periods throughout the day) exemplified in newborns, then transitions to a biphasic pattern (i.e., sleeping for two periods over the course of a day) at approximately three months of age in typically-developing infants, and then eventually evolves into a monophasic pattern (i.e., a singular sleep period each day) by three to four years (Ferber, 1996; Richdale, 1999). While sleep problems arising from such sleep patterns may be 'disruptive' in a child's early years, they are developmentally appropriate. On the other hand, a sleep/wake schedule would be considered 'disordered' if the presenting sleep problems were rooted in some abnormality in the physiology of that sleep/wake schedule (Ferber, 1996; Richdale, 1999). For instance, abnormally low levels of melatonin and abnormal regulation of basic circadian rhythms have been identified as having a causal role in disordered sleep/wake

schedules amongst children with higher prevalence developmental disorders, such as ASD (Melke et al., 2008; Tordjman et al., 2013), as well as with children with some RGNDs (Dosier, Vaughn, & Fan, 2017; Miyamoto et al., 1999; Takaesu, Komada, & Inoue, 2012).

The sleep difficulties and their hypothesized physiological underpinnings of the syndromes of participants included in this study are described below.

Sleep and Fragile X Syndrome. There is limited research analyzing sleep problems within samples of children with FXS, and even fewer examine FXS when it is not accompanied with a comorbid diagnosis of ASD or other conditions, however the existing research has shown that difficulties settling, increased wake-time after sleep onset, and shorter overall sleep duration are experienced by children with FXS (Gould et al., 2000; Miano et al., 2008; Richdale, 2003). An estimated 31-77% of individuals with FXS present with some form of sleep disturbance (Kronk et al., 2010; Richdale, 2003). The types of sleep problems exhibited in cases with FXS are akin to those experienced by the typically developing population (Harvey & Kennedy, 2002). In spite of the apparent similarities in sleep disturbance type across the two cohorts, research has found that individuals with FXS display sleep microstructure abnormalities, demonstrated specifically by lower transient slow EEG oscillations during non-REM sleep (Miano et al., 2008). In addition, the sleep architecture of children and young adults with FXS appears to comprise a high percentage of stage 1 sleep, and a low percentage of REM sleep, as well as having a low number of REM episodes compared to that of typically developing children (Miano et al., 2008).

Sleep and Prader-Willi Syndrome. Insomnia (i.e., difficulty settling, sleep onset delay, sleep maintenance), early morning waking, and excessive daytime sleepiness are typical sleep problems associated with PWS (Cassidy, McKillop, & Morgan, 1990; Cotton & Richdale, 2006; Gibbs, Wiltshire, & Elder, 2013; Richdale, Cotton, &

Hibbit, 1999). Prevalence of the above sleep difficulties range from 35 to 100% amongst children with PWS (Richdale, Cotton, & Hibbit, 1999; Tietze et al., 2012). Actigraphy, which is an objective measure of motor movement and provide a picture of sleep/wake patterns (Sadeh, 2011), and PSG studies have demonstrated that children with PWS have longer wake after sleep onset time, while at the same time having shorter sleep latency (Gibbs et al., 2013; Joo et al., 2010; Verillo et al., 2009; Vgontzas et al., 1996). Compared to typically developing children, those with PWS have longer wakeful periods and display more daytime sleepiness, however the two populations are similar with regard to total sleep time and frequency of night waking (Gibbs et al., 2013). Manni et al (2001) observed abnormal REM sleep patterns in people with PWS, specifically an earlier REM sleep onset compared to typically developing individuals.

Sleep-disordered breathing is often present in children with Prader-Willi syndrome, given the dysmorphism to the face, small nasopharynx, and small oropharynx with or without adenotonsillar hypertrophy (Pavone et al., 2015). CSA has been found to be prevalent in individuals with PWS, an observation that some have suggested may be due to their lower resting functional residual capacity (FRC) resulting from hypotonia and decreased respiratory muscle strength found in infants with PWS (Urquhart et al., 2013). This increases their risk for sleep-related hypoventilation, especially when in REM sleep. An additional contributing factor to such impairment in central respiratory control experienced by children with PWS, may be related to hypothalamic dysfunction and blunted chemoreceptor sensitivity following hypoxemia and hypercapnia which is common in such children (Pavone et al., 2015; Urquhart et al., 2013). Individuals with PWS are also frequently diagnosed with Obstructive Sleep Apnoea (OSA), purportedly linked to the increased incidence of obesity within this population (Lin et al., 2007; O'Donoghue et al., 2005).

Despite the evident physiologically-based differences in the sleep of children with RGND compared to the sleep of typically-developing children, there may also be a behavioural component to their sleep difficulties. The demonstrable role that environmental and learning factors have been shown to play in the sleep disturbance of children with higher prevalence developmental disorders (i.e., ASD, ADHD; Devnani & Hegde, 2015; Meltzer & Mindell, 2008; Reynolds & Malow, 2011; Sharma & Andrade, 2012) raises the question of whether such variables may be similarly implicated in the sleep disturbance of children with RGND.

Behavioural factors.

Behavioural model of sleep disturbance. A behavioural model of sleep disturbance has its basis in operant behavior theory (Didden et al., 2002). Operant behavior theory holds that the probability of a behaviour recurring under similar conditions is dependent on the antecedents (A) to the behavior/response (B), coupled with the consequences (C) that follow said behavior (Skinner, 1969). Specifically, the likelihood of behavioural repetition hinges on the nature of the contingencies and their interrelationship with the antecedents and the behavior/response itself. Antecedents are defined as discriminative stimuli that result in the occurrence of the behavior/response (Blampied, 2013). Consequences or contingencies are categorized as either reinforcers or punishers, respectively increasing or decreasing the likelihood of the behavior/response transpiring again (Blampied, 2013). Further, a reinforcing or punishing consequence may be deemed positive meaning that the contingency involves something being added into the situation or environment, or negative meaning that the contingency takes the form of something being removed from the situation or environment (Skinner, 1969).

While the behavioural model may be applied to sleep, it is important to note that sleep is not technically classified as a behaviour (Blampied & France, 1993;

Blampied, 2013). Sleep is instead deemed a biological state that does not appear to reliably fulfill the criteria of an operant behaviour, in particular, that the likelihood of its recurrence may be manipulated through its consequences (Blampied & France, 1993). It is held instead that the act of “falling asleep” may be better suited to serve as the instrumental behaviour in this model, as it is clearly under the control of discriminative stimuli, and is consequently reinforced by sleep itself (Blampied & France, 1993).

Of course, the behaviour of “falling asleep” does not occur in isolation, that is to say that there is typically a sequence of behaviours that lead to the ultimate act of falling asleep, and then sleep itself. In the consideration of behaviours that take place consecutively, the notion of ‘operant behavior chains’ come into play. Within such a chain, antecedents and consequences have dual-functionality, each acting to reinforce the behaviour that came before while simultaneously performing the role of discriminative stimuli, signaling for the next behaviour to occur. The discriminative stimuli present before and after each behaviour in the chain will either cause momentum toward the desired state (i.e., sleep), or disrupt the momentum (Skinner, 1969).

Discriminative stimuli in the context of sleep fall into three categories: interoceptive stimuli (e.g., fatigue, which relies on the quality and duration of recent sleep combined with the length of time since last waking), proprioceptive stimuli (e.g., from posture and body orientation), and exteroceptive stimuli (i.e., elicited from the physical environment and social context in which the wake-sleep transition takes place). Time of day at which an attempt at sleep initiation is to occur is another critical discriminative stimulus, and may be considered both an interceptive and external stimulus (Blampied & France, 1993; Blampied, 2013). It is said that the sleep initiation process is under stimulus control once an assortment of distinctive stimuli become associated consistently with the behaviour of “falling asleep” and are

consistently reinforced by sleep (Blampied, 2013). It follows then that when in pursuit of the desired behavioural outcome of falling asleep, the discriminative stimuli of the entire behaviour chain that prelude it must be consistently present to ensure its attainment, otherwise unintended reinforcers such as choice, disruption, or distraction may impede progression toward falling asleep (Blampied, 2013; Blampied & France, 1993). Bed refusal and sleep onset delay are possible consequences of inconsistent and/or indistinct exteroceptive cues involved in the behaviour chain concerning pre-sleep bedtime routines and falling asleep (Blampied & France, 1993). The risk of obstructed momentum toward sleep is especially high when sleep-competing discriminative stimuli are conspicuous in the environment or when the reinforcement that they offer is attractive and immediate (Blampied, 2013).

In order to maximize the likelihood of sleep initiation, having in place sleep-conducive antecedent stimuli during a bedtime routine is necessary (Blampied & France, 1993). Quiet, darkness, cool room temperature, and comfortable bedding are discriminative stimuli in the sleep environment that encourage a period of “behavioural quietude,” which is a state characterised by “the reduction of overt motor activity, covert cognitive activity, and the lowering of perceptual stimulation,” that ultimately results in falling asleep (Blampied, 2013; Blampied & France, 1993; Jin et al., 2013; Blampied, 2013, p. 174). Behavioural quietude is necessary in order for sleep-controlling internal cues to become salient and henceforth noticed, and that continued activity or stimulation may stifle the recognition of such (Blampied & France, 1993).

Co-sleeping, being placed into bed after falling asleep elsewhere, being fed, rocked, sung to, patted, walked, or driven around are steps that parents commonly believe are required in order for their child to sleep and they will willingly enable/enact for such purposes (Blampied, 2013; Blampied & France, 1993; Jan et al., 2008). While these rituals may aid in sleep initiation, they depend on the

participation of others (i.e., a parent/caregiver). The longer these patterns of inappropriate, parent-supplied discriminative stimuli are in place, the stronger their association with falling asleep becomes, such that even a slight deviation from the established pattern will hinder sleep onset (Blampied, 2013) or sleep reinitiation following night wakings (Blampied & France, 1993). For example, in the case of co-sleeping, the presence of the parent may become associated with sleep initiation and thus becomes a discriminative stimulus that is required for the child to fall asleep or re-initiate sleep after a night-time waking (France & Henderson, 1996).

Adding to the issue is the fact that parental attention brings with it comfort, engagement, and warmth, which can be a powerful and alluring reinforcer for children, given the immediacy of its effect (Blampied & France, 1993). Attention from a parent is oftentimes more enticing than sleep itself, meaning that it will likely compete with the biological state to become the dominant reinforcer (Blampied & France, 1993). When a child learns that such problem behaviour as bedtime refusal and expressions of distress will result in their parent attending to them, the behaviour is strengthened and the likelihood of its recurrence increased (Blampied, 2013). Further, double reinforcement occurs when an expression of distress is successful in gaining parental attention, where the behaviour is negatively reinforced by the escape it provides from the unpleasantness of solitude, while simultaneously being positively reinforced by the attention (Blampied & France, 1993). Then, with the provision of comfort and attention in response to their child's distress, parents experience relief from their child's aversive/disruptive behaviour, and they themselves are negatively reinforced. This mutually reinforcing interaction, creates a coercive behaviour trap, wherein both the child's and parent's behaviour are strengthened, and the likelihood of their future engagement in the same attention-seeking and attention-giving behaviours is increased (Blampied, 2013). Following the ongoing recurrence of this interaction, both the child and the parent refine their roles in order to avoid distress, through the increasingly earlier instigation of

engagement or attention on the part of the child, and the increasingly rapid response to their child, on the part of the parent (Blampied & France, 1993). Each party gets something out of the transaction that is beneficial in the short-term, but ultimately aversive in the long-term. As with any persistent discriminative stimuli, a coercive behaviour trap that is played out consistently will eventuate in the child requiring it in order to fall asleep (Blampied, 2013). What may have initially been a developmentally and socially acceptable set of discriminative stimuli, later produces bedtime tantrums, sleep-onset delay, and bed refusal (Blampied & France, 1993), and importantly highlights an inability to self-soothe and initiate sleep independently.

Alternatively, and more appropriately, positive sleep practices may be established wherein the child is able to fall asleep without parental presence and stimulation or activity. Then discriminative stimuli that are associated with the sleep environment or routines (e.g., soft toys, bedding, thumb sucking in younger children – or postural adjustments, relaxing, and turning away from sources of stimulation in older individuals), as opposed to discriminative stimuli associated with parental attention or reliance on other parent-supplied cues (Blampied & France, 1993; Blampied, 2013), can lead the child into the state of sleep initiation. These more appropriate discriminative stimuli are then reinforced by the sleep that follows. Bed-associated cues or self-produced comfort cues for sleep also help to encourage sleep re-initiation in children if they wake in the night, as the same stimuli that signaled for them to fall asleep at bedtime are present still in the child's immediate surroundings (Blampied & France, 1993; Blampied, 2013). Such self-soothing methods and objects will then be reinforced by the sleep that follows (Blampied & France, 1993; Blampied, 2013; Henderson et al., 2010). Ideally, reinforcement will follow sleep compatible behaviours comprising the behaviour chain, serving to strengthen and maintain them, and not behaviour that is incompatible with positive, self-regulated and self-initiated sleep.

Ultimately, given the multitude of different factors described above, it appears that sleep disturbance in children with RGND is the byproduct of an interaction between biological/physiological, behavioural, and environmental factors. The degree to which each of these factors is responsible for sleep problems is variable across each individual child, and thus cannot be assumed. Henceforth, each factor must be given consideration in the formulation of sleep interventions.

Common Interventions for Sleep Disorders

A variety of treatments have been developed to improve sleep (Richdale & Wiggs, 2005). The most common, empirically supported treatment approaches are pharmacological or behavioural (Braam et al., 2008; Malow et al., 2012; Mindell et al., 2006; Turner & Johnson, 2013; Vriend et al., 2011).

Pharmacological interventions. Medications are used frequently in the treatment of sleep problems in children. This is in part, due to the fact that they provide an effective and often expedient solution. Melatonin is a pharmacological treatment that has been the subject of a considerable amount of childhood sleep management research (Blackmer & Feinstein, 2016). Melatonin is a hormone produced by the suprachiasmatic nucleus via sympathetic β -adrenergic receptors in the pineal gland that promotes normal sleep-wake cycles (Moore; 2007). Melatonin is released in response to night (dark) and day (light) (Moore; 2007). The natural production of melatonin in neurodevelopmental disorders is commonly thought to be impaired, resulting high rates of melatonin use in the management of sleep disturbance, particularly among children with ASD (Cortesi et al., 2012; Damiani et al., 2014; Wasdell et al., 2008). Whilst melatonin is effective, it has a number of limitations, including a reduction in efficacy over time, and reports of headaches, dizziness, rashes, cough, increased seizure activity and asthma attacks (Blackmer & Feinstein, 2016; Braam et al., 2013; Bruni et al., 2018). Other medications used in

the treatment of sleep include antihistamines, clonidine, guanfacine, and benzodiazapines (Felt & Chervin, 2014).

Behavioural interventions. Behaviourally-based interventions are founded on the principles of operant behaviour and learning theories, and seek to target the environmental, behavioural, and cognitive causes of sleep problems (Owens, France, & Wiggs, 1999). These strategies emphasize the role of stimuli present surrounding sleep-related behaviours, including those that signal for the occurrence of such behaviour (i.e., antecedents), along with those that serve as positively or negatively reinforcement for such (i.e., consequences). Behavioural interventions are conventionally separated into antecedent-based procedures and consequence-based procedures, based on which type of discriminative stimuli they address. Antecedent-based approaches include sleep hygiene (as described above) and bedtime routine modification, visual supports, social stories, sensory modulation and stimulus substitution, faded bedtimes with and without response cost, scheduled wakings and chronotherapy (Blampied & France, 1993; Jan et al., 2008). Consequence-based techniques include standard extinction and modified extinction procedures (including graduated extinction, minimal check, and parental presence), and multimodal treatments (Blampied & France, 1993).

Antecedent-based procedures. Traditionally, behavioural sleep interventions begin by addressing the antecedents identified by functional assessment as being the discriminative stimuli preceding the problem behaviour. This is done using antecedent-based procedures which are formed on the principles of operant behaviour theory (Blampied, 2013; Skinner, 1969). They utilize stimuli that work on the basis that any stimuli present in the spatial and temporal vicinity of the behaviour will serve to reinforce said behaviour. Antecedent-based procedures may also be useful when attempting to chain behaviours together, such as in a bedtime routine, where activities and cues can be manipulated to facilitate progression to the next

phase of the routine. Antecedent-based interventions include sleep hygiene, visual supports, bedtime fading, sleep restriction, chronotherapy, and scheduled wakings. Of this assortment of therapies, establishment of good sleep hygiene and a bedtime routine, bedtime fading, sleep restriction, and bedtime scheduling has been used with children with RGND. The convention in behavioural sleep interventions is to begin with the establishment of positive sleep hygiene.

Sleep hygiene modifications. Good sleep hygiene has been found to be a critical component of overall sleep quality over the course of one's life (Brown et al., 2014; Mindell et al., 2009; Spruyt & Curfs, 2015). Modification to sleep-related behaviours that encourage good sleep, or "sleep hygiene," is the standard first step in treating sleep disturbance in children with neurodevelopmental disorders (Grigg-Damberger & Ralls, 2013; Jan et al., 2008; Stores, 2001). In establishing good sleep hygiene, activities and cues associated with the sleep environment (e.g., noise, light, room temperature, bed position), sleep schedule (e.g., regularity of sleep and wake times), physiologic factors (e.g., preventing hunger, avoiding overstimulation), and sleep practices/bedtime routines (e.g., relaxing activities before bedtime) are targeted to optimize sleep (Christodulu & Durand, 2004; Gradisar & Short, 2013; Jan et al., 2008; Owens et al., Schreck, 2001; Vriend et al., 2011). Establishing sleep-conducive bedtime routines is an especially important part of sleep hygiene, and may include any activities or rituals that are relaxing and enjoyed by the child (Christodulu & Durand, 2004; Kodak & Piazza, 2008; Midell et al., 2006; Schreck, 2001). Examples of such activities include bathing, getting into pyjamas, brushing teeth, reading, listening to calming music, and turning out the light before bedtime (Christodulu & Durand, 2004; Schreck, 2001). In addition to the establishment of routines that promote sleep, a sleep hygiene protocol should also seek to remove behaviours and cues that are not conducive to sleep and that reinforce wakefulness, such as technology/device use leading up to bedtime (Cortesi et al., 2010; Deliens et al., 2015; Gradisar & Short, 2013). In many cases, good sleep hygiene is achieved

through a process of trial-and-error for children with neurodevelopmental disorders, and the presenting disorder will determine the selection of which specific techniques to employ (Blackmer & Feinstein, 2016).

Bedtime scheduling and bedtime fading. Systematic and progressive alteration of bedtime can be an effective approach to the resolution of sleep onset delay/latency, night wakings, and early morning wakings (Piazza et al., 1997). Faded bedtime procedures close the temporal gap between one's bedtime and the time of their actual sleep onset, after which the bedtime is shifted forward in a sequential manner until a developmentally appropriate bedtime is attained (Mindell et al., 2006). A time at which a child will naturally fall asleep after 15 minutes of being put to bed is identified through assessment and baseline measures. With the commencement of treatment, a new bedtime introduced which aligns with this natural sleep time. The child's bedtime is then methodically brought forward to an earlier time over the course of treatment, whenever they have progressed to the point of reliably falling asleep within a few minutes of their designated bedtime (Kodak & Piazza, 2008; Richdale & Wiggs, 2005; Turner & Johnson, 2012; Vriend et al., 2011). As an alternative method, bedtime scheduling involves the enforcement of a nominated sleep and wake time (Piazza et al., 1997). A child partaking in bedtime scheduling is not permitted to sleep outside of their predetermined bedtime and morning wake time. If the child fails to fall asleep in this sleep period, they are prevented from sleeping until their next designated sleep period (Piazza et al., 1997). In both faded bedtime and scheduled bedtime procedures, children must be woken at a scheduled time in the morning and they must be prevented from sleeping during the day, as unplanned additional sleep will likely interfere with the goals of this intervention (Vriend et al., 2011).

While sleep hygiene implemented in isolation is not sufficient to resolve sleep problems completely, the effectiveness of other sleep intervention procedures is

contingent on the establishment of good sleep hygiene (Jan et al., 2008; Johnson, Giannotti, & Cortesi, 2009; Mindell, Telofski, Wiegand, & Kurtz, 2009; Singh & Zimmerman, 2015; Vriend et al., 2011). Modification to a child's sleep hygiene is thus generally carried out together with additional behavioural strategies tackling the antecedents preceding the problematic sleep behaviour, such as bedtime scheduling and bedtime fading.

Consequence-based procedures. Directing focus toward the consequence element of the A-B-C framework of the behavioural model of operant behaviour theory (Blampied, 2013; Skinner, 1969), consequence-based interventions concentrate on the factors immediately following an undesired behaviour. Such procedures function through the manipulation of these factors in order to decrease the incidence of the problematic behaviour in future (Wiggs & France, 2000). Some common consequence-based procedures used in the treatment of sleep disturbance include extinction or modified extinction procedures (e.g., standard extinction, graduated extinction, minimal check, and parental presence), and reinforcement schedules.

Extinction or modified extinction procedures. The standard extinction procedure addresses sleep problems related to settling and night waking, where functional behaviour assessment has concluded that the behaviour serves to instigate or retain parental attention (Didden et al., 1998). Extinction necessitates the consistent withholding of social reinforcement that has previously served as a factor maintaining undesired behaviour (Didden et al., 2002; Owens et al., 1999; Turner & Johnson, 2012; Vriend et al., 2011). A traditional extinction procedure would consist of the parent only attending to/engaging with the child when absolutely necessary (e.g., if the child's safety or health is in question), and otherwise ignoring the child's behaviour until an appropriate morning wake-time (Owens et al., 1999; Turner & Johnson, 2012, Vriend et al., 2011). In essence, parents would be required to avoid

responding to any sleep-resistant behaviour such as tantrums, crying, calling out, or leaving the bedroom (Allen et al., 2013). The anticipated result of the intervention is that problematic behaviour including bedtime resistance, sleep onset delay, and night wakings will no longer occur (Vriend et al., 2011).

Behavioural approaches are currently considered the gold standard in treatment of childhood sleep disturbance (Mindell et al., 2006b; Morgenthaler et al., 2006). Aside from their proven efficacy, behavioural intervention procedures have been shown to have greater social acceptability, due to longer-lasting effects, fewer negative side effects, the potential for treatment effects to generalize to daytime problems, and increased parental self-efficacy and coping abilities (Grigg-Damberger & Ralls, 2013; Stores, 2001).

Sleep Treatment in RGND

Despite the growing empirical support for the use of behavioural interventions in the treatment of sleep disturbance in typically developing children (Mindell et al., 2006b; Morgenthaler et al., 2006) and children with ASD (Richdale, 2013), medical and pharmacological approaches are typically opted for in cases involving children with RGND. This is evidenced by the comparatively abundant literature which investigates medical and pharmacological treatments (Bailey et al., 2012; Braam et al., 2008; Wong et al., 2015). For example, in a survey by Wong et al., (2015) it was evident that 43.4% of parents of children with Rett syndrome chose to treat their child's sleep problems through non-specific sleep medications and 28.3% through sleep medications. Conversely, only 12% of respondents choose to implement non-pharmacological treatments (Wong et al., 2015). This is in spite of the evidence that parents have trepidation over the side-effects that come with various medications, and their effectiveness in the long-term (Bramble, 1996). Underlying the preference for medications may be the fact that health professionals or parents often attribute sleep problems to being characteristic of the disorders themselves (i.e., as an

eventuality of cortical or chemical abnormalities) and therefore do not consider behavioural interventions appropriate (Allen et al., 2013; Walz, Beebe & Byars, 2005; Pelc et al., 2008). Furthermore, the lack of empirical evidence demonstrating the efficacy of behavioural approaches to sleep problems in children with RGND is likely to deter their implementation. It is important to note that the research that does exist which explores the effects of behaviourally-based treatments for sleep problems is promising. This research is reviewed extensively in chapter two of this thesis. Whilst these studies indicate the promising effects of behavioural treatments, further research in this area is required.

Functional Behaviour Assessment

Implementing an intervention without the function of the challenging behaviour being targeted is an ill-informed approach, and will likely lead to less effective treatment outcomes (Hanley, 2016). In many cases, treatments are prescribed based on surface evaluation of a problem behaviour, without careful consideration of purpose or function that the behaviour is serving (Brown & Piazza, 1999; Hanley et al., 2003). The result of treatments that do not consider the antecedents and consequences of the behaviour and how these factors may be functioning to facilitate to preservation of said behaviour, is merely modification without necessarily resolving the issue (Campbell, 2003; Hanley, 2016; Horner et al., 2002). Not to mention that ineffective treatments waste time, money, and can be distressing and disheartening to the child and family involved. Promisingly, the empirical evidence supporting interventions based on functional behaviour assessment (FBA; Brown et al., 2013; Didden & Sigafoos, 2001; Hanley, 2016; Hanley et al., 2014; Kodak & Piazza, 2008) is increasing in the field of clinical behavioural intervention.

Definition and process of functional behaviour assessment. An application of operant behaviour theory that has proven to be an effective tool in the elimination of problem behaviours is Functional Behaviour Assessment (FBA; Beavers, Iwata, &

Lerman, 2013; Blampied, 2013; Brown et al., 2013; Hanley et al., 2014; Hanley, 2016; Horner et al., 2002; Kodak & Piazza, 2008). FBA is a procedure through which the most pertinent discriminative stimuli and contingencies causing and maintaining the behaviour of interest are identified and used to inform treatment planning. The logic behind the approach is that one can alter a behaviour by manipulating the antecedents and consequences that either precede or follow it (Horner et al., 2002). The FBA process involves gathering information from a variety of sources regarding the problem behaviour and then using that information to generate a hypothesis concerning the function of the behaviour (Blampied, 2013). Interviews, checklists, ratings, self-report measures, questionnaires are among some of the tools used to compile objective information about the antecedents and consequences working to facilitate and encourage undesirable behavior (Blampied, 2013). Additionally, direct observations are utilized in FBA to provide insight into how the problem behavior presents in natural contexts (Blampied, 2013). The comprehensiveness of this assessment procedure enables inferences to be made regarding the factors that may be causing and maintaining the behavior in question. From there, evidence-based intervention procedures that address the various antecedents and consequences enveloping the behavior can be selected (Blampied, 2013; Horner et al., 2002).

The FBA process accounts for the fact that the aetiology of problematic behaviour is multifactorial, and henceforth it takes heed in ensuring not to confine a challenging behaviour to traditional categorisations, diagnostic labels, or norms (Horner et al., 2002; Blampied, 2013; Brown & Piazza, 1999; Kodak & Piazza, 2008). Additionally, in undertaking FBA the focus is set on present and recent occurrences related to the problem behaviour, rather than those of the past (Blampied, 2013).

Functional Behaviour Assessment for sleep problems. There are a number of examples of the use of FBA to inform the treatment of challenging behaviour in

typically developing children and children with developmental disabilities, including RGND (Anderson, Rodriguez, & Campbell, 2015; Arndorfer & Miltenberger, 1993; Radstaake et al., 2013). There are also a small number of examples in which FBA has been used successfully to inform the treatment of sleep problems in children (Blampied, 2013; Jin et al., 2013; McLay et al., 2017; Moore et al., 2004; Papadopoulos et al., 2015). The information obtained about sleep difficulties in FBA-informed sleep treatment comes predominantly from sleep diaries, but also through interviews, video analysis, and parent-report questionnaires (Blampied, 2013). Interviews are an efficient way to build a picture of the sleep disturbance (i.e., the frequency, duration and intensity), the setting/s that the sleep-related behaviour takes place within, and the preceding and consequent stimuli that frequently occur around the sleep problems (Blampied, 2013). An interview can also be used to procure other contextual information (i.e., developmental history, past intervention attempts) that might further ones understanding of the sleep disturbance and subsequently aid in the development of a treatment plan (Blampied, 2013). Parental preferences, concerns, and thoughts about their child's sleep are some other valuable details that can be uncovered through interviewing and can provide an opportunity for rapport-building between families and health professionals overseeing treatment (Blampied, 2013). Supplementary to interviews, parent-report questionnaires are also commonly administered to gain objective identification of the specific sleep problems at hand, and measurement of its frequency, duration, and intensity where appropriate (Blampied, 2013). Assessment packages in FBA of sleep disturbance often contain the Sleep Assessment and Treatment Tool (SATT; Hanley, 2005), Child Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000), and the Questions About Behavioural Function (QABF; Matson & Vollmer, 1995) questionnaires.

Direct measures comprise parent-reported sleep diaries and observations in the form of video recordings of children during the night (Blampied, 2013; Hanley et

al., 2014). Sleep diaries provide parental report of instances of sleep disturbance and may request description of the type (e.g., sleep-resistant behaviour, curtain calls, night-time wakings), frequency, duration, and context of sleep problems, in addition to any responsive action on the parents' part. Video recordings enable the first-hand viewing of sleep problems as they occur typically for the child (Blampied, 2013). Information of this kind can be used to validate parental report, and uncover any behavior that parents are not aware of (Jan et al., 2008; Richdale & Schreck, 2009). The aforementioned direct and indirect measures are used in conjunction to build a comprehensive picture of the problematic sleep behaviour, prevent reporting bias, and establish treatment effects via ongoing or repeated measurement (Knight & Johnson, 2014; Spruyt & Curfs, 2015).

Once all necessary information is accumulated through the various assessment techniques, a treatment plan that takes into account all that has been uncovered can be crafted to address the individual's personal sleep problems (Blampied, 2013). Antecedents and consequences that are typically identified as being maintaining factors in cases of childhood sleep disorders include poor sleep hygiene, parental presence at sleep onset, possibility of procuring desirable tangible items, and self-stimulatory behaviours. From there, a treatment plan would be developed based on the findings of the FBA. For instance, if it was suspected that the function of a presentation of co-sleeping was to gain attention from a parent, then the treatment approach would be to remove parental attention at times where the child would be falling asleep, or going back to sleep following a night waking.

Given that functional assessment places emphasis on recent behaviour and events (Blampied, 2013), it is often necessary to amend portions of the treatment plan over the course of intervention, to ensure that all treatment techniques continue to serve their purpose in addressing the function of the behaviour. In this sense, repeated measurement of sleep outcomes throughout baseline and treatment

phases using direct methods (i.e., sleep diaries and video recordings) is critical. In addition to making sure that intervention is responsive to the changing needs of the individual as they progress or regress in treatment, it is important that intervention approaches be kept to a minimum, only including techniques that are actively producing positive outcomes. Intervention procedures that become redundant at any point during treatment should be removed or discontinued.

Following an FBA-based sleep intervention, it is advisable to carry out a measure of social validity to gain insight into the family's thoughts about the intervention, their level of comprehension with respect to approaches used, any secondary effects that they observed relating to their child or themselves, their level of satisfaction with the treatment outcomes, and any recommendations they might offer (Finn & Sladeczek, 2001; Hanley et al., 2014). The greater the social validity of an intervention, the more likely it is that parents will adhere to treatment plans, and henceforth promote superior treatment outcomes (Brown et al., 2013). The Treatment Acceptability Rating Form (TARF-R; Reimers & Wacker, 1992) is a questionnaire designed for this purpose, and may be preferable to a semi-structured interview format, where parents may be less likely to offer honest opinions whilst face-to-face with a figure involved with the intervention. Establishing social validity is in keeping/compatible with the principles of FBA, as it maintains the emphasis around gathering information from various sources to inform treatment. While it is a measure obtained post-treatment and obviously cannot be used to develop the now completed intervention, such data can be used to inform future interventions with other families.

Family Collaboration

As is stipulated by the systems perspectives of human development, a child grows within the confines of multiple interrelated biological, psychological, social,

and cultural contexts (Bronfenbrenner & Morris, 2006; Ford & Lerner, 1992; Lerner, 2006). In an application of this theory, it may be posed that a child's familial system is partially responsible for modeling, shaping, and maintenance of problematic behaviour (Wahl, Johnson, Johansson, & Martin, 1974). With this being the case, it is important for the family context (particularly parent-child interactions) to be examined in the assessment of problem behaviour for intervention. Then in the same way that the family system aids in the development of maladaptive behaviour, it likewise plays a crucial role in the reversal of such behavioural challenges. Given that sleep typically occurs in the home, amongst family members residing in the shared space, parents are instrumental in the implementation of sleep interventions. In a review of the literature, Carr and colleagues (1999) found that treatment success was 37% more likely with behavioural interventions implemented by familiar support figures such as parents or teachers and carried out in natural settings, compared with interventions conducted by "atypical" support agents.

It is important that treatment plans are deemed socially acceptable by the parents who will be charged with the implementation of the plan, if the desired treatment outcomes are to be achieved (Moore, 2004; Turner & Johnson, 2012). Functional behaviour assessment requires the perspectives of the family to be gathered and henceforth enables/promotes the procurement of parental knowledge, preferences, goals, in addition to any cultural or ethical standpoints that should be incorporated in the resulting treatment design (Jin et al., 2013). Seeking familial input when designing interventions can assist in ensuring parents' adherence to interventions, as they are likely to feel that they have greater agency over the endeavor given that they were actively involved from the beginning (Blampied, 2013; France, 1994; France et al., 1996; Moore, 2004; Turner & Johnson, 2012).

Chapter 2

Literature Review

The purpose of this review is to appraise the scope of the existing/extant intervention research targeting/covering/on sleep disturbance experienced by children with RGND. Literature discussing both behavioural and pharmacological evidence-based treatment methods within this population is examined at length, including melatonin, human growth hormone (GH) replacement therapy, Diphenhydramine HCl, bedtime fading, bedtime scheduling, reward systems, extinction, desensitization procedures, psychoeducation, “Excuse-me” drill, sleep hygiene, sleep environment, prevention of daytime napping, and visual aids. Many multi-component interventions. The review closes with a summary of the findings and the state of the literature discussed, followed by the rationale for the current study.

Search Process

Given the relative scarcity of empirical research published on/regarding evidence-based behavioural procedures, this review also covers non-behavioural interventions in order to develop a more comprehensive understanding of the customary treatment approaches exercised with children with RGND for their sleep difficulties. The selection of RGNDs included in this search was decided based on a comprehensive internet search, as well as correspondence with individuals via the PedSleep forum. Their resulting list of RGNDs to include were FXS, AS, RTT, SMS, Williams syndrome, Cri-du-chat syndrome, Turner syndrome, DiGeorge syndrome (velocardiofacial syndrome or 22q11.2 deletion syndrome), Sotos syndrome, septo-optic dysplasia, PWS, and agenesis of the corpus callosum. Additional syndromes were also added if later identified during literature searching, though this was not the case. As with the McLay and colleagues’ (2019) search procedure, developmental disabilities that were deemed degenerative, proliferative, or resulting from infection or environmental causes were excluded from the search list, as were disorders that

did not feature developmental or intellectual disability (e.g., epilepsy). Inclusion required that an article (a) be published in and English-language, peer-reviewed, academic journal; (b) involve at least one participant with a primary diagnosis of RGND that was under the age of 18; (c) employ a behavioral treatment (Cooper, Heron, & Heward, 2007); and (d) report quantitative data on sleep-related treatment outcomes.

PsychINFO, PsychARTICLES, Psychology and Behavioural Sciences Collection, and Education Resources Information Centre (ERIC) were the electronic databases used to conduct the literature search. Search terms input into the above databases were 'sleep', keywords relating to intervention ("treatment", "intervention", "therapy"), and the names of each listed RGND individually. Finally, an ancestry search was conducted for the articles that had been detected during initial database searches.

Pharmacological Interventions for Sleep Disturbance in Children with RGND

Melatonin. There were five articles sourced that treated sleep disturbance in children with RGND with melatonin. The studies describe participants ranging from two to 20 years of age, and included diagnoses of AS, Smith-Magenis syndrome, and RTT. Sleep onset latency, night wakings, and reduced total sleep time are among the sleep difficulties treated by melatonin in the studies found.

Braam and colleagues (2008) randomly assigned 8 children with AS between ages four and 20 to a treatment or placebo-controlled group. The former was administered 2.5 mg/5 mg (age-dependent) of melatonin mixed with carboxymethylcellulose in a fast-release tablet, and the latter were given an identical-looking placebo. Participants received melatonin treatment for a period of four weeks, during which time average sleep onset latency decreased by 32 minutes, and average total sleep time increased by 56 minutes. Night wakings

decreased from a mean of 3.1 to 1.6 over the course of a week. These results contrast with those of the placebo control group, who experienced an average decrease in SOL of .75 minutes, an average decrease in total sleep time (TST) of 9 minutes, and a reduction in average number of night wakings from 1.7 to 1.8 (Braam et al., 2008). Additionally, the authors conducted an open treatment trial with 20 new participants, and observed a lessening in therapeutic gain in some participants by the fourth week of treatment. In these cases, salivary endogenous melatonin levels had become exceptionally high, to the point where normal melatonin rhythms had been expunged (Braam et al., 2008).

In another study targeting 13 children with AS (aged two to 10), Zhdanova and others (1999) implemented a daily treatment of a 0.3 mg dose of melatonin. Blood samples were taken every hour for two 21-hour periods, one pre-treatment and one during treatment, to establish the participants' individual endogenous serum melatonin levels and the levels brought about by melatonin treatment, respectively. Actigraphy was also used to measure motor activity 24 hours per day for seven days prior to treatment and then five days during treatment, corroborated by parent recorded sleep diaries. The results indicated that melatonin treatment had significantly improved the participants' sleep, with a notable reduction in motor activity during specified sleep periods and an increase in total sleep time overall. Parents of the participating children noted a decrease in SOL and a lowered susceptibility to being woken by noises in and around their sleep environment following the commencement of melatonin treatment (Zhdanova et al., 1999).

In a randomized, double-blind, placebo-controlled trial, Gringas and others (2017) assessed the utility of PedPRM (a prolonged-release melatonin) in treating sleep problems in a sample of 125 children between the age of two to 17.5. Within their participant group/pool 96.8% had ASD and 3.2% (four children) had Smith-Magenis syndrome. The children received either a 2/5mg dose of PedPRM over the

course of 13 weeks, or a placebo. A “Sleep and Nap Diary,” composite sleep disturbance index (CSDI), and actigraphy were included as measures of treatment effects. By the end of the 13-week treatment period, TST increased on average by 57.5 minutes in the treatment group. Contrastively, the placebo group demonstrated an average increase in TST of 9.14 minutes. The treatment group also showed improvement in SOL, with a mean decrease of 39.6 minutes, compared to a mean decrease of 12.5 minutes in the placebo group. CSDI scores indicated a decrease in overall sleep disturbance amongst treatment group participants (Gringas et al., 2017).

Nine girls with RTT with a mean age of 10.1 years were prescribed 2.5 to 7.5 mg of immediate release melatonin or placebo in a double-blind, placebo-controlled, crossover study with randomized treatment order (McArthur et al., 1998). Sleep onset latency and total sleep time data were gathered through actigraphy and sleep diaries during one week of baseline assessment, the 1st four-week treatment regimen, a one-week “wash-out” period, and a second four-week treatment period using the alternate medication. With regard to SOL, the treatment group showed a considerable decrease within the first three weeks of treatment, with improvement noticeable during the first nights of melatonin supplementation in a number of instances (McArthur et al., 1998). Gains in TST were detected for the three participants who had the poorest sleep efficiency at baseline. The group mean TST was not statistically significant however, nor were the mean group differences. Frequency of night wakings were similarly unaffected by melatonin treatment in this sample. Overall, there was considerable variability in the individual responsiveness to the melatonin treatment and the subsequent improvement in sleep outcome variables across participants (McArthur et al., 1998).

Miyamotoe and colleagues (1999) describe their treatment of a seven-year-old (patient 1) and a 13-year-old girl (patient 2) with RTT and sleep disturbance, using

melatonin. Patient 1 demonstrated a free-running sleep-wake cycle (i.e., sleep pattern that does not adhere to the typical 24-hour cycle in humans) prior to treatment, while patient 2 displayed a fragmented sleep pattern and parasomnia characterized by night screaming. Both girls were administered 5 mg of melatonin and blood samples were drawn at hourly intervals which were then examined through radioimmunoassay every few months for blood cell counts, urinalysis, serum chemistry, luteinizing hormone, follicle-stimulating hormone, prolactin and estradiol. In the case of patient 1, a normal sleep-wake cycle was established and progress was maintained after two years. Free-running sleep onset reemerged on occasion when melatonin supplementation was suspended, however these cleared up when melatonin treatment was reinstated. Patient 2 was consistently falling asleep within 30 minutes of taking melatonin each night, thus demonstrating some improvement in sleep pattern. However, early morning wakings did not improve with treatment and fragmented sleep returned when treatment stopped (Miyamoto et al., 1999).

Other medications. Two articles were found that addressed non-melatonin pharmacological interventions, namely human growth hormone (GH) replacement therapy and diphenhydramine HCl. The two articles presented single case descriptions of a 5-year-old girl with Smith-Magenis syndrome, and a 9-year-old boy with AS. The sleep disturbance exhibited in these two cases included early morning wakings, reduced total sleep time, and daytime sleep.

A single-case study conducted by Itoh and others (2004) reports on the elimination of sleep disturbance of a 5-year-old girl with Smith-Magenis syndrome who received human growth hormone (GH) replacement therapy for dwarfism. The particular variables of interest were sleep-wakefulness circadian rhythm, which were measured through direct observation on a daily basis and polysomnography for one night four months before and one night four months after treatment. Morning wake times shifted from an average time of 4-4.30am before treatment was introduced to

approximately 6am. TST also improved, increasing from a mean of 399 minutes pre-treatment to 502 minutes. Low percentage of REM sleep before treatment was increased following human growth hormone (GH) replacement therapy, specifically from 2 to 5 REM episodes (Itoh et al., 2004).

Using a treatment package consisting of both pharmacological and behavioural components, Summers and colleagues (1992) treated a 9-year-old boy with AS who was experiencing sleep problems. Diphenhydramine HCl was prescribed, and the behavioural procedures put in place included restriction of daytime sleep, establishing a consistent sleep schedule, and reducing parent-child interactions during sleep onset and night wakings. The intervention was conducted at an inpatient behavioural treatment unit primarily and implemented by staff, however the treatment setting was transferred to the participant's home after 55 days of treatment, implemented from then on by his parents. TST increased from a mean of 1.9 hours per night and 1.3 hours during the day at baseline to a mean of 8.3 hours per night and .8 hours during the day. Following these results, medication was stopped, and subsequently TST decreased slightly to a mean of 7.8 hours during the night and .7 hours during the day. Treatment effects were maintained at a 45-day follow-up, with a mean TST of 7.1 hours at night and .29 hours during the day (Summers et al., 1992).

The small body of existing research examining pharmacological treatments for sleep problems in children with RGND points to the short-term effectiveness of medications such as melatonin, human growth hormone (GH) replacement therapy, and Diphenhydramine HCl. Across the studies, improvement was observed in the participant's sleep behaviours following treatment. This improvement was greater for treatment groups compared with placebo control groups. The only exception to these results was found by McArthur and others (1998), for whom results were mixed for TST and night wakings.

Two studies utilized a randomized, double-blind, placebo-controlled trial (Gringas et al., 2017; McArthur et al., 1998), which is deemed the 'gold standard' research design for drug trials due to its ability to reduce bias, balance participant characteristics between groups, and provide insight into the cause-effect relationship between treatment and outcome (Hariton & Locascio, 2018). One study implemented a randomized, placebo-controlled trial (Braam et al., 2008), and four studies followed a single-case research design (McArthur et al., 1998; Miyamoto et al., 1999; Summers et al., 1992; Zhdanova et al., 1999). Although these studies describing single cases are not protected from bias, they do allow for the examination of causal relationships due to their comprehensiveness. The overall high quality of the research designs used subsequently lends credibility to the findings of these studies. Importantly however, the long-term maintenance of initially positive treatment was largely unexplored. The studies that did conduct long-term follow-up of effects found mixed results, with Summers and others (1992) observing sustained improvement, and Miyamoto and colleagues (1999) seeing a return to pre-treatment levels of sleep disturbance.

Behavioural Interventions for Sleep Disturbance in Children with RGND

Six articles were uncovered that described the treatment of children with RGND using behavioural interventions. The participants in these studies ranged from 2 to 18 years old and had RGND diagnoses of PWS, FXS, AS, and Williams syndrome. The behavioural procedures implemented across the included articles were faded bedtime with a response cost, bedtime scheduling, graduated and non-graduated extinction procedures, parental psychoeducation, modification to sleep environment, manipulation of sleep/wake schedule, "Excuse Me Drill", visual aids, reinforcement procedures, and prevention of daytime napping. The types of sleep problems treated with the aforementioned procedures included SOL, night wakings, early morning wakings, co-sleeping, sleep-related anxiety, bedtime resistance, difficulty

independently initiating sleep without parental presence, and excessive daytime napping.

In a study of 14 participants (including one child with PWS) aged from 4 to 14, a behavioural treatment involving faded bedtime with a response cost (the response cost being to remove the child from bed and keep them awake for one hour when the child did not fall asleep within 15 minutes of their bedtime), or bedtime scheduling was employed to treat sleep difficulties (Piazza, Fisher, & Sherer, 1997). Participants underwent a baseline period, followed by one of the two behavioural treatment procedures, to which each child was randomly assigned. Measures that were taken at pre- and post-treatment to evaluate treatment efficacy comprised direct observation through momentary time sampling over 24 hours at every half hour interval, and a global measure of sleep disturbance, which was calculated by adding the duration of sleep that occurred outside of appropriate sleep times plus the duration of wake time that took place within the defined appropriate sleep time. Within the bedtime scheduling group, the mean hours of disturbed sleep decreased from 1.37 during baseline to 1.10 hours post-treatment, however this effect was not significant. For the group that were prescribed a faded bedtime procedure (with response cost), the mean hours of disturbed sleep decreased to a significantly greater degree, beginning with 1.44 hours at baseline and ending treatment with 0.53 hours of disturbed sleep. The child with PW had presented with some very early morning wakings and some night wakings, both of which improved greatly following his faded bedtime with response cost. Of particular note was the finding that it was daytime sleep was only able to be eliminated once the faded bedtime was coupled with the response cost, signifying the importance of this intervention component.

Didden and others (1998) implemented behaviourally-based interventions with six boys ranging from two to seven years of age. Within the participant group, there was one boy with PWS presenting with settling difficulties and night wakings, and

one boy with FXS presenting with sleep-related anxiety and co-sleeping. During a baseline phase, interviews and sleep diaries were completed which informed a functional analysis conducted for each of the children to ascertain the function of their problematic sleep behaviour and point to potentially effective solutions.

Throughout all phases of the study, parents took note of the number of minutes that target behaviours occurred on recording sheets. The children then went through a treatment phase that varied in duration across participants between 45 to 55 nights.

For the boy with PWS, treatment followed an AB-design (comprising a baseline phase followed by treatment) and consisted of a non-graduated extinction procedure wherein his parents bid him goodnight and then left him alone until morning. This technique was combined with the provision of positive attention in morning when he had remained quiet during the night. The behavioural techniques used to treat the boy with FXS were a desensitization procedure coupled with differential reinforcement of positive behaviour, as his functional assessment had indicated that his sleep problems were rooted in anxiety. For this boy's intervention, a B-design was followed (treatment phase followed by the cessation of intervention, before then reinstating intervention a second time). A clear, consistent, and calming bedtime routine was put in place. The child's bedtime routine consisted of 19 steps, including being taken to bed at 8pm, and his mother reading him a story in bed before being left to sleep. If he kept quiet and complicit for the entire duration of a step, he earned a preferred edible reward, and then they would continue with the next step in his routine. If he failed to do this during any step, his mother would stay with him in his bed until morning. The study included a follow-up period of about five days approximately six months after treatment had ended. The results for the participant with PWS showed a decrease in the number of minutes of nighttime disruption from an average of 90 minutes at baseline to an average of 22 minutes' post-treatment, and a further decrease to zero minutes at follow-up. The participant with FXS demonstrated a sudden increase in inappropriate bedtime behaviour with the reinstatement of intervention, followed subsequently by the successful completion of

the last step in his bedtime routine, along with the elimination of co-sleeping and all symptoms of anxiety (Didden et al., 1998).

Another study which carried out a multi-component behavioural treatment package was led by Allen and colleagues (2013). Through a multiple baseline design across participants, five children between 2 and 11 years with AS were assessed for co-sleeping, bedtime resistance, and night wakings using sleep diaries and actigraphy before individualized treatment packages were developed to optimize their sleep environment, sleep-wake schedule, and any parent-child interactions during sleep times. Some of the treatment techniques included delayed bedtimes, sleep restriction during the day, extinction procedures, and in cases where parents did not feel comfortable with the standard extinction procedure, a modified extinction procedure known as the “Excuse-Me Drill” was implemented. The “Excuse-Me Drill” involves periodic checking in on children in the time before sleep onset and during night wakings, contingent on the child’s demonstration of calm and quiet behaviour whilst staying in bed (Allen et al., 2013; Kuhn, 2011). In this way, parental attention and presence is purposefully utilized to reinforce sleep-conducive behaviour and provide the opportunity for self-soothing skills to be practiced, and subsequent independent sleep initiation at bedtime and reinitiation to take place in response to night-time wakings (Allen et al., 2013). The authors also requested that parents establish a sleep-compatible environment by keeping visual and auditory stimuli to a minimum (i.e., turning off television and stereo), removing any light sources to create a no light or low light setting, and adjusting the room temperature if needed. Following the commencement of treatment, all five participants showed decreases in disruptive behaviour and independent sleep initiation over the next month. Prior to intervention, all participants needed their parents to be with them in order to fall asleep. Within the first week of treatment, the children were all falling asleep independently. Throughout treatment and follow-up these effects were found to be stable and maintained (Allen et al., 2013).

Moss and others (2014) trialed the “Sleepwise” program (O’Connell, 2005) to address sleep problems of children with various developmental disabilities. 26 children between 8 to 18 years of age, of whom 5 were listed to have diagnoses of “syndromes including Angelman, Down and others”. The participants were randomized to a treatment or the wait list control group. The researchers measured child sleep habits using the Child Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000), child daytime behaviour with the Developmental Behaviour Checklist: Parent Version (DBC-P; Einfeld & Tonge, 2002), and parent stress with The Parenting Stress Index: Short Form (PSI-SF; Abidin & Brunner, 1995). Outcome measures were administered prior to the commencement of intervention (week 1), and post intervention at week 10 and week 18. The *Sleepwise* program (O’Connell, 2005) that was implemented with each of the participants included is a manualised intervention for sleep disturbance, and is formulated designed for youth with developmental delay. The program first involved two 3-hour educational workshops that provided psychoeducation about sleep, atypical sleep, and sleep treatment. Second, a comprehensive sleep assessment was undertaken, which included an interview. Lastly, an individualized program and plan was developed for each child using the information gathered through assessment. Average total sleep disturbance scores in the treatment group decreased from 56.20 at baseline to 46.50 post-treatment. This compared to the wait-list control group scores which showed no significant change (51.38 pretreatment and 51.12 at post-treatment). Total Problem Behaviour scores decreased from 66.20 at baseline to 57.70 post-treatment in the treatment group, which contrasts with average scores of 72.29 at baseline and 69.25 post-treatment in the wait-list control group. These results indicate that daytime problem behaviour was no significantly changed as a result of treatment. PSI scores similarly showed no significant change from over the study phases in either group (Moss et al., 2014).

Multi-component behavioural interventions were formulated for a group of children with a mean age of 5 who had various developmental disabilities and were suffering from sleep disturbance (Weiskop et al., 2005). 7 of the 13 participants had FXS. The participants were initially assessed through an interview and sleep diaries and a functional assessment was completed. The study then followed a concurrent multiple baseline design, and began with a period of baseline, followed by at least seven weeks of treatment, and a 2-week follow-up period 3 months posttreatment. Sleep diaries were completed during all study phases by parents, who recorded the number of pre-sleep disturbances per week, number of nights per week that the child fell asleep alone in their own bed, average sleep latency, number of night wakings, number of nights per week that the child co-slept, and the average duration of night-time sleep. The intervention itself entailed 3 weekly individual parent-training sessions which covered topics such as goal setting, the principles of learning theory, the role of antecedents and consequences, individualized bedtime schedules and routines, reinforcement procedures, visual aids, partner support strategies, extinction techniques, and general psychoeducation around sleep. The participating families had weekly contact with therapist for support and guidance. Complete data was obtained from 5 of the participants with FXS. The results showed an overall improvement with regard to independent sleep initiation, co-sleeping, and night wakings across participants for whom these were presenting issues. The treatment effects appeared to have been maintained at follow-up for most participants. It should be noted however that these positive treatment effects emerged quickly following the introduction of an extinction procedure (Weiskop, Richdale, & Matthews, 2005).

Montgomery, Stores, and Wiggs (2004) delivered a behavioural sleep intervention to 66 families with severe learning disabilities including Angelman syndrome (n=2), Williams syndrome (n=1), and Fragile X syndrome (n=1), wherein parents were educated regarding good sleep habits. Sleep hygiene topics that were

covered included “creation of an appropriate sleep environment, prevention of daytime napping, the importance of clear routines, putting children to bed while awake but drowsy, removal of bottle, as well as how to deal with possible physical contributors of sleep disturbance (e.g., wet nappies, being too cold/hot). Following treatment, which also involved further psychoeducation, including information about extinction, and reinforcement procedures, a significant portion of the participating children demonstrated improvement in their sleep onset latency and night wakings. Those who had initially taken more than 30 minutes to fall asleep on at least 5/7 nights, reduced a sleep onset latency of only a few minutes, and/or demonstrated significant sleep onset delay only once or twice per week. A similar pattern of change was found for night waking in this group. Maintenance of these treatment gains was discerned at a three-month follow-up (Montgomery, Stores, & Wiggs, 2004).

The literature described above has demonstrated the effectiveness of behaviourally-based interventions used to treat children with RGND presenting with sleep problems. The positive treatment effects observed were also found to be maintained at follow-up in each of the studies which included follow-up assessment (Allen et al., 2013; Montgomery, Stores, & Wiggs, 2004; Weiskop, Richdale, & Matthews, 2005). Despite the strengths of this body of literature, which include the use of comprehensive assessment protocols using reliable and valid objective and subjective measures (and FBA in two instances; Didden et al., 1998; Weiskop et al., 2005), the implementation of multi-component intervention packages comprising empirically supported treatment procedures, and high quality methodological approaches, including randomization to treatment or control groups (Didden et al., 1998; Moss et al., 2014; Piazza, Fisher, & Sherer, 1997) and multiple baselines (Allen et al., 2013; Weiskop et al., 2005), it is not without its limitations.

Limitations of the Literature

Several methodological limitations should be considered when interpreting the existing research on this subject of behaviourally-based sleep interventions in their use with children with RGND, including small sample sizes, invariability in study designs, non-representative samples, unequal control and treatment groups, the lack of objective measures for primary sleep variables, the lack of measurement of parental wellbeing and general child behaviour effects of treatment, and the lack of utilization of FBA as an assessment protocol and tool for informing intervention formulation.

A considerable proportion of the studies found had a small sample size. Although single-case designs, which were a common study design utilized in the literature covered in this review, boost internal validity and allow for the clear demonstration of the functional relationship between treatment and outcomes, the generalizability or external validity of findings is concurrently restricted (Allen et al., 2013; Didden et al., 1998; Piazza, Fisher, & Sherer, 1997; Weiskop et al., 2005). A small sample also decreases statistical power and simultaneously increases margin of error (Faber & Fonseca, 2014). Secondly, the few studies which do include larger sample sizes do not always describe the RGND participants in isolation (Montgomery, Stores, & Wiggs, 2004; Moss et al., 2014). The results of the few participants with RGND in these studies are considered within group means, making it difficult to draw conclusions specifically pertaining to RGND. Therefore, despite the good internal validity afforded by single-case studies, the research area would certainly benefit from future assessment of the effects of behavioural sleep interventions in larger populations and using different study designs, such as randomized controlled trials or experimental designs which incorporate multiple baselines, to provide a more rigorous test of treatment efficacy (Smith et al., 2007).

Further diminishing the external validity of the research described in this review is the limited representation of different RGND diagnoses. PWS (Didden et al., 1998), FXS (Didden et al., 1998; Montgomery, Stores, & Wiggs, 2004; Weiskop et al., 2005), AS (Allen et al., 2013; Montgomery, Stores, & Wiggs, 2004; Moss et al., 2014), and Williams syndrome (Montgomery, Stores, & Wiggs, 2004) are the only RGNDs included in the studies in this literature review. The lack of diversity in diagnoses restricts the extent to which research findings can be generalized to other RGNDs, of which there are many. This is especially the case with children with RGND, considering the uniqueness in the diagnostic/clinical features across different RGNDs. As such, the efficacy of behavioural sleep interventions with other RGNDs still requires investigation.

Also, while not addressed by the authors in the article, Moss and colleagues' (2014) study found significantly higher scores (in the 'Clinically Significant' range) in the wait-list control group at all data collection time points compared to the treatment group could suggest that it was not an equivalent point of comparison and therefore any conclusions drawn regarding between-group results may be disputable. There should have been no significant difference between groups at baseline, so that it could be accurately inferred that differences between the groups following treatment were attributable to one group receiving the intervention and the other not receiving intervention, and not some other extraneous variables (Bailey, 2008).

Across half of the studies, there was an apparent reliance on parent report as opposed to more objective measures (Didden et al., 1998; Moss et al., 2014; Weiskop et al., 2005). While subjective, self-report measures have their value, they are subject to bias and therefore call into question the generalizability and certainty of results, especially when reliability data is not collected and analyzed, as was the case in these studies (Turner & Johnson, 2013). Future research needs to prioritize

the triangulation of multiple methods of assessment, including objective measures, in order to procure more credible results.

There was only one study which included measures of parental wellbeing and general child behaviour variables prior to and following intervention (Moss et al., 2014). These are important secondary treatment effects which future empirical research should investigate, as all behavioural interventions should endeavor to have a positive impact on the broader quality of life of the child and their family. By including measures of these variables in intervention studies, it may be ascertained whether the effect of treatment does in fact have further reaching repercussions for general child behaviour and parental wellbeing variables, or whether adjustments could be made to the intervention to maximize the benefit in this regard.

FBA was similarly underused in past literature on the effectiveness of behavioural sleep interventions for children with RGND, with only two studies (Didden et al., 1998; Weiskop et al., 2005) reporting to have employed the assessment protocol. Both studies demonstrated the utility and benefits of conducting FBA to not only assess pre-treatment sleep difficulties and their related functions, but to inform the development of treatment plans for participants. Future research should heed the endorsement that these and other studies treating the sleep problems of children with developmental disabilities (Brown et al., 2013; Didden & Sigafos, 2001; Hanley, 2016; Hanley et al., 2014; Kodak & Piazza, 2008) give to FBA and incorporate it into methodologies in order to enhance the empirical evidence-base for the procedure.

All in all, while the existing literature presents promising findings which point to the potential efficacy of behaviourally-based sleep interventions for children with RGND, it is evident that more research is needed to properly gauge the effect of

such treatments in addressing the sleep difficulties of children in this particular population.

Rationale

The review presented in this chapter has highlighted the literature on the effectiveness of pharmacological and behaviourally-based sleep interventions used in the treatment of sleep problems in children with RGND as it currently stands. The studies that met the inclusion criteria have demonstrated positive sleep outcomes through the implementation of a small selection of medications, including melatonin (Braam et al., 2008; Gringas et al., 2017; McArthur et al., 1998; Miyamoto et al., 1999; Zhdanova et al., 1999), human growth hormone (GH) replacement therapy (Itoh et al., 2004), and diphenhydramine HCl (Summers et al., 1992), antecedent-based procedures, such as sleep hygiene modification (Allen et al., 2013; Didden et al., 1998; Montgomery, Stores, & Wiggs, 2004; Weiskop, Richdale, & Matthews, 2005), establishing consistent sleep-wake times (Allen et al., 2013; Piazza et al., 1997), and bedtime fading (Allen et al., 2013; Piazza et al., 1997). While generally positive treatment effects were found across the reviewed studies, the extant literature on the subject is minimal and suffers from a number of limitations. This, in conjunction with the high prevalence of sleep disturbance within this population, and the considerable direct and indirect consequences of such issues on the affected children and their families, provides abundant justification for the expansive investigation of the effectiveness of FBA-based behavioural interventions at treating the sleep disturbance of children with RGND. Given such, this study will add to the literature on this subject by investigating the effectiveness of FBA-informed, behaviourally-based interventions in the treatment of sleep disturbance amongst children with RGND, as well as examining any changes to child and parent wellbeing and quality of life following the sleep intervention. Additionally, this research will establish the treatment acceptability of the interventions implemented by parents with children with RGND experiencing sleep problems.

This study is guided by three research questions:

1. Are FBA-based, behavioural sleep interventions effective in the treatment of sleep difficulties in children with RGND?
2. Is parental wellbeing and general child behaviour effected by FBA-based, behavioural sleep interventions for children with RGND experiencing sleep disturbance?
3. Are FBA-based, behavioural sleep interventions, and the assessment and treatment processes carried out as part of said treatment pursuit, acceptable to parents of children with RGND?

Chapter 3

General Method

The Sleep Research Team

This investigation into the effectiveness of FBA-informed treatments for sleep disturbance in children with rare genetic neurodevelopmental disorders (RGND), is part of an expansive programme of research being undertaken by a team of researchers at the University of Canterbury. The wider research study endeavours to explore behaviourally based assessment and treatments for sleep problems in children with developmental disabilities. The wider research team is led by senior researchers and also includes a number of Masters and PhD students, intern and registered psychologists, and research assistants.

Ethics and Participant Consent

Ethical approval for this study was obtained from the University of Canterbury Human Ethics Committee (HEC 2018/48). Parents in this study provided written informed consent, and children provided assent, in alignment with their developmental level. Attached in Appendix A is a copy of the child's information sheet and in Appendix B is the child assent form. Copies of the parent information sheet and the parent consent form are included in Appendices C and D respectively. Additional parental consent was provided for the video recording of children's sleep. The child and parent audiovisual recording consent forms can be found in Appendix E and Appendix F.

Design

The current study followed a multiple-baseline-within-participants design. This type of research design allows researchers to examine individual participant's intervention outcomes, as a measure of change from baseline. Single-case multiple

baseline designs also enable conclusions to be drawn from the replication of treatment effects across multiple participants. Given the distinctiveness of each child's background, familial context, RGND diagnosis, and presenting sleep problems, and the need for individualization of treatment plans, a single-case multiple baseline research design was selected. When best practice procedures are adhered to, participants, inclusion/exclusion criteria, setting, dependent variables and their measurement, and independent variables are meticulously described in single-case research designs (Horner, Carr, Halle, McGee, Odom, & Wolery, 2005). Such thorough examination facilitates tight experimental control over extraneous variables and yields high internal validity (Cohen, Feinstein, Masuda, & Vowles, 2014; Kratochwill et al., 2013; Sidman, 1960). A single-case research design also allows for flexibility with regard to modification to independent variables amid the study, such as altering components of a treatment. This is particularly useful in applied behavioural research (Cohen, Feinstein, Masuda, & Vowles, 2014).

Data Analysis

Graphs were created which represented the data collected over the baseline, intervention, and follow-up phases for each of the targeted dependent variables for each child. Dependent variables that were graphed included sleep onset latency (mins), frequency of night wakings, duration of night wakings (mins), and the percentage of total sleep time spent co-sleeping. Systematic visual analysis of the graphed dependent variables over the course of each study phase within cases, as well as across participants, constituted the main method of data analysis in this study. Recognition of visual analysis as being an effective method of assessing treatment effects/outcomes spans the field of experimental research, particularly in single-case multiple baseline across participant studies where the method is commonly employed (Blampied, 2013; Hanley et al., 2003). Within the current study, visual analysis made it possible determine whether or not change in behaviour over

time could be attributed to the intervention. Visual inspection of the mean, level, trend, variability, latency, and consistency of sleep-related behaviours were all factors that were examined (Cohen, Feinstein, Masuda, & Vowles, 2014).

Analysis of pre- and post-treatment scores on all psychometrics, namely the CSHQ, VABS-II, DASS-21, PSQI, RQI, and CBCL, as well as the calculations of SPS scores, were compared within and between participants in order to ascertain whether any of the child or parent variables measured by the questionnaires changed as a consequence of intervention.

Psychometrics completed by families at a single time-point, either at pre- or post-treatment (i.e., the QABF and TARF-R) were also examined.

Participants

Recruitment. Participants were recruited throughout New Zealand. Flyers providing information about the study were disseminated to relevant organisations and agencies that provide services for children with RGND and their families (e.g., the Angelman Network, Fragile X New Zealand Trust, and the Prader-Willi Syndrome Association of New Zealand). These organisations were asked to share study information across their networks, with those who may be interested. Study flyers invited participant self-referral or referral from relevant organizations or service providers.

Screening and confidentiality. Potential participants were screened over the telephone, to determine whether they were eligible for inclusion in the study. Prior to asking screening questions, parents/caregivers were provided with a brief explanation of the purpose of the study and relevant procedures and matters related to confidentiality and anonymity were discussed. Parents were then asked a series

of questions to determine eligibility for inclusion in the study. The screening call took approximately 15 minutes.

Inclusion/exclusion criteria. Children were deemed eligible to participate in this study provided that the following inclusion criteria were met: (a) they were between the ages of 2 and 18 years; (b) they had a diagnosis of a Rare Genetic Neurodevelopmental Disorder, as verified by a paediatrician, psychiatrist, registered psychologist, or other relevant medical professional; (c) they presented with parent-reported sleep problems, including delayed sleep onset latency, bedtime resistance, frequent or prolonged night wakings, and/or unwanted co-sleeping. Children were excluded from the study if they had physical or medical comorbidities that may have compromised the effectiveness of treatment or that may have made it unsafe to implement intervention. Similarly, children were not included if it was established from the initial screening contact or clinical interview that the parents were unable or unwilling to adhere to the assessment or treatment processes.

Participant characteristics. The participants included in this study were two boys and one girl ranging from 6 to 12 years of age. The children presented with a diverse array of RGND diagnoses, specifically FXS, PWS, and a child with various chromosomal deletions (details omitted given the rarity of the disorders and potential for identification of participants). Details such as individual ethnicity and actual names have not been reported, to ensure the protection of participants' confidentiality and anonymity, and pseudonyms have been used in place of the children's actual names. Participant characteristics are summarized in Table 1.

Table 1. *Summary of Participant Characteristics*

Name	Sex	Diagnosis	Mode of diagnosis	Comorbid health issues	Presenting sleep problems	Past and present sleep medication	Previous sleep intervention attempts	Pre-treatment VABS-II scores (<i>Note: X-X = year-month</i>)	Pre-treatment CBCL scores (<i>Note: R= Range; N=Normal; B=Borderline; C=Clinical</i>)
Hannah	Female	PWS	Pediatrician	Cataplexy	Night wakings, co-sleeping, early morning waking, day-time napping	N/A	Groclock	Receptive communication = 2-4; Expressive communication = 3-11	Internalising behaviours = 59 (N); Externalising behaviours = 73 (C); Total Score = 72 (C)
Michael	Male	Chromosomal abnormality *	Geneticist	Dairy allergies, eczema, hay fever, mild asthma	Bedtime resistance, sleep onset delay, night wakings, co-sleeping, day-time napping	Melatonin (past), Phenergan (past), Vallergran (past)	White noise, Groclock	Receptive communication = 2-3; Expressive communication = 1-10	Internalising behaviours = 52 (N); Externalising behaviours = 57 (N); Total Score = 58 (N)
Robert	Male	FXS	Pediatrician	N/A	Sleep onset delay, night wakings, co-sleeping, early morning waking	N/A	Music, sitting with him, rocking him	Receptive communication = 1-8; Expressive communication = 1-11	Internalising behaviours = -; Externalising behaviours = -; Total Score = -

* = due to the rarity of Michael's chromosomal abnormality, the specific details of his diagnosis have been withheld to ensure that the participant is unable to be identified.

Setting. As the participating families all lived outside of Christchurch, the clinical intake interviews and contact across study phases, was conducted via telephone or Skype™. Resources and equipment were delivered via post or email. The interventions were all implemented by the primary caregiver/s and extended family members of the children, and conducted in the family home. Ongoing communication between members of the sleep team and the families was facilitated by phone, text, and/or email.

Measures

FBA measures. A combination of clinical interviews, analysis of video content and sleep diaries, and data from the Sleep Assessment Treatment Tool (SATT; Jin et al., 2013) and the Questions About Behavioural Function (QABF; Mason & Vollmer, 1995) were used to inform each child's FBA. Conclusions drawn from the FBA aided in the development of comprehensive treatment plans tailored to each individual child.

Sleep Assessment Treatment Tool (SATT; Jin et al., 2013). Data from the clinical interview pertaining to the child's sleep was used to complete the SATT. The SATT enabled sleep problems and parent goals to be clearly identified and defined, for the purpose of guiding the FBA. The SATT includes questions aimed to assess: a) the history of the presenting sleep problems; b) parents sleep-related treatment goals; c) the specific sleep problems, including bedtime routine resistance, sleep onset delay, sleep interfering behaviour, co-sleeping, night wakings, and early wakings; d) any antecedents or consequences occurring before or after the behaviour that parents can identify; e) the sleep schedule currently in place for the

child; f) the child's routine leading to eventual sleep onset; g) the child's sleep environment; and h) any sleep dependencies.

Questions About Behavioural Function (QABF; Mason & Vollmer, 1995). The QABF is a 25-item psychometric that aids in hypothesizing about the function of target behaviour (Freeman, Walker, & Kaufman, 2007; Healy, Brett, & Leader, 2013; Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000). The QABF comprises five subscales: Social Attention, Escape, Non-social Reinforcement, Physical Discomfort, and Tangible Reinforcement. Each subscale contains five items, which are scored on a four-point scale, indicating the frequency with which each item is applicable: Doesn't apply (Never), 1 (Rarely), 2 (Some), or 3 (Often). Subscale scores and their corresponding severity ratings are calculated separately.

The QABF is a standard measurement component of functional behavioural assessment, and has the strongest psychometric properties of all functional assessment scales currently (Matson, Tureck, & Rieske, 2012). The QABF has good test-retest reliability ranging between 0.81 to 0.82, moderate to good interrater reliability from 0.63 to 0.68, and good internal consistency across the five subscales, ranging from 0.89 to 0.96 (Freeman et al., 2007; Healy et al., 2013; Matson et al., 2012; Paclawskyj et al., 2000; Zaja et al., 2011).

The QABF was completed by one parent during baseline, focusing on a specified target behaviour.

Primary sleep measures.

Video recordings. Night-time video recordings were made using either a Swann-Advanced-Series DVR4-1200 camera, or a D-Link HD Cloud Camera. All cameras were infrared and had the capacity to record multiple nights. A monitor was

included in the DVR4 package, and connecting this to the camera made it possible for parents to check live footage. Parents were instructed to situate the video camera so that it was inconspicuous to the child, and so that it gave a clear and unobstructed view of the child and their bed. Video recordings made by the DVR4 camera were stored in an internal hard drive, and a micro SD card for the DSLR cameras. Families were provided with a set of written instructions with corresponding photographs to guide the set-up of the video equipment. Those families that received a DVR4 camera were told to turn the camera on just before they bid their child goodnight and to turn off the camera when their child woke for the day the following morning. The D-Link cameras were preset to record from half an hour before the child's specified bedtime to one hour after their anticipated morning wake time. The video data was intended to supplement the subjective measures used throughout study phases, such as sleep diaries. Video recordings allow the child's behavior and sleep-wake phases to be coded objectively, by an independent observer in order to fill in any information not reported in the diaries and to collect interobserver agreement data. Video recordings were obtained for a minimum of 30% of nights.

Sleep diaries. Sleep diaries are a popular tool for examining children's sleep (Blampied, 2013; France & Blampied, 2005; McLay & France, 2016). The diaries were formatted with columns designated for each day of the week, which were intersected by rows that allowed for parents to report such dependent variables as day-time sleep (including the setting, time asleep, and time awake), night-time sleep (including setting, time put to bed, frequency of curtain calls, nature of curtain calls, parental responses to curtain calls, and estimated time of sleep initiation). On a second attached sheet, and in a continuation of the same format, rows provided space for parents to note down two night wakings, each including cells for description of the time and duration of the waking, the child's behaviour while awake, and the parents' responses. Then lastly, a row was included for the reporting of morning wake time. All requests for the duration of wakings were in the unit of

minutes. At the end of the two-page document was a definition of curtain calls for the parents' reference, and space for any additional notes. Parents could fill in the sleep diaries by writing a description of behaviours. An example of a sleep diary used by families participating in the study is attached in Appendix G. Parents were asked to complete sleep diaries each night across study phases. Collection of sleep diaries during the assessment phase helped to ensure that the families understood how to complete the diaries accurately, while also providing valuable information that would be used to develop the child's treatment plan. Over the course of the intervention phase, the researcher and/or psychologist would maintain regular contact with the families to collect the latest sleep diaries. The frequent sharing of sleep diary data ensured that the sleep team had up to date information regarding the children's progress, and were therefore able to promptly react to the children's responses to intervention, and adjust the intervention plan when necessary. Sleep diary data was graphed and analyzed visually for the purposes of assessment and the monitoring of progress.

The researcher provided the families with sleep diaries either in printed form or as an electronic copy shared over email, and explained to the parents how to fill in the necessary information.

Sleep Problem Severity Score. A score was calculated which summarized the severity of the children's total sleep disturbance at each measurement time-point in the study, in emulation of the methodology carried out by Lawton, France, and Blampied (1991). For each sleep variable, a score of 1 is given if there was an occurrence of that sleep problem to any extent during the night (i.e., co-sleeping at sleep onset = 1; co-sleeping following NW = 1; SOL > 15 min = 1; and no NW = 0), and conversely if a feature of sleep disturbance was not present, a score of 0 would be given. The scoring of the duration of NW however followed a different method, with total NW duration < 5 min receiving a score of 1; or total NW duration > 5 min receiving a score of 2. The maximum possible score per night was 5. All of the total

nightly scores over the course of each study phase were summed to give an SPS score for baseline, intervention, and follow-up (Lawton, France, & Blampied, 1991).

The Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000). The CSHQ is a 45-item parent-report measure that is used to identify sleep problems such as bedtime resistance, sleep onset delay, sleep duration and night waking (Owens et al., 2000). One parent was instructed to complete the CSHQ at assessment and again during maintenance in the current study. The CSHQ constitutes eight subscales which target specific sleep-related problems. These subscales include *Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness*. Parents indicate the frequency of various sleep behaviours exhibited by their child over the past week on a three-point scale - 'usually' (5-7 nights per week), 'sometimes' (2-4 nights per week), or 'rarely' (0-1 night per week). The questionnaire also asks parents to specify whether each of their child's sleep behaviours are problematic for the family. The eight scores derived from each subscale are added together to obtain a total sleep disturbance score.

The CSHQ has been administered extensively in sleep research involving typically developing children (Krakowiak et al., 2008; Markovich, Gendron, & Corkum, 2015), and children with neurodevelopmental disorders such as ASD (Lambert et al., 2016; May et al., 2015; Mazuerk & Sohl, 2016). The CSHQ is widely used in clinical practice and research and has acceptable psychometric properties (Hodge, Parnell, Hoffman, & Sweeney, 2012; Hoffman, Sweeney, Gilliam, & Lopez-Wagner, 2006). The questionnaire has a sufficient internal consistency in a clinical sample ($\alpha=.78$), as well as a community sample ($\alpha=.68$) (Owens et al., 2000). The CSHQ also has adequate test-retest reliability (ranging from .62-.79) (Owens et al., 2000). On the matter of validity, the CSHQ is capable of differentiating between

clinical and control groups, where sensitivity was 0.80 and specificity was 0.72 (Owens et al., 2000).

The CSHQ was administered during baseline, and upon completion of treatment. The repeated measurement allowed for the assessment of change in dependent variables.

The outcome of the measures discussed in the above section informed the development of children's individualized treatment plans.

Measures of parental sleep, well-being, and relationship satisfaction. In the current study, the DASS-21, RQI, and PSQI were administered to all primary caregivers of participating children during assessment and maintenance to assess any change in levels of parental depression, anxiety or stress, perceptions of parental relationship quality, and parent sleep quality, following the intervention.

Pittsburgh Sleep Quality Index (PSQI; Buysse, Reynolds, Monk, Berman, & Kupfer, 1989). The PSQI is a self-report measure of sleep quality in adults containing 18 items targeting the frequency with which certain sleep behaviours occur, as well as sleep quality, sleep onset, sleep efficiency, sleep disturbance, sleep medication, and daytime sleepiness (Buysse et al., 1989). In the current study, the PSQI was administered during assessment and maintenance to all primary caregivers. Four items are posed in question-form and respondents are instructed to answer them in reference to their sleep over the past month. 13 items relate to the frequency of sleep behaviours and are rated on a four point Likert scale ranging from 0 (not during the past month), 1 (less than once a week), 2 (once or twice a week), or 3 (three or more times a week). The final item asks for a rating of the respondent's overall sleep quality and is rated as either 0 (very good), 1 (fairly good), 2 (fairly bad), or 3 (very bad). A global score is determined by adding seven component scores together.

Reports of satisfactory psychometric properties have been made, including an internal reliability of .83, and a test-retest reliability of 0.85 (Buysse et al, 1989). The PSQI is frequently use in both clinical and research settings to measure the sleep quality of parents and children, typically developing or with developmental disabilities such as ASD (Carpenter & Andrykowski, 1998; Giallo, Wood, Jellet, & Porter, 2011; Hodge et al., 2013; Hoffman et al., 2008; Lopez-Wagner et al., 2008; McBean, Schlosnagle, 2016; Meltzer, 2008).

The Depression Anxiety and Stress Scales (DASS-21; Lovibond & Lovibond, 1995). The DASS-21 is a self-report measure containing 21-items relating to symptoms of depression, anxiety, and stress (Henry & Crawford, 2005). All primary caregivers in the current study completed the DASS-21 during assessment and maintenance phases. The DASS-21 requires the informant to note the extent to which the various statements were relevant for them over the past week. Respondents rate each ... on a four-point scale – ‘0’ (did not apply to me at all), ‘1’ (applied to me to some degree, or some of the time), ‘2’ (applied to me a substantial amount of the time), and ‘3’ (applied to me most of the time). In scoring the DASS-21, severity levels for the depression, anxiety, and stress axes may be determined, and severity labels of “normal”, ‘mild’, ‘moderate’, ‘severe’, or ‘extreme’ are provided.

The DASS-21 has demonstrated good psychometric properties, particularly with respect to reliability ($\alpha=.82-.90$ for the subscales), internal consistency (ranging from 0.87 to 0.94 across subscales) (Antony, Bieling, Cox, Enns, & Swinson, 1998), and convergent and discriminative validity (Henry & Crawford, 2005). Use of the DASS-21 has been widely used with adult populations in clinical and research contexts, and with samples of parents of children with neurodevelopmental disorders such as ASD (Al-Farsi, Al-Farsi, Al Sharbati, & Al-adawi, 2016; Giallo et al., 2011).

Relationship Quality Index (RQI; Norton, 1983). The RQI is a tool that is a 6-item measure of a couple's perceptions of their satisfaction with their relationship, as well as their perception of the quality of their relationship (Sanders, Markie-Dadds, & Turner, 2001). For two of the participants in the current study, the RQI was administered to both of their parents during assessment, and then again during maintenance. Participants rate the degree to which they are in agreement with various statements concerning their partner and relationship. Ratings are given on a 7-point Likert scale (1 = very strongly disagree through to 7 = very strongly agree). Global relationship satisfaction is ascertained through the summing of scores, with higher scores denoting higher satisfaction.

Adequate psychometric properties have been established for the RQI, including an interrater reliability of .70, and internal consistency ranging from .25 to .65, which researchers have contended illustrates the RQI's ability to measure elements of relationship quality that are simultaneously related and yet distinct (Lawrence, Brock, Barry et al., 2008).

Measures of General Child Behaviour. The VABS-II and the CBCL were administered by an intern psychologist or researcher at the baseline and maintenance phases of the study to provide an impression of potential secondary effects of treatment.

The Vineland Adaptive Behaviour Scales-II, Parent/Caregiver Rating Form (VABS-II; Sparrow, Cicchetti & Balla, 2005). The VABS-II measures a child's adaptive functioning, including their level of proficiency at navigating social contexts and functioning within their everyday environment (Gleason & Coster, 2012; Sparrow et al., 2005; Tassé et al., 2012). The the Communication Domain segment of the VABS-II was completed at pre- and post-intervention by one parent. It was used to measure the participants' receptive and expressive communication abilities prior to

and following intervention for their sleep problems. The VABS-II is a parent/caregiver report form for individuals between 0 to 90 years of age. Parents were instructed to select the option that best describes the frequency with which their child engages in certain behaviours using a three-point scale, where '*usually*' is coded as '2', '*sometimes or partially*' is coded as '1', and '*never*' is coded as '0'. The questionnaire also provides a '*don't know*' option in instances where the parent or caregiver is unsure of the frequency of a particular behaviour.

The VABS-II has been used extensively in research and clinical practice, and consequently has acquired a considerable amount of normative data (Achenbach & Rescoria, 2001; Tassé et al., 2012). Good psychometric properties have been found for the VABS-II, including an internal consistency of .93 to .97 across age groups, and a test-retest reliability of ranging between .76 and .92 across the five domains (Sparrow, Cicchetti, & Balla, 2005). There has been ample utilization of the VABS-II in populations of children and adolescents with intellectual disabilities, independence issues, and developmental disabilities, including autism (Gabriels et al., 2005; Gleason & Coster, 2012; Sikora et al., 2012; Tassé et al., 2012). The measure has shown good reliability in being able to differentiate clinical and non-clinical samples, with clinical groups reliably producing an adaptive behaviour composite score approximately two standard deviations below the non-clinical mean, whilst still generating distinct profiles across different clinical groups (Sparrow, Cicchetti, & Balla, 2005).

Child Behavior Checklist (1 ½ - 5 years) and (6 – 18 years) (CBCL (1 ½ -5), Achenbach & Rescorla, 2000; CBCL (6-18), Achenbach & Rescoria, 2001). The CBCL (1 ½ - 5) is a standardized measure based on parent report that includes 100-items assessing internalizing and externalizing behaviours in preschoolers aged between 18 months and 5 years. For older children (6 to 18 years), the school-age form of the CBCL is administered to either parents, or teachers. Parents noted the

frequency of behaviours on a three-point Likert scale: 0 (not true), 1 (somewhat or sometimes true), or 2 (very true or often true). Across the seven syndrome scales (emotionally reactive, anxious/depressed, somatic complaints, withdrawn, sleep problems, attention problems, and aggressive behaviour) and five scales associated with DSM-5 diagnoses (depression, anxiety, ASD, ADHD, and oppositional defiance), scores are added up and converted into T scores. T scores from the various scales are combined to provide internalizing problems, externalizing problems and total problems composite scores, which indicate problem severity as falling within normal, borderline, or clinical ranges.

Adequate reliability and validity have been demonstrated with the CBCL. An internal consistency of .63 to .97 across scales has been found (Tehrani-Doost, Shahrivar, Pakbaz, Rezaie, & Ahmadi, 2011), test re-test reliability ranging from .80 to .94 (Achenbach & Rescoria, 2001), and good convergent and divergent validity (Nakamura, Ebesutani, Bernstein, & Chorpita, 2008). The CBCL has been commonly used in the assessment of sleep problems in populations with ASD (Anders, Iosig, Schwichtenberg, Tang, & Goodlin-Jones, 2012; Delahaye et al., 2014; Fadini et al., 2015; Goldman et al., 2009; Hollway, Aman, & Butter, 2013; Lambert et al., 2016; Moon et al., 2011; Sikora, Johnson, Clemons, & Katz, 2012).

Treatment acceptability measures.

Treatment Acceptability Rating Form – Revised (TARF-R; Reimers & Wacker, 1992). The TARF-R is a brief 20-item questionnaire used to measure treatment acceptability ratings (Reimers & Wacker, 1992). In the current study, parents completed the TARF-R during the maintenance phase. For 17 of the items, parents were asked to rate their perception of how appropriate, fair, and effective the intervention was. Parents' perception of the severity of their child's present behaviour and parents' understanding of the intervention procedures was assessed in the remaining three items. The 20 items of the TARF-R were rated using a 7-point

Likert scale. The questionnaire produces scores that are added to give a total acceptability score.

The TARF-R has been shown to have good reliability ($\alpha=.92$) and clinical utility (Finn & Sladeczek, 2001; Reimers & Wacker, 1992). The measure of treatment acceptability has been used in studies examining the effectiveness of treatments for challenging behaviours in many different populations, including children with autism (Lee, Anderson, & Moore, 2014; McLay, Carnett, van der Meer, & Lang, 2015). The TARF-R and post-treatment interview were completed during the maintenance phase.

Post-treatment interview. On conclusion of intervention, a semi-structured interview was undertaken by a member of the research team who had not had any previous contact with the the family. During this interview parents were asked about their experience with the assessment and treatment process, the degree to which they were satisfied/dissatisfied with the treatment outcomes and their thoughts on how the treatment process could have been improved.

Dependent Variables

The type and topography of the sleep problems and parents' treatment goals were used to determine the dependent variables measured for each child. Dependent variables were recorded from the beginning of the bedtime routine to the time that the child woke for the day. Common dependent variables are defined in the following paragraphs.

Awake. To be considered *awake*, a child would have to exhibit open eyes, and/or some form of sleep-interfering behaviour (as defined below), vocalization, or excessive bodily movements that would suggest a wakeful state.

Asleep. To be deemed asleep, a child would need to be lying in bed with closed eyes, and not voluntarily moving about or vocalizing. Movements that were characteristic of REM sleep were differentiated from wakeful movements.

Co-sleeping. A child would be reported to have engaged in co-sleeping if they lay in the same bed with another person for any length of time during the night. This included the sleep onset period as well as co-sleeping in response to a night waking. Co-sleeping may have been initiated by the child or parent. Co-sleeping was determined by calculating the percentage of total sleep time that was spent sleeping with a parent.

Parental presence. Parental presence included any occasion in which a parent was in physical or visual vicinity of the child during the onset of sleep (e.g., sitting in a chair beside the bed). This dichotomized as a presence or absence of a parent.

Sleep-interfering behavior. Sleep-interfering behaviour encompassed any behaviours that occurred upon being bid goodnight that interfered with the with the child's ability to establish the behavioural quietude needed to initiate sleep. For example, vocalising (e.g., talking, screaming, crying, calling out, singing, humming, or laughing), physical actions (e.g., sitting, standing, getting out of bed, playing with objects) and stereotypic behaviours (e.g., repetitive movements).

Sleep onset latency. Sleep onset latency was defined as the amount of time in minutes, that elapsed between the child being bid goodnight, and the onset of sleep.

Night waking. A night waking was defined as an arousal that occurred following initial sleep onset that interrupted a period of sleep that lasts longer than 2 minutes. Any waking that took place before the child's agreed morning wake time and/or that was prior to 6am was classified as a night waking. The frequency and duration (in

minutes) of a night waking was recorded in sleep diaries. The duration of a night waking was calculated by adding up the time spent awake during every NW that occurs over the course of an evening.

Curtain calls. Curtain calls were defined as behaviours that occurred during the initial sleep onset period, which served the purpose of obtaining parental attention (e.g., calling for a parent to enter their bedroom, leaving their bedroom). The child may have remained in bed or left their bed/bedroom in order to access parental attention or preferred items. The frequency of curtain calls was recorded in sleep diaries.

Procedure/Study Phases

Participants and their families persevered through each phase of the study, beginning with an initial assessment, followed by baseline, intervention, maintenance, and short-term follow-up phases.

Assessment. The assessment phase included a clinical interview undertaken by a registered intern psychologist. The open-ended clinical interview followed the structure of the standard intake interview used at the Pukemanu Dovedale Centre Clinic at the University of Canterbury. The clinical interview sought to obtain details concerning any previous attempts made to improve sleep, medical or physical conditions that may contribute toward the child's sleep problems, the child's RGND diagnosis, and other relevant contextual information about the child's life and family. On conclusion of the clinical interview, parents were given the opportunity to ask any questions that they had. The interviews were conducted via phone call or Skype. The clinical interviews were usually 1 ½ hours in length, with flexibility to suit the time-restraints of the families, as well as the variable duration of the open-ended dialogue. Examples of questions used in the clinical interview are included in Appendix H. The assessment phase also included the administration of the FBA

measures, sleep outcome measures, and measures of children's general behaviour and daytime functioning. Also completed during the assessment phase were a selection of parental sleep quality, parent well-being, and relationship quality measures. Data gathered during the clinical interview was used to aid in the formulation of individualised treatment plans.

Baseline. Sleep patterns and sleep-related behaviours were established during the baseline phase using a combination of daily sleep diaries and video footage. Parents were instructed to record daily sleep diaries along with video during their child's assigned baseline period. Children were randomly assigned to a baseline period of one, two or three weeks. The baseline start date was arranged so that it would be followed immediately by the commencement of intervention. During baseline, parents were asked not to make any alteration to their child's sleep schedule, bedtime routine, or their responses to their child's sleep-related behaviour. This stipulation was put in place to ensure that any change in the dependent variables that were observed following the commencement of intervention could be attributed to the intervention procedures, rather than variations in the environmental context or parents' behaviour.

Intervention. Intervention began immediately following the end of their assigned baseline period. Each family was provided with an individually-tailored, multi-component intervention plan which was informed by the outcomes of the FBA. The general goal across the participating families was to increase sleep conducive behaviours, and decrease sleep interfering or sleep incompatible behaviours. During the preceding baseline phase, families were provided with a proposed treatment plan, which described the particular intervention methods and instructions for their implementation. Families had the opportunity to comment on the drafted plan and offer any input before the final intervention plan was put into effect. Once the intervention plan was agreed upon by all parties, families were provided with the

resources necessary to carry out their child's intervention (e.g., social stories, Grocloclock).

Throughout the intervention phase, frequent contact was maintained between the researcher or psychologist and the family, via phone calls or emails. During the early stages of intervention, daily contact was maintained with the families. Sleep diaries were also received on a weekly basis, which permitted the allowed for close monitoring of progress and any problems that may have been encountered during intervention. If necessary, this made it possible to amend the intervention plan in a timely manner. Ongoing communication between the families and the research team also allowed for encouragement and emotional support to be offered to the parents. It was thought that this would bolster their sense of self-efficacy and competence, in turn increasing the likelihood of treatment adherence and desired treatment outcomes (Sanders & Burke, 2014).

Interventions were implemented until the sleep problem had been resolved, the family and/or the researchers were content with the level of improvement in sleep-related dependent variables, or until the parents decided to withdraw from the study. At this point, the family moved into the maintenance phase.

Common treatment components. There were a number of common treatment components across participants. This included social stories, use of visual aids as discriminative stimuli to facilitate the understanding of appropriate sleep and wake times, modification to sleep/wake schedules, faded bedtime, and rewards.

Social story. Each of the children participating in the study were provided with an individually-designed social story to use during intervention. Social stories generally depicted the steps in the bedtime routine, any changes in sleep setting, changes in expectations/goals, visual supports to be used, and reinforcement

contingencies. Parents provided the images but the social stories were constructed by the researcher. As is recommended by Gray (2010), the written text that accompanied the photographs was kept very brief, developmentally-appropriate, and was written in the first-person. These guidelines aim to hold the attention of the child and facilitate comprehension (Gray, 2010). Social stories are read with the child each night to help them to acclimate to their new sleep routine and any rules regarding their night-time behaviour by providing information relating to any who, what, where, and why questions that the child might have (Gray & Garand, 1993). The stories were presented in the form of a laminated and bound booklet containing photographs and accompanying text representing and describing each step in the child's bedtime routine, how they are advised to behave during night wakings, and any rewards that would follow the successful completion of this routine. The photographs showed the child him or her-self modelling the various steps in their routine, as well as images of any relevant sleep settings or items. The text was worded in a first-person, present tense narrative, and framed in a positive and empowering manner. The social stories were made to be concise, only including and describing steps that were necessary, as well as developmentally-appropriate for the individual child.

Discriminative stimuli for sleep and wake time. Visual aids and Groclocks were used with the children as prompts to help them learn to differentiate appropriate sleep and wake times. Two of the three children's intervention plans involved a Groclock. A Groclock has a screen which changes at preset times, showing a sun when it is time to wake up and a star when it is time to sleep. Groclocks provide a discriminative stimulus for the child to support them to distinguish between sleep and wake time. In the instance of bedtime resistance, a night waking, or early morning waking, parents could direct their child's attention to the Groclock and explain to them that they can "get up when the sun comes up", or point to their visual aid (in the

case of the other participant), which displays a sun, and explain that they are not to get up for the day until their surroundings resembled those shown in the visual aid.

Establishing a consistent sleep/wake schedule. A consistent sleep schedule was established through the enforcement of consistent sleep and wake times, and the elimination of daytime naps. These methods were implemented for all three children and served to establish a consistent sleep pattern by instigating a fixed duration of sleep. Parents were instructed to put their child to bed at specified times, and similarly wake them up at certain times in the morning. With regard to the prevention of sleep during the day, this was prescribed so that sleep pressure could be built in the lead up to bed time, as well as to facilitate good sleep hygiene practices at night time. The children were provided with preferred alternative activities to promote wakefulness in these instances, such as snacks and games.

Faded bedtime. Faded bedtimes were used with all three of the participating children and involved the shifting of a child's pre-intervention bedtime to a later time. The goal with this treatment method is to decrease sleep onset latency, with the child initiating sleep shortly after being bid goodnight by their parent. By delaying the child's bedtime, their need for sleep is increased and in turn so is the likelihood of them falling asleep quickly. The exact time of the delayed bedtime was established as being within 15 minutes of the time that the child typically fell asleep during baseline. Over the course of the intervention phase, faded bedtimes were at times shifted earlier or later, to promote progress in the reduction of sleep onset latency.

Rewards. While recommended for use with all three children, only 2/3 parents implemented a reward system with their child. Parents provided rewards in the form of verbal praise and/or a preferred tangible reward (for example; food, stickers, or an enjoyable activity), contingent upon their child's demonstration of target behaviours, such as staying in their own bed the whole evening. In both cases these rewards

were given in the morning and tangible rewards were provided by and at the cost of the parents.

Maintenance. During the maintenance phase, the parents and researchers had no contact. This break in communication was necessary for families to fully embed new sleep-conducive behaviours into their everyday lives and allow for progress to generalize outside of the context of treatment (Blampied, 2013; Sanders & Burke, 2014). Parents completed post-treatment psychometrics and a post-treatment interview during this phase.

Short-term follow-up. At roughly four weeks post-treatment, families were asked to record seven nights of video footage and sleep diaries. The data gathered at this stage provided insight into the retention and permanency of the original treatment effects.

Individual Cases

A summary of the participant's sleep problems as reported by their parents, the precipitating and/or maintaining factors identified through FBA, the hypothesized function of the sleep problems, and the selected interventions for each of the three children is presented in Table 2.

Hannah.

Family's goals for treatment. The parents' goals included: 1) for Hannah to take herself back to her own bed after waking to use the toilet and for her to remain there for the duration of the night; 2) for Hannah to settle herself back to sleep independently upon waking in the night; 3) for Hannah to learn to settle herself independently (i.e., without parental presence in the bedroom) during sleep onset; 4) for Hannah to wake after a more appropriate time of 6am in the morning.

FBA outcomes. Hannah's FBA indicated that multiple factors may be responsible for the maintenance of her sleep difficulties. The functions behind Hannah's difficulty self-settling following night wakings and subsequent co-sleeping were established as being a desire for parental attention, in the form of conversation, as well as a desire for physical contact. The enabling of co-sleeping by Hannah's parents provided such positive reinforcement and henceforth cemented these sleep problems. Hannah's early morning wakings and disruptive behaviour in the morning were found to be similarly maintained by the social reinforcement of parental attention. Additionally, having access to preferred items (e.g., toys in the lounge) and the opportunity to roam the house and property unsupervised were identified as functions of these undesired sleep-related behaviours.

Method.

Baseline (BL). Hannah's family carried out baseline phase with a randomly assigned duration of one week.

Intervention. The treatment plan designed for Hannah included faded bedtime, sleep/wake scheduling, elimination of daytime naps, discriminative stimulus for sleep/wake time, a social story, removal of parental presence, and reinforcement. Any variation on the common intervention components described on pages 77 to 79 is outlined below.

Faded bedtime. Hannah's bedtime was shifted from 7.30 to 8.30pm. In this case, even though Hannah's sleep onset latency was not deemed problematic at assessment, it was hypothesized that increase in the biological need for sleep may result in a reduction in the frequency of night wakings and early morning waking, which were indicated as being problematic.

Sleep/wake scheduling. Hannah's parents were asked to ensure that Hannah was awake for the day by 6.30am.

Reward system. Hannah was provided with a tangible reward, paired with praise and social attention, contingent upon her compliance with intervention, namely staying in her own bed for the entire evening.

No procedural modifications to the treatment plan in Hannah's case.

Robert.

Family's goals for treatment. Robert's parents' goals for intervention were to: 1) fall asleep independently in his own bed, without exhibiting signs of distress, and remain there for the whole night, and 2) resettle himself independently following a night waking.

FBA outcomes. Results from the FBA indicated that Robert's sleep onset delay and difficulty independently initiating sleep, inability to resettle himself following night wakings, and co-sleeping were likely maintained by social reinforcement. It is clear that Robert was seeking out parental attention, given that he would follow them around during the night upon waking. In receiving this desired attention from his parents, his behaviour was henceforth reinforced. With respect to Robert's early morning wakings, his FBA revealed that access to preferred activities and tangible items, such as television and toys, was a maintaining factor.

Method.

Baseline (BL). Robert was assigned a two-week baseline period.

Intervention. Interventions strategies implemented included a faded bedtime, consistent sleep/wake scheduling, discriminative stimulus for sleep/wake time, a social story, and removal of parental presence.

Faded bedtime and consistent morning wake-time. Robert's bedtime was changed from 7.00pm until 8.30pm. His parents were also asked to enforce a consistent wake time of 6.30am each day.

Discriminative stimulus for sleep/wake time. Robert's parents were asked to turn the Groclock on when Robert was put to bed at 8.30pm (initially), and set to display the sun at 6.30am when he was able to get up for the day.

No procedural modifications were made to Robert's treatment plan.

Michael.

Family's goals for treatment. The family's goals for treatment were 1) for Michael to remain in his own bed for the entire night and subsequently eliminate co-sleeping; 2) for Michael to be able to independently resume sleep following night wakings; 3) to reduce sleep onset duration; 4) increase compliance with the bedtime routine; and 5) eliminate curtain calls.

FBA outcomes. The results of Michael's FBA suggested that his sleep problems were being maintained by multiple potential factors. The positive reinforcement that Michael gained from his maternal attention and physical contact through co-sleeping was a possible factor that was strengthening his undesired curtain calls, night wakings, and inability to self-settle. In summation, Michael's sleep problems were determined as performing the functions of social attention and to obtain sensory stimulation.

Method.

Baseline (BL). Michael was randomly assigned a three-week baseline phase.

Intervention. Treatment recommendations included a social story, discriminative stimulus for sleep/wake time, extinction, elimination of daytime naps, and a reward system.

Faded bedtime. Michael's bedtime was delayed from 8.30pm to 9.00pm.

Discriminative stimulus for sleep/wake time. The visual aid displayed a photograph of his clock with hands showing 7 o'clock in the morning. There is also text underneath the photograph saying 'Morning Time' along with a cartoon image of a sun. The visual aid was situated on the wall of his bedroom and was used to model the time of day that Michael was able to get up and go and see his mother.

Extinction. If Michael left his bed after being bid goodnight, his mother was instructed to return him to bed, say "goodnight," and then leave the room. She was to do this with minimal communication and in a calm manner. His mother also reminded him that he was to remain in his bed until his clock matched his visual aid (7:00am) or when she comes to wake him in the morning. In cases where Michael's mother was concerned for his health or safety, she was to check on him with as minimal attention as was necessary.

Reward system/reinforcement procedure. A reward system was put in place where Michael received tokens for falling asleep in his bed independently and for staying in his bed all night. These were later exchanged for tangible rewards.

Procedural modifications. On night 58 of intervention, a further delay to Michael's bedtime was instituted in an attempt to further reduce his sleep onset latency. His bedtime was delayed until 9.15pm to 9.30pm.

Table 2. Parent reported sleep problem/s, precipitating and/or maintaining factors, hypothesized function, and intervention for each of the three children

Child	Parent reported sleep problem/s	Precipitating and/or maintaining factors	Hypothesized function	Intervention
Hannah	Night wakings, co-sleeping, early morning waking, day-time napping	Parental attention, preferred activities (i.e., exploring house) and tangible items (i.e., toys)	Attention, escape from bed	Social story, Groclock, faded bedtime, sleep/wake scheduling, removal of parental presence, reward system, elimination of daytime naps
Michael	Bedtime resistance, sleep onset delay, night wakings, co-sleeping, day-time napping	Parental attention, physical contact	Attention, sensory stimulation	Social story, visual aid, faded bedtime, extinction, rewards system, elimination of daytime naps
Robert	Sleep onset delay, night wakings, co-sleeping, early morning waking	Parental attention, preferred activities (i.e., television) and tangible items (i.e., toys)	Attention, escape from bed	Social story, Groclock, faded bedtime, sleep/wake scheduling, removal of parental presence

Chapter 5

Results

Chapter 5 presents data on sleep outcomes following sleep intervention for each of the three participants. Data relating to parental sleep quality and well-being, children's daytime behaviour and communication at pre- and post-intervention is also presented. Finally, treatment acceptability data gathered post-intervention is also reported.

Although she completed intervention, follow-up data for Hannah had not been collected at the time of submission of the current study. Data from Hannah's baseline and intervention phases alone, is presented in the results.

Sleep Outcome Measures

Sleep diaries. Each child's sleep intervention varied in length, with the specific duration dependent on the rate of the individual child's progress and/or any obstacles or setbacks that may have delayed goal attainment. Michael's intervention was suspended on 18 out of 33 nights due to illness. Sleep diary data concerning at least one sleep variable is missing for days 38, 48, 49, 50, 51, and 62 as his mother did not report all data on these days. Intervention was implemented for a total of 54 days for Michael before the decision was made to end treatment. It was at this point that his mother determined that she was satisfied that her treatment goals had been achieved and that she no longer required the involvement of the research team.

For Robert, co-sleeping data is not reported in the sleep diaries from nights 21-30 during intervention and as such, this data is not included in the results. Data is reported for his 24 nights of intervention.

Sleep diary data was not reported for one night of Hannah's intervention, and is therefore not included in any analyses. Duration of night waking data is missing on

nights 1, 2, 6, and 7 during baseline, as these times were not reported in the corresponding sleep diaries. Data is also missing for various sleep variables on nights 9, 10, 11, 12, 13, 14, 20, 22, and 24 during intervention.

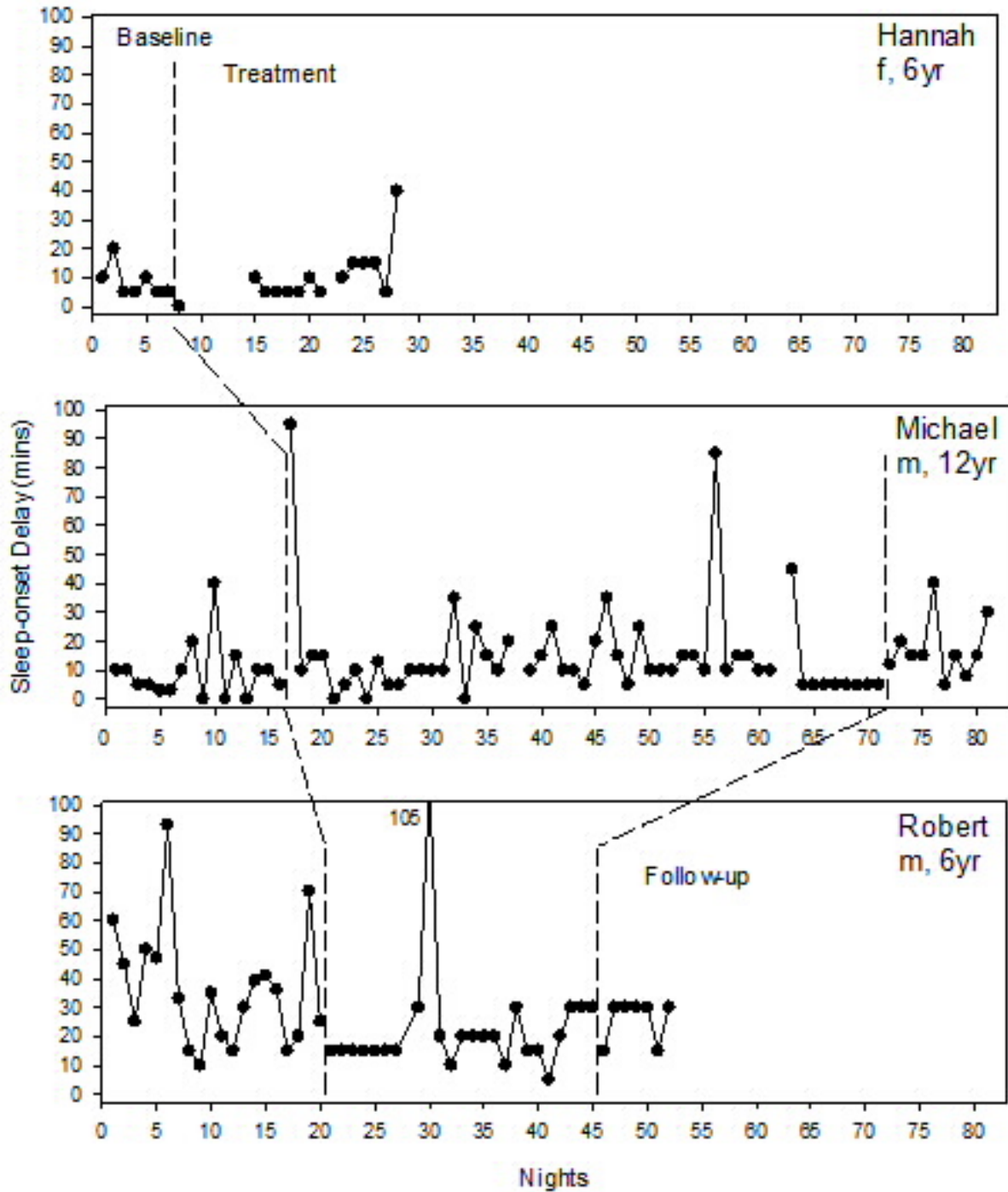


Figure 1. Duration of sleep onset latency (mins) during baseline and intervention for the three participants, and follow-up data for Michael and Robert.

Effect on sleep onset latency. Figure 1 displays baseline, intervention, and (where available) follow-up data for sleep-onset latency (SOL) for all three participants. Considerable variability in SOL during baseline was observed for Michael and Robert. Hannah on the other hand presented with a comparatively less variable SOL over baseline. A variable pattern of SOL continued throughout intervention for Michael, while Robert's SOL appeared to have consolidated and become less variable. Some of the data pertaining to Hannah's SOL during intervention is missing, however the available data indicates that there was no substantial change from baseline patterns. The follow-up sleep diary data available for Michael reveals a return to his variable pretreatment SOL, and within the same temporal range reported at baseline. Robert's reduction in SOL was maintained at follow-up, and there was a decrease in the amount of variability when compared to intervention. Follow-up data had not been collected from Hannah's parents at the time of submission of this thesis.

Hannah. For Hannah, a variable pattern of SOL was reported during her baseline period. SOL ranged from 5 to 20 minutes over the course of this study phase. On the first night of intervention, Hannah's SOL was reported to be within 1 minute, and then SOL data was not reported for the next 7 nights. From night 16 to the end of Hannah's intervention phase (night 29), her SOL ranged from 5 to 15 minutes, with the exception of night 23, when her SOL was not reported, and night 29, when her SOL increased to 40 minutes. The reason for this was unexplained.

Michael. For Michael, his SOL was variable during the baseline phase, ranging from 3 to 40 minutes. His longest SOL periods during this phase aligned with a bout of illness he experienced from night 8 to night 16. A sharp and immediate escalation in SOL (95 minutes) occurred on night 17 when Michael's intervention phase began. This spike is likely to be the result of the removal of parental presence

as part of intervention. Following this initial burst, there was no change from baseline levels up until with the exception of an 85-minute SOL on night 56 and a 45-minute SOL that occurred on night 63. On both occasions Michael had been experiencing stomach pains. Following this last burst, there appeared to have been a resolution of SOL for the remainder of intervention, however there was a return to baseline levels over follow-up.

Robert. For Robert, his baseline SOL was variable (between 10 and 93 minutes) and typically lasted longer than 30 minutes. From the commencement of intervention, his SOL became more consistent, settling within 15 minutes for the first 7 days of intervention. On night 29, when Robert was staying at his grandparents' house, he exhibited a spike in SOL to 105 minutes. For the remainder of his intervention phase (from night 30 to night 42), SOL ranged from 5 to 30 minutes. During follow-up, SOL was relatively consistent compared to baseline and intervention. During 5/7 nights of follow-up his SOL was 30 minutes and on the other two nights his SOL was 15 minutes.

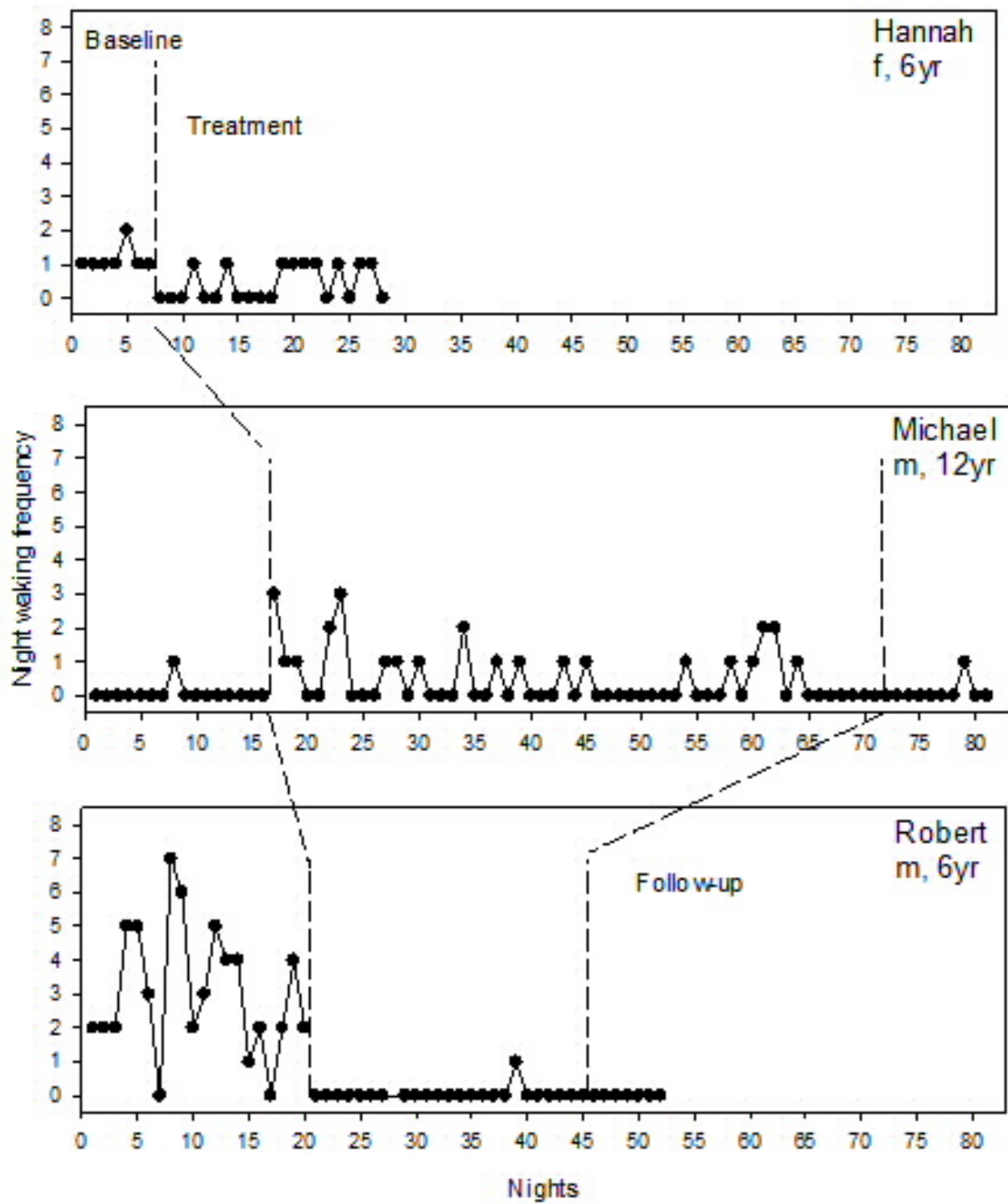


Figure 2. Frequency of night wakings during baseline and intervention for the three participants, and follow-up for Michael and Robert.

Effect on frequency of night wakings. Figure 2 presents data for the frequency of night wakings (NW's) for each of the three children in the study over the course of their baseline, intervention, and follow-up phases. During baseline, the frequency of NW's was variable for Robert, almost non-existent for Michael, and consistent but low for Hannah. Throughout intervention, the frequency of NW's increased for Michael, and decreased for Hannah and Robert. At follow-up, NW's were non-existent for Robert, and almost non-existent for Michael.

Hannah. During her seven-day baseline period, Hannah displayed a relatively consistent pattern of NW's, having one NW on six nights and two NW's on one night. Following intervention, the frequency of NW's decreased, specifically with nine nights of one single NW, and 12 nights with no NW's.

Michael. Contrary to pre-baseline parental report, Michael displayed a very low frequency of NW's during baseline, with zero NW's barring one that occurred on night 8. The reason for this is likely related to his sleep setting over those particular evenings and the subsequent fulfilment of his co-sleeping needs from initial sleep onset. During intervention, NW's occurred during 19 out of 54 nights and ranged from one to three NW's per night. The nights during which NW's took place were typically followed by an absence of NW's for a period lasting between one to eight consecutive nights. One period of consecutive nights without NW's spanned from night 29 to night 53, and then again during the last seven nights of intervention. Michael's lowered frequency of NW's was sustained at follow-up, during which time he had one NW.

Robert. For Robert, NW's occurred on all but two nights during the baseline phase. The frequency of NW's over those evenings was highly variable and ranged from one to seven. From the commencement of intervention, NW's were virtually

non-existent, with the exception of one NW on night 38. During the follow-up phase no NW's were reported.

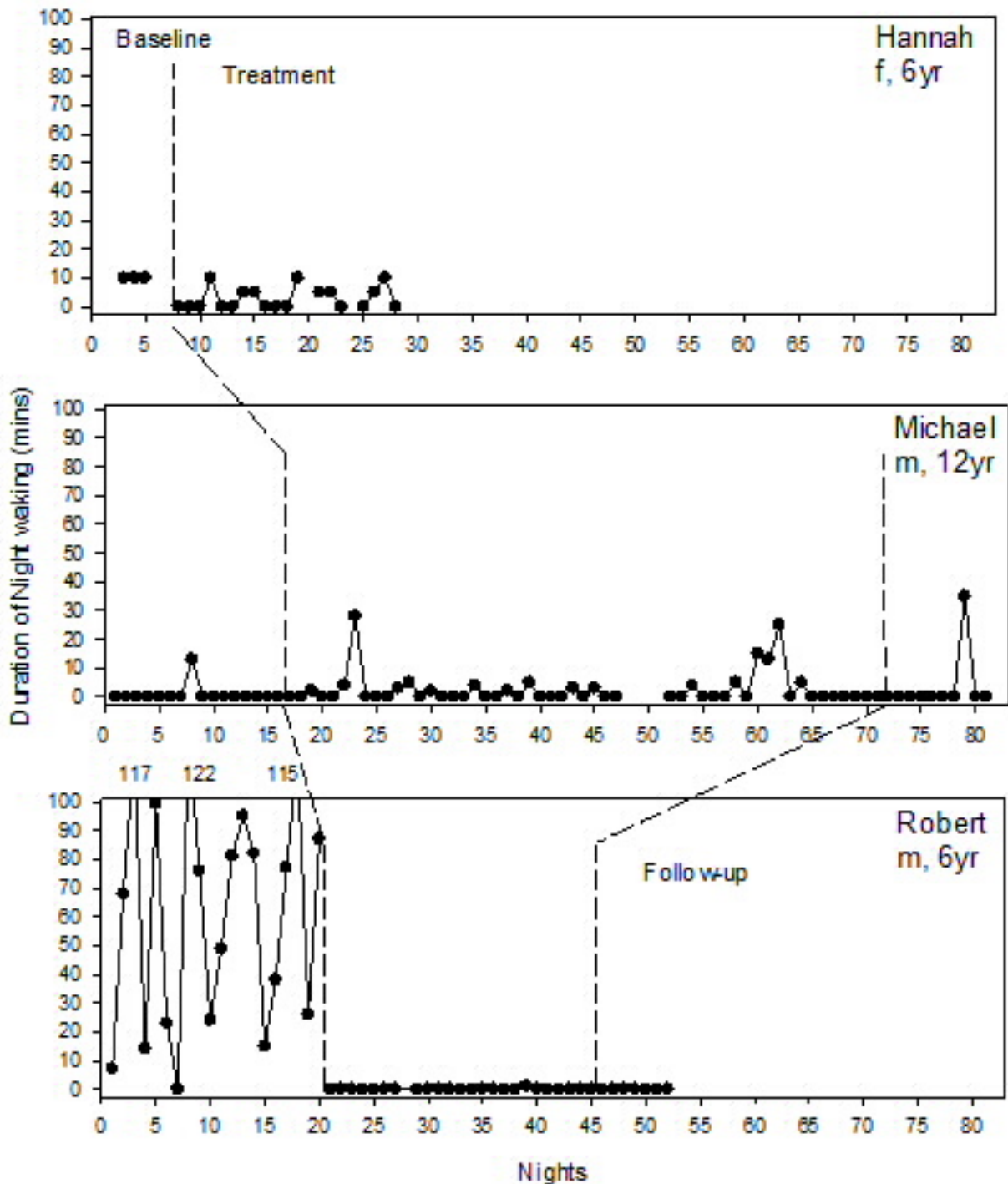


Figure 3. Duration of night wakings during baseline and intervention for the three participants, and follow-up for for Michael and Robert.

Effect on duration of night wakings. Data pertaining to the duration of NW's for the three children in this study across baseline, intervention, and follow-up are depicted in Figure 3. The duration of NW's during baseline was highly variable for Robert, and less variable for Hannah. Michael's single NW during baseline did not reveal a pre-treatment pattern of NW duration. Throughout intervention, duration of NW's did not fluctuate significantly for Michael or Hannah. An almost entire elimination of NW's was reported from the introduction of intervention and through to follow-up for Robert, and during follow-up for Michael.

Hannah. Over the three consecutive nights of her baseline phase when night wakings were reported, the night wakings lasted for 10 minutes. Although NW's were reported as having occurred on nine nights of Hannah's intervention, data regarding the duration of NW's was not reported for three of those nights. For those reported, the duration ranged from 5 to 10 minutes, indicating low variability.

Michael. Michael's baseline period had one NW that lasted 13 minutes, however the single NW did not enable the identification of a pattern of NW duration and was at variance with the parent's previous report. The explanation for this is alluded to in the previous section regarding the frequency of NWs. The duration of his NW's was under 5 minutes on 13 of the 17 nights of intervention during which at least one NW occurred. During the other five nights of intervention which featured at least one NW, Michael's NW's lasted between 13 to 28 minutes. With Michael's NW's not exceeding 30 minutes in length on any occasion over the course of baseline or intervention, a relatively consistent pattern of a low duration was observed. Michael's single NW during his follow-up phase lasted for 35 minutes, which is a very slight increase from the range of durations that he exhibited previously. However, as with his baseline, the one NW during follow-up is not enough data to establish a pattern of NW duration following treatment.

Robert. For Robert, his duration of NW's was highly variable, lasting from 7 to 122 minutes. Robert's reduction in the frequency of NW's during intervention was paralleled by a reduction in NW duration. NW's did not occur during follow-up; therefore, no data was reported concerning the duration of NW's.

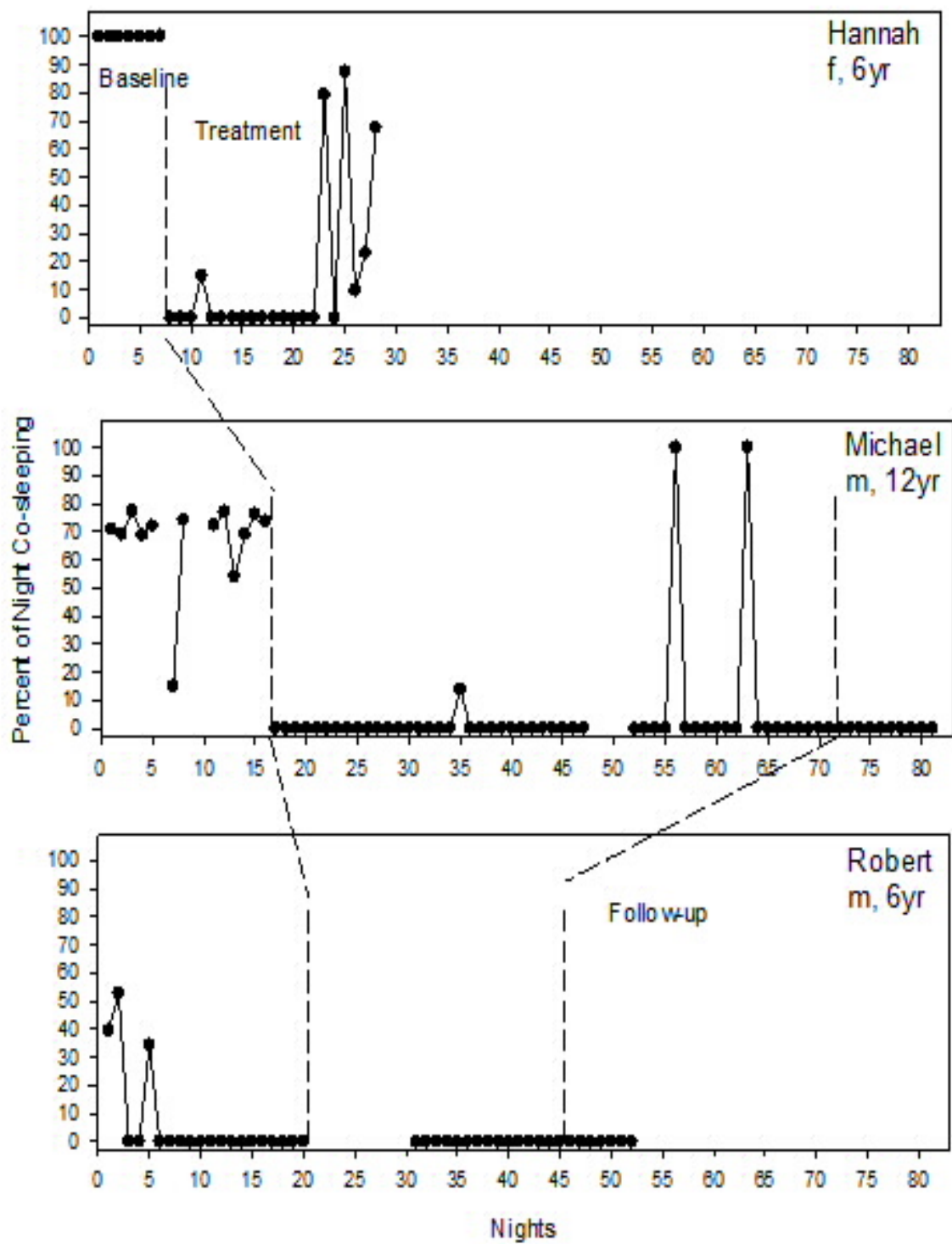


Figure 4. The percentage of the night spent co-sleeping during baseline and intervention for the three participants, and follow-up for Michael and Robert.

Effect on co-sleeping. Figure 4 displays the baseline, intervention, and follow-up data for the percentage of the nightly sleep period spent co-sleeping for the three children participating in the study. Michael and Hannah engaged in co-sleeping every night during the baseline period, and for high percentages of their total nightly sleep period. Co-sleeping occurred in Robert's case over the first few nights of baseline, but from then and through to the end of his follow-up, it was no longer a presenting problem. Michael and Hannah's co-sleeping also decreased to 0% with the introduction of intervention, and was sustained at that percentage for most of their intervention phases, with the exception of a few evenings. For the entirety of Michael and Robert's follow-up phases, co-sleeping sat at 0%.

Hannah. Hannah co-slept for 100% of her total nightly sleep period during each night of baseline. Her co-sleeping fell to 0% for almost every night of intervention, until night 24. Then for five of the remaining six nights of intervention, her percentage of total nightly sleep period spent co-sleeping rose and ranged from 9.7% to 87.5%. This is probably a consequence of lapse in treatment adherence on the part of the parents at this time. As follow-up data is yet unavailable, it is not possible to determine whether the parents reinstated any procedures pertaining to the elimination of parental presence/co-sleeping at any point after the cessation of Hannah's intervention phase.

Michael. Co-sleeping occurred every night during Michael's baseline and for a large percentage of those nights. Co-sleeping constituted between 54% and 77% of his total nightly sleep period, with the exception of night 7, when 14.9% of the night was spent co-sleeping. The percentage of the night that was spent co-sleeping reduced to 0% for the first 18 nights of intervention until night 35, when Michael co-slept with his Mother 13.9% of his nightly sleep period. His co-sleeping reduced back down to 0% for the remainder of his intervention phase, with the exception of two nights (night 56 and night 63) during which he co-slept with his Mother 100% of his

total nightly sleep period. The reduction in co-sleeping that was observed over intervention was maintained at follow-up for Michael, during which time his co-sleeping sat at 0% for all 10 nights of follow-up.

Robert. Within the first five days of Robert's baseline, there were three nights during which he co-slept between 34% and 52% of his total nightly sleep period. Co-sleeping then displayed a descending trend from night six of his baseline phase, and remained at 0% for every night of intervention and follow-up.

Sleep Problem Severity. The SPS scores for each of the three participants at each of the three study phases is presented in Table 3. Higher SPS scores indicate higher prevalence of co-sleeping, sleep onset delay longer than 15 minutes, and night wakings. The trend in the scores over the three phases of the study was variable across participants.

Table 3. Comparison of baseline, treatment, and short-term follow-up SPS scores for the three participants

	Hannah	Michael	Robert
Baseline	1	4	7
Treatment	4	1	1
Short-term follow-up	-	0	2

Hannah. Hannah's low pre-intervention SPS score of 1 increased following intervention to 4. Short-term follow-up data was not yet collected for Hannah at the time of submission.

Michael. Michael's SPS scores show a steady reduction in sleep problem severity over the three stages of this study. His baseline score of 4 decreases to 1

following the commencement of intervention, and then decreases slightly again by his short-term follow-up.

Robert. For Robert, his baseline score of 7 was the highest of the sleep problem severity scores across the three participants. His score then fell significantly to 1 at the time of treatment, and then saw a small increase to 2 at his short-term follow-up.

Child Sleep Habits Questionnaire. The results of the CSHQ (Owens et al., 2000) are displayed in Table 4. Higher scores convey greater difficulties in the particular sleep-related variable rated. Total sleep difficulty ratings across the three participants at baseline were mixed. At post-intervention, total sleep difficulties decreased for Michael and Robert, while slight increase was reported for Hannah.

Table 4. Comparison of Pre- and Post-Intervention Scores on the CSHQ for the three participants

Variable scores	Hannah		Michael		Robert	
	Pre	Post	Pre	Post	Pre	Post
Bedtime Resistance	8	9	11	6	6	5
Sleep Onset Delay	1	1	2	1	2	1
Sleep Duration	4	5	7	3	3	3
Sleep Anxiety	6	7	7	4	4	4
Night Wakings	8	6	9	4	9	3
Parasomnias	10	11	10	8	10	10
Disordered Breathing	5	5	4	3	3	4
Daytime Sleepiness	12	11	11	11	10	11
Total difficulties	54	55	61	40	47	41

Hannah. There was considerable variability across Hannah's pre- to post-intervention scores, with the CSHQ variables showing either an increase, decrease, or constancy. His night wakings and daytime sleepiness scores decreased, from 8 to 6 and 12 to 11 respectively. A slight increase in bedtime resistance (from 8 to 9), sleep duration (from 4 to 5), sleep anxiety (from 6 to 7), and parasomnias (from 10 to 11) was seen over the two time points. The remaining variables of sleep onset delay (both: 1) and disordered breathing (both: 5) showed no change from pre- to post-intervention. Despite the above-described variability, Hannah's total difficulties score reflected an overall, albeit minor, increase in sleep disturbance.

Michael. For Michael, his post-intervention scores on all variables were lower than his pre-intervention scores, with the exception of daytime sleepiness, which remained at 11 at both time-points. The most sizeable decreases were in bedtime resistance (from 11 to 6), sleep duration (from 7 to 3), and night wakings (from 9 to 4). Overall, his total sleep difficulties score decreased significantly from 61 to 40 following treatment.

Robert. Robert's scores remained constant from pre-intervention to post-intervention or decreased on the CSHQ, except for daytime sleepiness, which increased from 10 to 11, and disordered breathing, which increased from 3 to 4. His scores for sleep duration (both: 3), sleep anxiety (both: 4), and parasomnias (both: 10) did not change following intervention. Improvements in bedtime resistance (from 6 to 5), sleep onset delay (from 2 to 1), and most notably, night wakings (from 9 to 3), were reported. His total sleep difficulties score reflects an overall decrease, from 47 at pre-intervention to 41 at post-intervention.

Parental Sleep, Well-being, and Relationship Satisfaction Measures

PSQI. The global score of the PSQI for the parents of all three children participating in the study is presented in Table 7. This gives an impression of the quality of the parents' sleep at pre- and post-intervention. Lower scores on the PSQI indicate better sleep quality. The results are mixed across the respondents.

Table 5. Comparison of Pre- and Post-Intervention Global scores on the PSQI for the parents of the three participants

	Hannah				Michael				Robert				Max
	Mother		Father		Mother		Father		Mother		Father		
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	
Global PSQI score	7	5	4	5	6	7	N/A	N/A	9	6	4	5	21

Hannah. Hannah's mother's global sleep quality score was 7/21 at baseline and decreased to 5/21 following treatment. Her father's global score increased slightly from 4 to 5/21.

Michael. Michael's mother's global sleep quality score increased from 6/21 to 7/21 following the completion of his intervention.

Robert. For Robert's mother, her global sleep quality score decreased from 9/21 to 6/21, while his father's global score showed a slight increase, from 4/21 to 5/21, over the two time-points.

DASS-21. The participants' parents' results for the DASS-21 are presented in Table 5. A greater likelihood of psychological distress in the form of each of the three dimensions is represented by higher scores. Across the participants' parents, anxiety scores either decreased or remained constant following intervention. Conversely,

there was more variability in depression and stress scores. For both variables, three of the five respondents scored higher at the second measurement time-point, while the remaining two respondents' scores decreased. All scores were within the normal range.

Table 6. Comparison of Pre- and Post-Intervention Scores on the DASS-21 for the three participants

Variable scores	Hannah				Michael				Robert			
	Mother		Father		Mother		Father		Mother		Father	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Depression	2	1	3	2	0	3	N/A	N/A	1	2	0	2
	(N)	(N)	(N)	(N)	(N)	(N)			(N)	(N)	(N)	(N)
Anxiety	12	1	2	0	2	0	N/A	N/A	0	0	0	0
	(N)	(N)	(N)	(N)	(N)	(N)			(N)	(N)	(N)	(N)
Stress	11	8	3	5	6	4	N/A	N/A	1	3	2	7
	(N)	(N)	(N)	(N)	(N)	(N)			(N)	(N)	(N)	(N)

Hannah. Hannah's parents both scored within the normal range at the two measurement time-points. Her mother produced lower scores at post-intervention than at pre-intervention for depression, anxiety, and stress. Her father's scores similarly decreased for depression and anxiety, however his stress score increased at post-intervention. The decrease in depression scores was slight, from 2 to 1 for her mother and 3 to 2 for her father. Her father's anxiety score decreased from 2 to 0, and her mother's anxiety score showed a marked decrease from 12 to 1. Hannah's mother's stress score decreased from 11 to 8, while her father's stress score increased from 3 to 5.

Michael. Michael's mother scored within the normal range for all dimensions at pre- and post-intervention. His mother's depression score increased from 0 to 3. For

the anxiety and stress dimensions, his mother scored two points lower at post-intervention compared to pre-intervention, from 2 to 0 and 6 to 4 respectively. No data was collected for Michael's father, as his Mother is his primary caregiver.

Robert. For all dimensions, both of Robert's parents scored within the normal range at pre- and post-intervention. His parents' depression and stress scores increased post-intervention. The increase was slight for depression scores, from 1 to 2 for his mother and 0 to 2 for his father. Robert's mother's score increase was also minor for stress (1 to 3), however his father's stress score increased considerably from 2 to 7. His parents both scored 0 at pre- and post-intervention for the anxiety dimension.

RQI. The results of the RQI are presented in Table 6, showing the pre- and post-intervention global scores for the parents of two of the children in the study (Robert and Hannah). Greater partner satisfaction is indicated by higher global RQI scores. Across the responding parents, scores either did not change from pre- to post-intervention, or increased. Michael's mother did not complete the questionnaire, as she is not in a relationship.

Table 7. Comparison of Pre- and Post-Intervention Scores on the RQI for parents of two of the participants

	Hannah				Robert				Max
	Mother		Father		Mother		Father		
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	
Global RQI	31	33	39	39	44	44	45	45	45

Hannah. A higher level of partner satisfaction was indicated by Hannah's father (39/45) at baseline than her mother, who rated her partner satisfaction at 31/45

before intervention commenced. Hannah's father's post-intervention global RQI score did not change from pre-intervention. Her mother on the other hand indicated a higher level of partner satisfaction post-treatment (33/45) compared to her pre-treatment rating.

Robert. Robert's mother and father indicated similar levels of partner satisfaction, with scores of 44/45 and 45/45 respectively, and at both measurement time-points.

General Child Behaviour Measures.

VABS-II. The results of the VABS-II at pre- and post-intervention for the three children participating in the study are displayed in Table 8. The receptive communication scores represent the proficiency with which an individual listens, pays attention, and understands, and the expressive communication scores represent the level of sophistication in the words and sentences they use in their communication. The reported scores indicate the neurotypical age, in years and months, that is equivalent to their ratings on various dimensions of the questionnaire. A higher score, which reflects an older age, signifies greater ability. For Michael and Robert, their age-equivalent scores on both the receptive and expressive communication dimensions either increased or did not change following treatment. Contrastively, Hannah's scores showed a decline in receptive and expressive communication.

Table 8. Comparison of Pre- and Post-intervention receptive and expressive communication age-equivalence scores on the VABS-II for the three participants

	Hannah		Michael		Robert	
	Pre	Post	Pre	Post	Pre	Post
Receptive communication	3-2	2-4	2-3	2-9	1-8	1-10

Expressive communication	4-10	3-11	1-10	1-11	1-11	1-11
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(Note: X-X = year-month)

Hannah. For Hannah, there appears to have been a significant regression in both her receptive and expressive age-equivalent communication scores from pre- to post-intervention. Her pre-intervention receptive score of three years and two months decreased to two years and four months at post-intervention. Likewise, Hannah's expressive score at baseline was four years and 10 months, which then decreased to three years and 11 months following intervention.

Michael. Receptive and expressive communication age-equivalent scores increased from pre- to post-intervention for Michael. His pre-intervention receptive score of two years and three months increased by six months following intervention, while his expressive score increased by one month, from one year and 10 months to one year and 11 months. This slower increase for expressive communication is likely related to some developmental factor, perhaps highlighting an especially challenging area of development for Michael.

Robert. Robert's age equivalent scores on the receptive and expressive communication domains remained fairly consistent over the two time-points. Robert's receptive score was one year and 8 months at baseline and then increased by two months following intervention. His expressive communication score of one year and 11 months was sustained from pre- to post-intervention.

CBCL. The pre- and post-intervention results of the CBCL for the three children are displayed in Table 9. The CBCL, which is completed by parents, brings to light possible problems of a behavioural or emotional nature in a child. A greater level of problem is indicated by a higher score.

Table 9. Comparison of Pre- and Post-Intervention Internalising, Externalising, and Total T-scores on the CBCL for the three participants

	Hannah				Michael				Robert			
	Pre		Post		Pre		Post		Pre		Post	
	Score	R	Score	R	Score	R	Score	R	Score	R	Score	R
Internalising behaviours	59	N	48	N	52	N	57	N	-	-	52	N
Externalising behaviours	73	C	62	B	57	N	55	N	-	-	51	N
Total Score	72	C	63	B	58	N	59	N	-	-	54	N

(Note: R= Range; N=Normal; B=Borderline; C=Clinical)

Hannah. Hannah's parents gave her scores that reflect a reduction in internalizing and externalizing behaviours following intervention and a shift in score ranges. For internalizing behaviours, her score remained within the normal range, but decreased from 59 at pre-intervention to 48 at post-intervention. With regard to externalizing behaviours, Hannah shifted from the clinical range to the borderline range with her scores of 73 and 62.

Michael. For Michael, his mother's ratings did not change significantly from pre- to post-intervention. His mother gave him a rating of 61 for internalizing behaviours at baseline, which placed him in the normal range. Michael remained within the normal range for internalizing behaviours, with a score of 57 following intervention. Similarly, for externalizing behaviours, Michael fell within the normal range at pre- and post-intervention, with scores of 57 and 55 respectively.

Robert. Pre-intervention CBCL data was not collected for Robert, however his parents scored him in the normal range for internalizing and externalizing behaviours at post-treatment.

Treatment Acceptability

Post-treatment discussions.

Hannah. Hannah's mother stated that they were satisfied with the intervention process and the treatment procedures suggested, especially the Sleep Story, Groclock, and restriction of daytime napping, saying that they were "easy to implement and effective". Of the Sleep Story, she said that she felt that it was particularly impactful for Hannah, and that Hannah "enjoyed that it was about her" and that it "told her really clearly what [they] wanted her to do". Her mother reported that Hannah had become very familiar with her bedtime routine following the consistent reading of the Sleep Story over the course of intervention. Some disruption that Hannah's mother felt that the study caused involved the amount of psychometrics that they had been asked to complete at pre- and post-intervention. Additionally, she felt unsure as to "whether the [psychometric] data gathered was meaningful" and necessary. Hannah's mother also reported that the camera equipment stopped working during treatment and that the red light on the equipment disrupted her own and Hannah's sister's sleep, when they co-slept together in Hannah's room. According to her mother, Hannah's only persisting sleep problem following treatment involved toileting difficulties, wherein Hannah was still wearing a nappy during the night, and sometimes wet the bed. She also stated that Hannah's behaviour at school had improved in response to intervention, specifically that she is "more attentive to learning" and she is able to "concentrate easier". Hannah's mother noted that her daughter's improved sleep as a result of intervention had positively impacted herself and Hannah's younger sister, with both family members consequently having experienced improved sleep.

Michael. Michael's mother described the intervention as good overall, and that having a treatment formulated by someone with a fresh perspective brought to light the "common sense" solutions to which she had become "oblivious". She noted that

the bedtime routine, goal setting aspects of treatment were helpful in achieving the desired sleep outcomes, but the social story was particularly effective. She reported that Michael was very engaged with the social story and would enjoy reading it multiple times each night. Michael's interest in the social story lessened over the course of the intervention as he was eventually able to grasp the routine and expectations that it set out. Michael's mother expressed that the intervention could be improved through better communication regarding deadlines for the completion of psychometrics and the organization of intervention resources such as rewards. She suggested that a flow chart could be provided that clearly stipulates the entire intervention process including such deadlines, to give families the opportunity to prepare and consequently prevent any delays in the intervention process. She also noted that she would have liked to have seen some graphs or visual representation of Michael's progress during each of the study phases, along with a written summary at the end of their involvement in the study. Michael's mother noticed that Michael's mood was better following the good night's sleep that he was now able to enjoy following treatment. She stated that they were "still working on things but there has been a huge change" with respect to Adam's ability to sleep through the night, as well as her own sleep. However, she reported that she still struggled to relax once in bed herself, as she is still "alert to any sounds" and anticipates Michael waking.

Robert. The intervention and overall study process was "great" according to Robert's mother. She noted that despite the simplicity of Robert's intervention, positive change in his sleep-related behaviour was achieved. His mother reportedly believed that the faded bedtime procedure was particularly effective in reducing his SOL, and that Robert had become rather fond of the Groclock and enjoys switching it on at night. Robert apparently understands what the sun and the star displays on the Groclock signify, and is able to communicate to other what they mean with respect to his sleep and wake times. She also felt that the mere involvement of the research team and Robert's awareness of being monitored by the camera may have

prompted the immediate reduction seen in his bedtime resistance. Robert's mother said that she had found the amount of psychometrics and paperwork overwhelming, and did not understand their purpose. She suggested that it might be helpful for parents to be given a better explanation for the reasoning behind the administration of the questionnaires and also to be provided with some information regarding the changes seen in the various measured variables from pre- to post-intervention. Robert's mother recognized that sleep had improved for everyone in the family following Robert's intervention, and in addition stated that she and Robert's father were no longer worried about Robert staying in his own bed throughout the night.

TARF-R. The results of the TARF-R are presented in Table 10. Overall, the three families that completed the intervention regarded the treatment to be highly acceptable, reasonable, comprehensible, affordable, and effective. There was some variability in the parents' perceptions of the disruption caused by the treatment and the time it took to carry out intervention.

Table 10. Post-Intervention Treatment Acceptability Scores from TARF-R for the three participants

Variable scores	Hannah		Michael		Robert		Max score
	Mother	Father	Mother	Father	Mother	Father	
Total Acceptability	93.5	93.5	99	N/A	110	110	121
Reasonableness	18.5	18.5	18	N/A	21	21	21
Willingness	15	15	21	N/A	21	21	21
Effectiveness	17.5	17.5	17	N/A	16	16	21
Cost	13	13	14	N/A	14	N/A	14
Negative Side-Effects	14	14	18	N/A	18	18	21
Disruption/Time	15.5	15.5	11	N/A	20	20	21
Problem Severity °	8	8	8	N/A	10	10	14

Understanding of Treatment °	6	6	6	N/A	5	5	7
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° = not included in the Total Acceptability score

Hannah. Hannah's mother and father were completely consistent in their ratings on all dimensions of the TARF-R, beginning with a high rating of overall treatment acceptability (both: 93.5/110). Her parents found the intervention to be reasonable (both: 18.5), effective (both: 17.5/21), and cost-effective (both: 13/14). Hannah's parents reported that while they have a high level of understanding of the intervention (both:6/7), their willingness to implement the intervention was at a moderate level (both: 15/21). They perceived the Hannah's intervention as moderately disruptive and time-consuming (both: 15/21) and bringing with it some negative side-effects (14/21). Hannah's parents considered her sleep behaviours post-intervention to be moderately problematic compared to her peers of the same age (both: 8/14).

Michael. Michael's mother reported a relatively high level of overall treatment acceptability (99/121). She also considered the intervention to be reasonable (18/21) and effective (17/21). His mother rated her willingness to implement the intervention very highly, with a maximum score of 21/21, and she reported a high level of understanding of treatment (6/7). Michael's mother also perceived the intervention to be very economical (14/14). She reported there to have been some negative side-effects (18/21) of treatment, and a high-level of disruption, with a rating of 11/21. Michael's mother perceived his sleep behaviour problems as moderate compared to his same-aged peers following intervention (8/14).

Robert. For Robert, his parents were in complete agreement in their ratings on the TARF-R. Both his mother and father showed a high-level of treatment acceptability overall (both: 110/121) and perceived the treatment to be very

reasonable (both: 21/21). Results suggest that both parents were very willing to carry out the intervention (both: 21/21) and had a moderate understanding of treatment (both: 5/7). They rated the effectiveness of the intervention a 16/21. Robert's parents perceived the intervention to have been very affordable (both: 14/14), and caused a low-level of disruption and did not require a lot of time to implement (both: 20/21). They rated the overall effectiveness of the intervention a 16/21. They also reported that intervention produced some negative side-effects (both: 18/21). Following intervention, Robert's mother and father deemed his sleep behaviour problems to be moderate in comparison to his same aged peers (both: 10/14).

Chapter 6

Discussion

This study provides some encouraging evidence for the effectiveness of FBA-based behavioural interventions in the treatment of sleep difficulties of children with RGND. Each of the participants exhibited improvements for most sleep-related variables following intervention, illustrating the effectiveness of the selected sleep interventions within this cohort. Consistent improvement across neither parental wellbeing measures, nor measures of general child behaviour, was observed as a result of treatment. Despite this, parents of all three participants proved to be highly satisfied with the intervention process and intervention itself.

Research Questions

Three research aims guided the current study: 1) to investigate the effectiveness of FBA-based interventions in the treatment of sleep disturbance amongst children with RGND; 2) to explore any changes to general child behaviour and parent wellbeing following the treatment of sleep disturbance; and 3) to examine parents' ratings of treatment acceptability and their experiences regarding the assessment and treatment process.

Study findings

The sleep diary data revealed an overall sustained improvement across dependent sleep-related variables from pre- to post-intervention for the children in this study. There was no change in SOL found for Hannah following intervention. SOL remained variable throughout all phases for Michael, but there was a clear PERB at beginning of intervention, and Robert progressed from highly variable SOL at baseline to a more consistent pattern. A consistently low frequency of NW's was

observed in Hannah's case over all phases, while still displaying an overall decrease with intervention, whereas Michael's low frequency of NW's at baseline also demonstrated a PERB with the introduction of intervention, followed by a return to baseline levels at follow-up. NW's appeared to have been eliminated from the beginning of intervention, and the effect was maintained at follow-up. Co-sleeping was eliminated for both Hannah and Michael as intervention commenced, however Hannah showed some deterioration nearing the end of treatment.

The results of the parent-reported CSHQ and SPSS echoed these findings regarding the participants' sleep following treatment for the most part, with Michael and Robert being reported to have shown reductions in total sleep difficulties and sleep problem severity respectively, while Hannah demonstrated a slight increase.

The utilization of FBA could be credited with the progress made by the three children, given that the assessment protocol allowed for the formulation of individualized interventions which targeted the specific sleep problems being presented by each of the participants, addressed the precipitating factors and function of the behaviour in question, and took into consideration the child's and their family's needs, preferences, and goals. In the few instances where the children exhibited a behavioural regression following the introduction of intervention, or no change, it may be that extraneous factors interfered with the intervention process (e.g., reduction in parental motivation to implement intervention; changes to sleep or home environment such as sleep-overs or visitors staying over) or the child's capacity to respond positively and optimally to treatment (e.g., illness; developmental changes).

Outliers, anomalies, and interesting observations.

Hannah. Despite a large immediate treatment effect, Hannah's progress with regard to co-sleeping deteriorated towards the end of treatment. The information

offered in her sleep diaries indicate that the parents' adherence to treatment was low, which could explain the decline. The parent-reported TARF-R willingness scores support this explanation as they imply that Hannah's parents were not highly motivated to implement the recommended intervention procedures, at least at the time that the TARF-R was completed by parents, upon completion of the intervention phase.

Hannah had a low and decreasing SOL in baseline, however there was no evidence of systematic improvement in treatment. The descending trend in SOL before the introduction of treatment may suggest that this problem was improving naturally and without intervention. The lack of progress observed in response to intervention may simply indicate that by that point her SOL had improved independently to an appropriate duration for Hannah, that being between five to 15 minutes.

There was an increase in Hannah's SPS and CSHQ scores following treatment, which was echoed in her sleep diary data regarding co-sleeping. As has been previously discussed and in fact self-reported, Hannah's parents were low in motivation by the end of intervention. No specific explanation for this was provided, however there are numerous possible reasons for anyone to lose motivation, and it is especially understandable given the intensity of the intervention and the challenges that present when attempting to change a child's behaviour (e.g., PERBs, lack of parental sleep, stress over progress).

Robert. Following his unstable and variable SOL during baseline, Robert's SOL became much more stable from the beginning of intervention, with the exception of one spike on night 30. On this night Robert had a SOL of 105 minutes. The only explanation that could be drawn from his sleep diaries to account for this anomaly is the fact that he spent that night at his grandparents' house. After this instance, his SOL returns to a more stable, but steadily increasing trend toward the end of

intervention. It is plausible then to posit that the change in Robert's environment (sleep conducive discriminative stimuli), and the circumstances and people around which he was to fall asleep disrupted his ability to fall asleep. This behavioural response to inconsistency in his routine and surroundings demonstrates the importance of upholding sameness when it comes to sleep interventions and seeing desired behavioural change.

Similar to Hannah's case concerning SOL, the descending pattern seen in Robert's co-sleeping during baseline may signify organic improvement in independent sleep initiation without parental presence. This could have resulted from developmental factors, or possibly a change in the parents' method or ability to manage Robert's co-sleeping.

Michael. Michael's data concerning NW's during baseline did not reflect earlier descriptions of his sleep disturbance given by his mother. While he had been reported to have consistently woken during the night when he fell asleep initially in his own bed, at which time he could join his mother to co-sleep with her in her bed, he only had one brief NW during his entire baseline phase. This discordance is likely resulting from the fact that for the majority of nights during baseline, Michael began his night sleep in his mother's bed, and therefore did not need to wake and instigate co-sleeping, as he was already in the presence of his mother.

After a short period of eliminated NW's around the mid-point of intervention, Michael's progress began to deteriorate from night 56 until night 64, after which time he had no NW's for the remainder of his treatment phase, and for almost his entire short-term follow-up. The reason for this 10-day long period of increased NW's appear to be related to the sleep resistance that was reported in his sleep diaries over that time. It was noted in the sleep diaries that Michael's grandfather was the one responding to his wakings during these nights, which could suggest that the

presence of someone other than his mother at night may have unsettled him somewhat and provoked him to resist his typical nighttime routine. If this is the case, then it emphasizes Michael's need for consistency in routines to support positive sleep behaviour.

While co-sleeping was effectively eliminated for Michael following the introduction of intervention, he co-slept for 100% of the night with his mother on nights 56 and 63, creating two spikes in his data-set toward the end of intervention. The reason for these outliers is easily explained by the fact that Michael and his mother were staying at his grandfather's house on these instances and had to share a bed.

Research Question One

Not only do the results of this study lend some support for the effectiveness of FBA-based behavioural interventions for sleep problems, they further the research through the application of such approaches with children with RGND.

Through FBA, NW's were identified as being a feature of all three children's sleep disturbance prior to treatment, although this was not reflected in Michael's sleep diary data for his 16-day long baseline period immediately preceding intervention, which was largely free of NW's. Subsequently, a combination of procedures including a social story, discriminative stimulus for sleep/wake time, extinction, and a reward system was devised to resolve the children's NWs, on the grounds that the function of this behaviour was found to be to procure parental attention, as well as the sensory stimulation of physical contact for Michael. This was also determined to be the function behind the pre-treatment co-sleeping in which both Michael and Hannah had been engaging, henceforth these intervention procedures were intended to treat this problem also. A treatment effect for the

frequency of NW's was demonstrated early in treatment for Hannah and Robert. Robert's decreased frequency in NW's from high variable numbers at baseline to 0 upon commencing with treatment was maintained at follow-up, whereas Hannah returned to her baseline frequency by the end of intervention, from night 19. Night wakings did not occur for Michael until a clear response to the introduction of intervention, when his frequency of NW's increased from 0 to 3. Following considerable variation during intervention, Michael's NW's returned to his baseline frequencies at follow-up. The duration of NW's treatment effects mirrors the pattern observed in the frequency of NW's across the participants. With respect to co-sleeping, Hannah and Michael displayed a large and immediate treatment effect. For Hannah, these treatment gains deteriorated rapidly towards the end of intervention, however for Michael the effect was maintained through to follow-up. The improvement seen in the boys' co-sleeping, and downturn seen in Hannah's co-sleeping may be a result of differences in parental adherence to treatment.

Then again for early-morning wakings and sleep onset latency problems observed in Hannah and Robert, FBA served to inform treatment. It was reported at initial assessment that the two children would in these occurrences engage in preferred activities or entertaining oneself with preferred objects (e.g., toys). From such information, the function of these problematic sleep behaviours was determined to involve escape from bed in order to relieve boredom from lack of sleepiness. The rationale behind this conclusion was that the children were not fatigued enough to either fall asleep at their pre-intervention bedtimes or wake at an appropriate time in the mornings, evidenced by the fact that they were seeking out stimulating activities and objects that one would in a wakeful state. The proposed tactics to resolve these issues were to enforce a faded bedtime procedure, keep to a specific sleep/wake schedule, and to prevent daytime napping. It was hypothesized that these procedures would work to increase biological sleep pressure. Following the implementation of these treatment methods, Robert displayed a reduction in SOL

and presented with a much more consistent range of SOL durations, compared with his baseline patterns. Hannah's SOL appeared to have been improving independently during baseline, before the introduction of intervention. Nevertheless, her SOL continued to consolidate throughout intervention, suggesting that even if it was not intervention that was the catalyst for behavioural change, it did not make matters worse and perhaps even facilitated the maintenance of her improvement. Although not graphed, early morning wakings were eliminated for both children upon the commencement of intervention. These findings suggest that the intervention procedures that were suggested based on the results of FBA had a positive effect on the SOL and early morning wakings of the two participants.

FBA has been shown to be more effective than those that do not conduct this assessment procedure (Beavers et al., 2013; Hanley, 2016; Spruyt & Curfs, 2015). FBA has received some attention in the literature, and demonstrated its ability to guide the development of interventions for children with developmental disabilities (Brown et al., 2013; Didden & Sigafos, 2001; Hanley, 2016; Hanley et al., 2014; Kodak & Piazza, 2008). A substantial evidence-base supporting the use of FBA within populations with higher prevalence developmental disabilities has subsequently amassed (Campbell, 2003; Hanley et al., 2014; Hansen & Wadsworth, 2015), however only two studies have made use of a formal functional assessment procedure in the formulation of sleep interventions for children with RGND (Didden et al., 1998; Weiskop et al., 2005). Utilization of FBA increases the likelihood of the overall effectiveness of treatment, by only suggesting the inclusion of methods that are proven to be effective at rectifying the specific established causal association with the specific problematic behaviour. Furthermore, FBA cuts down on redundant treatment procedures that would likely complicate matters and unnecessarily overburden the parents, who are typically charged with carrying out such interventions. The treatment effects produced in the current study provide support for the existing body of literature that attests to the notion that multiple factors are at play in the

presentation of sleep problems, and that multi-component interventions are therefore necessary to treat them (Singh & Zimmerman, 2015; Spruyt & Curfs, 2015), by providing further validation for the effectiveness and applicability of the procedure in the treatment of sleep problems in children with RGND.

While the progress made by the participants implies that the intervention package as a whole was effective, as evidently was the use of FBA to inform the selection of specific procedures, it is difficult to attribute the treatment effects to the specific intervention procedures, especially considering that all procedures were implemented simultaneously at the commencement of the intervention phase. One way in which this investigation could be expanded upon could be to stagger the different interventions such that each individual procedure is introduced one by one, so that they can be measured separately. This would therefore enable more precise inspection of associations between certain intervention procedures and change in outcome variables.

Research Question Two

The impact of successful treatment on general child behaviour, communication, and RGND symptomatology. The three children in this study completed their individualized sleep interventions successfully. Improvement in their child's daytime behaviour following treatment, specifically in relation to mood, was noted by the parents of two of the participants, as reported in the post-treatment interviews. These reports of perceived behavioural change were somewhat supported by the children's scores on the CBCL. The participants' CBCL data uncovered other behavioural changes that were not detected by the parents. Similarly, some gains in receptive and expressive communication were seen in two of the participants according to their pre- and post-intervention VABS-II scores.

CBCL. The internalising behaviour scores of Michael and Hannah at baseline were within the normal range, but for externalising behaviours and their total scores, Michael scored within the normal range while Hannah scored within the clinical range. The CBCL was not completed at baseline by Robert's parents, however his post-intervention scores were all within the normal range. Michael's scores did not change significantly following treatment, and he remained within the normal range for internalising and externalizing behaviours. Hannah on the other hand produced significantly lower scores following intervention, shifting from the clinical range for externalising behaviours and total score at baseline, to the borderline range post-intervention. Her post-intervention internalising behaviour score remained within the normal range. In the case of the participants of the current study, it appears that there was little change in the children's internalising and externalising behaviours when they were within the normal range prior to commencing with treatment, but a sizeable improvement in said variables was observed when they fell within the clinical range, and thus more severe, before intervention was introduced.

The improvement seen in Hannah's externalising behaviours following the sleep intervention could be attributed to a number of possible causes. One such explanation could be that there are similar mechanisms behind sleep difficulties and other problem behaviours. The same antecedent- and consequence-related factors that are maintaining sleep difficulties may also be maintaining externalising behaviours in a child. Alternatively, it may be that the lack of sleep resulting from sleep disturbance acts as a catalyst for other challenging behaviours. It is also possible that the skills and techniques learnt by parents through the sleep intervention, and the improved sleep that they might enjoy as a consequence of their child's improved sleep behaviours, may aid the parents in being able to better manage their child's externalising behaviours in a more informed and rested manner.

VABS-II. Before intervention, the three participants' age-equivalent scores on receptive communication put them at a significantly younger age and correspondingly lower level of competency than their neurotypical, same-aged peers according to the VABS-II. This discrepancy was even greater for expressive communication for two of the participants. The largest disparity in this sense was observed in Michael, who had an age-equivalent score in receptive communication at baseline that was eight years younger than his actual age of 12 years at the time of intervention. His baseline age-equivalent score in expressive communication was 10 years younger than his actual age. For Robert, the difference between his actual age at the time of intervention (six years) and his age-equivalent receptive and expressive communication scores was smaller, but still a sizeable gap of approximately five years at baseline. Of the three children, Hannah produced pre-intervention scores that were the closest to her actual age of six, with an age-equivalent score three years younger for receptive communication and two years younger for expressive communication. Michael's and Robert's age-equivalent scores on both the receptive and expressive communication dimensions either remained constant or increased following intervention. A regression in receptive and expressive communication scores was observed in Hannah's case.

The variable directions of change in VABS-II scores across participants could indicate that the relationship between the communication variables and intervention is correlational rather than causal. It may be that the changes in receptive and expressive communication observed following intervention were instead a result of other extraneous factors. These could include a developmental regression as part of their RGND, or perhaps some events or stress experienced by the child causing them to revert back to a simpler level of communication. Further research is needed to explore whether there is a causal interaction between communication and sleep interventions.

The effectiveness of successful treatments on parent wellbeing. Improved parental sleep following their children's interventions was reported in all three post-treatment interviews. The mothers of the three participants stated that they experienced better sleep as a result of their children's progress. Michael's mother also noted that she enjoyed more personal space with the elimination of co-sleeping, and Hannah's mother reported that she felt less stress following intervention, which she attributed partially to the improvements in Hannah's sleep behaviours. Robert's mother no longer worried about Robert during the night and therefore was able to sleep more peacefully. These findings mirror the testimonies reported in the literature related to behavioural sleep interventions carried out within the family context of children with RGND and higher prevalence neurodevelopmental disorders, such as ASD (Allen et al., 2013; Brown et al., 2013; Cortesi et al., 2010; Weiskop et al., 2005). These accounts given at post-treatment by the parents were supported somewhat by the pre- and post-intervention data obtained from the DASS-21, PSQI, and RQI.

DASS-21. Before intervention, depression, anxiety, and stress scores were all within the normal ranges for the parents of the three participants, which is out of line with what would be expected, based on the literature. Past research has found that parents of children with RGND, such as RTT, AS, PWS, Cornelia de Lange syndrome, and Cri du Chat syndrome, are at risk for higher levels of depression, anxiety, and stress in general (Griffith, Hastings, et al., 2011; Hodapp et al., 1997a; McDougall, Kerr, & Espie, 2005; Richman et al., 2009; Sarimski, 1997; Van den Bourne et al., 1999; Wulffaert et al., 2009). Although their baseline results displayed healthy levels of depression, anxiety, and stress, the changes observed in the parents' DASS-21 scores following intervention were mixed. Anxiety scores either decreased or remained constant following intervention for all of the participants' parents. Of note were Hannah's mother's anxiety scores, which decreased significantly from 12 to 1. While the DASS-21 does not provide any explanation for

the changes observed in the parents' scores (Moore, 2004), it is possible that decreases in parental anxiety may be related to the higher quality sleep experienced by parents, afforded by their children's improved sleep following intervention. It must also be considered that extraneous variables, such as a decrease in stress relating for instance to work or other family members, may be responsible either partially or entirely for this progress. Depression and stress scores on the other hand showed greater variability in their direction of change. Depression scores increased slightly for Michael's mother, who stated that she was still struggling to relax still in the night, and was still alert to any sounds, despite Michael's improvement. Her stress scores conversely showed a decrease. Both Robert's and Hannah's parents' stress scores increased following intervention. Depression scores increased slightly for Robert's parents, and decreased slightly for Hannah's parents. Robert's, Hannah's, and Michael's parents' post-treatment interviews elucidated an issue they had with the amount of psychometrics that they were required to complete at baseline and then again following the end of treatment. This could be a potential factor behind the increase in their depression and stress scores, however it is also possible that extraneous variables are the cause.

PSQI. Across the three families that participated in the current study, the impact of intervention on parental sleep quality, according to the results of the PSQI, were mixed. Improved global sleep quality scores were seen following treatment for Robert's father, Hannah's father, and Michael's mother. Contrarily, lower global sleep scores were obtained post-intervention for Robert's mother and Hannah's mother, compared to their pre-intervention scores. This finding is inconsistent with other research which has found positive correlations between child sleep problems and lower parental sleep quality (Boergers et al., 2007; Lopez-Wagner et al., 2008; Robinson & Richdale, 2004; Wiggs & Stores, 2001). The post-treatment interviews with Robert's and Hannah's mothers do not provide any unequivocal explanations for their worsened sleep. However, it is reported that Hannah's mother was still co-

sleeping with her younger sister, which could be a possible contributor to the decrease in her global PSQI score at that time. For both mothers, it is conceivable that some unrelated factors may be behind their reduced sleep quality reported at post-treatment, such as stress relating to work or relationships with friends or family members.

RQI. Prior to commencing with intervention, Robert and Hannah's parents had moderate to very high global scores on the RQI, which signifies a high level of satisfaction with their relationships. Comparing these scores to the post-intervention scores, it appears that across the parents, their relationship satisfaction remained consistent, or increased slightly, as was the case for Hannah's mother. It could be theorized that this increase in ratings of relationship satisfaction for Hannah's mother reveals a secondary effect of treatment, whereby improved child sleep behaviours subsequently decreased stress that may have been afflicting familial relationships. It also is possible that other extraneous variables might be responsible for the increased relationship satisfaction reported by Hannah's mother, such as the reduction in work and study stressors that she described in her post-treatment interview.

Minimal attention has been paid to secondary outcomes of behavioural sleep intervention for children with RGND, such as those explored in this study (i.e., general child behaviour and parental wellbeing, sleep, and relationship satisfaction). Moss and colleagues (2014) did not find a statistically significant change in parental stress following treatment, however, a shift from "clinical" to "normal" range from baseline to intervention respectively, was found for the treatment group. This was contrasted by the control group, which remained in "clinical" ranges over all study phases (Moss et al., 2014). These results mirror those of some of the parents of participants in the current study.

Due to the results from psychometrics measuring parental wellbeing and relationship satisfaction being largely mixed within and between respondents, a definitive treatment effect cannot be confirmed. While anecdotal evidence offered through post treatment interviews with the parents indicate that there were some self-perceived improvements in personal well-being and sleep which they felt were associated with their child's sleep intervention, which is in line with past research examining the association between the TD children's sleep and parental wellbeing (Hauck et al., 2012; Hiscock et al., 2008; Lam et al., 2003; Mindell, Telofski, et al., 2009b), it is highly plausible that any positive effect that treatment might have had on such secondary outcomes could be nullified by life stressors unrelated to their child's sleep behaviours (e.g., work-related or financial stress). It is possible that this was the case for those parents whose psychometric results showed a decline in wellbeing and/or sleep variables.

Research Question Three

Parent understanding and acceptability of treatment. All in all, the results of the current study convey a sufficient level of satisfaction felt by the parents of the three participating children, with regards to the intervention process, and interventions themselves. The participants' parents' personal accounts of the intervention and intervention process, communicated by way of the post-treatment interviews, also highlighted an unanimity of perception that the treatment was effective and easy to implement. Whilst the post-treatment interviews contained many positive opinions, all three sets of parents emphasized that they were inconvenienced by the amount of psychometrics that they were instructed to complete at pre- and post-intervention. They also added that they did not understand why this psychometric data was needed.

These parental perceptions of the intervention process and outcomes were corroborated by the results of the TARF-R. Across the all parents, treatment acceptability scores were high, as were their ratings on the reasonableness and effectiveness of treatment. The parents' scores also indicate that they had a good understanding of treatment. Maximum scores for willingness were given by all parents, except Hannah's mother and father, who gave moderate scores. These lower self-report ratings of willingness to carry out the intervention with Hannah were validated by her parents' noncompliance with various aspects of her intervention. Michael's mother's and Hannah parents' scores indicate moderate levels of disruption and time required to implement the interventions. Conversely, Robert's parents scores represent a low level of disruption and time required, which was reiterated in their post-treatment interview when they stated that Robert did not require a very intensive intervention for his behaviour to change. The results also suggest that all of the parents encountered some negative side-effects of intervention.

The incorporation of FBA into the assessment and treatment development processes could be credited for these high treatment acceptability scores and positive personal narratives concerning the intervention. FBA is conducted not only to facilitate the development of interventions tailored to the individual child, but also to their family who serve as the agents of change in the implementation of intervention. Through FBA, the child's unique presentation of problem behaviours, their developmental history, their family and home environment, and the level of support and motivation offered by their parents/caregivers, are ascertained and used to inform their treatment plan. As such, the consideration given to the families and the effort made to create the most feasible and non-disruptive intervention possible is likely to enhance the social validity, comprehensibility, and achievability of treatment. As intervention progresses, collaboration with the family may be continued in a similar vein, taking into account any observations made by the

families and adjusting the intervention to meet their changing preferences and needs. Moreover, interventions that are developed following this FBA-informed protocol are more likely to be implemented as instructed and adhered to, consequently achieving more positive treatment outcomes (Moore, 2004; Reimers, Wacker, & Koeppel, 1987). The benefits of conducting FBA and collaborating with families throughout the intervention process that is highlighted by the current study echoes the findings of other studies that have attributed their treatment success to their use of these approaches (Derby et al., 1997; Jin et al., 2013; Moore, 2004; Piazza, Hanley, & Fisher, 1996; Turner & Johnson, 2012; Wacker et al., 1998; Weiskop et al., 2005).

Future research of this kind could benefit from a reduction in the amount of pre- and post-treatment psychometrics, including only those measures that are absolutely necessary to the investigation. In addition, the provision of a more concrete explanation and justification for their purpose and importance to parents is advised, based on the findings of this study. This should not only increase treatment acceptability, but may also improve treatment adherence and parents' willingness to implement the intervention accurately.

Limitations of the study

Despite the encouraging results of the current study, several limitations must be considered alongside the findings discussed. These include the inability to attribute treatment effects to a specific intervention component exclusively, its dependence on psychometrics for general child behaviour and parental wellbeing, sleep, and relationship satisfaction data, hindered generalizability of results, absence of inter-observer agreement for primary sleep data, and a shortage of follow-up data.

The experimental design of the current study allows for certain conclusions to be derived from the results. That is to say that just as the function of problematic sleep behaviour unearthed through FBA is regarded as instigating and maintaining said behaviour, the function of the alterations in the sleep environment made through intervention may be recognized as being responsible for the behavioural change observed (i.e., reward/reinforcement; sense of self-efficacy or competency on the part of the child). Nonetheless, the inferences that can be made with regard to the contribution of individual treatment components are limited. This is at least in part due to the multiple intervention procedures that were implemented with each of the children in the treatment of their sleep difficulties. The various intervention procedures were introduced simultaneously for each child, rather than in a staggered fashion. This means that it is not possible to isolate the specific methods were responsible for the treatment effects seen, or determine whether the effects were a result of the synthesis of treatment procedures. This is a common limitation referred to in relevant literature, as a drawback of multi-component interventions (Knight & Johnson, 2014; Moore, 2004). Despite this limitation, the multi-component interventions carried out in the current study, and the FBA used to construct them, were successful in treating the participants' sleep disturbance. As such, the findings of the current study still have significant clinical implications.

A second limitation of the current study concerns the reliance on psychometrics to track any change to the children's general behaviour and their parents' wellbeing as a result of treatment. Psychometrics are susceptible to response bias on the part of the parents, responding on behalf of their children regarding their behaviour, and self-report measures of their own wellbeing, as the parents are not blind to the intervention (Weiskop et al., 2005). Further, psychometrics of these sorts only measure perceptions and do not provide objective data (Moore, 2004). A more accurate method of data collection for child-related variables could be to administer these measures to multiple appropriate respondents/sources, which would allow for

the triangulation of data and increase the validity of the results found. Greater precision of measurement could also be achieved by recording the frequency and duration of any behaviours of interest, such as externalizing behaviours. The psychometric measures used in the current study have elicited some useful preliminary insights within the scope of this investigation, which would in future benefit from further research utilizing more quantitative and objective measures.

Thirdly, the current study is limited by the restricted generalizability of the results, due to a number of factors. Among these factors is the small sample size, which despite of its provision of robust internal validity of the findings through the enablement of extensive examination of the functional relationship between the intervention and the outcomes, restricts the external validity. The lack of representation of other RGND diagnoses across the current study's participants also limits the generalizability of the results, given the distinctiveness of each RGND. Furthermore, the variability in the clinical features and behavioural presentation within each RGND hinders the applicability of the results to children with the same diagnoses as those included in this study. Most pertinently, it is not certain that the sleep disturbance exhibited by the children in the current study are representative of all children with RGND.

Another limitation involves the lack of reliability data procured through the examination of video footage, without which inter-observer agreement cannot be established. Due to time constraints, the video footage collected from each of the families had not been coded by the time of submission and the triangulation of sleep measures was not possible. Therefore, parental report via sleep diaries was the sole means of obtaining primary sleep data. Because of this, reliability of their reporting was not confirmed, which may pose as a threat to the internal validity of any conclusions drawn from the results (Didden et al., 2002). Other studies that have encountered this methodological limitation (Weiskop et al., 2005).

The time restrictions surrounding the current study also limited the collection of short-term follow-up data for Hannah, and long-term follow-up data for all of the participants. Consequently, maintenance of the treatment effects observed was not confirmed at the time of submission. It is intended that long-term follow-up phases will be carried out 12-weeks post-intervention for the three participants.

Future Directions

The findings and limitations of the current study provide ample justification and direction for future research.

One possible research endeavor within this area could strive to assess the effect that sleep interventions have on general child behaviour and parental wellbeing, using more objective measures. The current study depended exclusively on psychometrics to obtain this information about the participants and their families, leading to the reduced internal validity of the overall study. A more comprehensive method of measurement could be executed wherein the frequency and duration of specified child behaviours of interest could be charted in a manner similar to that used with the primary sleep variables in the current study. Similarly, variables pertaining to family wellbeing could be measured more objectively by employing such methods.

On the subject of the impact of sleep interventions on the child's family, the findings of the current study point to another encouraging area of interest, that being the effect of treatment on the child's siblings. Hannah's mother reported that following the implementation of Hannah's sleep intervention, her younger sister's sleep also improved. This information was communicated anecdotally through the post-treatment interview, but future research could investigate this effect using more

quantitative measures in order to draw more valid conclusions about the relationship between sleep interventions and sibling's sleep.

Within the current study, the sample size was small and included only a couple of different RGND diagnoses. Due to this, the findings may not be generalizable to the wider RGND population, which contains numerous genetic profiles and phenotypic presentations which may not respond to the interventions employed in this study in a manner alike with the current participants. Therefore, future research should strive to include larger samples, with a greater representation of different ages and RGND diagnoses, so that FBA-based behavioural sleep interventions may become more confidently prescribed within this cohort.

Given that the treatment carried out in this study involved multiple intervention procedures that were implemented at once, it is not possible to determine which individual procedures had the most potent effect. However, there were techniques carried out across all of the participants that were highly acclaimed by the parents, and may therefore be worth consideration for use in clinical practice. The social stories, Groclocks, and sleep-wake scheduling were applauded by the parents in their post-treatment interviews as having made a visible difference to their child's sleep-related behaviours. While this is only anecdotal evidence, these reports along with the success of the overall intervention package should warrant consideration for utilization in future research. In addition, the extinction-based procedures that saw the removal of parental presence at sleep onset and during night wakings were also implemented across the three participants. Although not highlighted in the post-treatment interviews, the amplified behavioural responses or PERBs exhibited by Michael following the removal of parental presence could signal its appropriateness as a treatment technique in this case. The increase in curtain calls, sleep onset latency, and night wakings that occurred for Michael resembles a post extinction response burst, which commonly occurs following the implementation of such

procedures, when parental presence is in fact the reinforcement maintaining their behaviour. The extinction bursts were not long-lasting, and sleep variables did improve over the course of intervention, which lends support for its effectiveness and advocates for further exploration in future research with children with RGND.

Conclusion

The objective of the current study was to gauge the effectiveness of FBA-based behavioural interventions in the treatment of sleep disturbance amongst children with RGND, as well as to ascertain whether there were any secondary effects of treatment related to general child behaviour and parental wellbeing, sleep, and relationship satisfaction, and finally to determine the level of treatment acceptability held amongst the parents of the participants. In conclusion, the study found variable levels of improvement, ranging from no change to complete elimination of sleep disturbance, across the measured sleep variables for all three children, as well as maintenance of treatment effects in cases. Considerable variation was similarly observed in the results of general child behaviour measures and measures of parental wellbeing, sleep, and relationship quality. Following intervention, parents rated their acceptance of treatment highly, and provided praise for the intervention and study process overall in their post-treatment interviews. The overall success of the interventions developed for and carried out with the children in this study provides substantiation for the use of FBA, specifically with children with RGND, however the variability in the results for secondary outcome measures calls for further investigation within this population.

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Appendices
Appendix A: Child Information Sheet



An investigation into the effectiveness of treatments for sleep disturbance in children with rare genetic neurodevelopmental disorders (RGND)

Young Person Information Sheet

Hello. My name is XXX and I am a researcher at the University of Canterbury. I am doing a project about how to help young people sleep better and I would like for you to help me with this.

I am going to be talking to you and your parent/s about ways to help you sleep better. This means I might be Skyping, phoning or texting you, coming to your house, or your parent/s will be coming to see me at the University.

I will be teaching you strategies that you can use, to help you to sleep better. I will ask you to complete some questionnaires so I can find out more about your sleep and the impact it may be having on other areas of your life.

There may be a video camera in your bedroom sometimes. This will help me to understand what you do when you are awake and asleep. Only your parents and other people working on this project will be able to see this video. We may ask you to wear an actigraph. An actigraph is worn on your wrist like a watch and it tells us when you are asleep and when you are awake.

If you do not want to be a part of this project, you can tell me or your parents at any time and you won't need to be a part of it anymore.

If you have any questions you can ask me or your parents whenever you like.

If you would like to be a part of my project then you can sign the attached form. If you do not want to be a part of this project then you can say "no" and no one will mind.

Appendix B: Child Assent Form

Children's Assent Form



The project that XXX and XXX wants to do to help me with my sleep has been explained to me. I know I don't have to be a part of it if I don't want to. If I have any questions I can ask XXX, XXX, or my parents.

- I am happy to be a part of the project and for XXX and XXX to help me with my sleep so I have coloured in the happy face.

OR

- I don't want to be part of the project or to have any help with my sleep so I have coloured in the sad face.



My name: _____

You can give this form back to XXX now.

Appendix C: Parent Information Sheet



An investigation into the effectiveness of treatments for sleep disturbance in children with rare genetic neurodevelopmental disorders (RGND)

Information for Parents/Caregivers

This research has been assessed and approved by the University of Canterbury Human Ethics Committee (HEC 2018/48).

Dear Parent/ Caregiver,

We are a group of researchers at the University of Canterbury. Dr Laurie McLay is a Senior Lecturer in the School of Health Sciences at the University of Canterbury. Laurie has many years experience in working with children and young people with developmental disabilities and their families. Associate Professor Karyn France has lectured here for many years, has conducted research into the treatment of paediatric sleep disturbance and is a registered clinical psychologist with considerable clinical experience in this area. Professor Neville Blampied has a similar history of teaching and research. A number of Masters and PhD students and registered Child and Family Intern psychologists or registered psychologists also work on this project.

We would like you and your child with a rare genetic neurodevelopmental disability (RGND; e.g., Angelman syndrome, Fragile X syndrome, or Smith-Magenis syndrome) to consider participating in this research study. The primary purpose of this study is to investigate the effectiveness of treatments for sleep disturbance in children with RGND. Treatment can include a range of strategies, including both non-traditional approaches (such as white noise) and behavioural interventions. These approaches have been designed to minimise stress as much as possible for the parents and children using them. We are also interested in parents' and children's experiences in using the treatments and any changes to their lives, or their child's lives, which result.

As a part of this study we would also like to investigate the experiences of families in implementing treatments for sleep disturbance, those treatments that they consider to be most acceptable, and the impact of successful treatment of sleep problems on parent and child wellbeing and quality of life. In order to do this we will ask you to complete some questionnaires about you and your child's wellbeing and behaviour at the commencement and conclusion of treatment. We will also ask your perspective on the treatment that was provided. We will do this either during visits to your home, Skype interviews or in a clinic at the University of Canterbury.

If you agree to allow your child to be a part of this study, we will meet with you, or Skype you, to discuss your child's sleep behaviour and find out more about him/her and your family. This initial meeting will last for approximately 1-1 ½ hours. We will then ask you to complete sleep diaries in which you will record further information about your child's sleep patterns. Sleep diaries will be recorded each day throughout all phases of the study, as this will allow us to monitor the effectiveness of the treatment approach. The sleep diaries will take you up to five minutes to complete each night. You will also be asked to complete commonly used questionnaires in order to obtain information about your child's sleep behaviour and the effects of treatment. It will take approximately 15 minutes to complete each questionnaire. When we have established an understanding of your child's sleep behaviour, we will work with you to develop sleep-related

goals for your child. This will involve a second treatment-planning session which will last 1-1 ½ hours.

To help us gather further information about your child's sleep patterns we will bring or send a video camera to your home for some nights over the course of the programme, which is capable of recording all night sleep. In addition we may ask you, if possible, to use an actigraph with your child. This watch-like device records the movements associated with sleep and can be worn on the wrist or ankle, or secured into a pocket on your child's pyjamas. This may offer an alternative to video cameras when appropriate. These methods will allow us to measure sleep behaviour at times when an adult is not present. We will demonstrate and explain how to use each of these methods for gathering information.

When information about your child's sleep behaviour has been gathered, treatment will commence. You will be offered a choice of treatment options which you will then implement with the support of the research team. If you are dissatisfied with the treatment approach or the degree of progress that is being made then you will be offered a choice of another treatment option. We will provide you with all of the necessary information about each treatment approach and we will maintain regular contact with you during treatment. It is anticipated that your involvement in the study will occur over the course of a few months, but will depend on the rate of your child's progress as well as your satisfaction with the progress.

For the purpose of this project, myself, Laurie McLay (lead investigator) and Jenna van Deurs (registered intern psychologist) will be working closely with you to conduct the necessary assessments and formulate interventions. XX, a research Masters student who also works as a part of our sleep team, may look at some of the information that we collect, such as video recordings and actigraph data.

Your child will be assigned a code name to ensure anonymity and anything that you or your child says or does will be kept confidential. The results of the study may be submitted for publication to national or international journals and may also be presented at conferences. No identification of the child or family will be possible from any report, publication or presentation.

If you want to withdraw from the project before completion, you can do this at any time without penalty or repercussions.

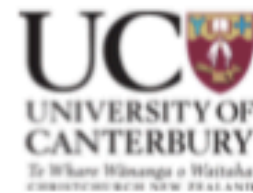
Should you require any additional information about the study or if you would like to access the study findings you are able to do so at any stage. The data which is produced from the research will be kept in a locked cabinet at the University of Canterbury for a minimum of ten years.

If you agree for your child to take part in the research, please sign the consent form that is attached.

If you have any complaints you may contact the Chair of the University of Canterbury Ethics Committee. The contact details are given below.

If you have any questions about this project please feel free to contact Dr Laurie McLay: Phone (03) 369-3522 or, email: laurie.mclay@canterbury.ac.nz

Appendix D: Parent Consent Form



An investigation into the effectiveness of treatments for sleep disturbance in children with rare genetic neurodevelopmental disorders (RGND)

CONSENT FORM FOR PARENTS/ CAREGIVERS

This research has been assessed and approved by the University of Canterbury, Human Ethics Committee (HEC 2018/48).

- I wish to participate in the project, "An investigation into the effectiveness of treatments for sleep disturbance in children with rare genetic neurodevelopmental disorders (RGND).
- 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
- I understand that I am able to request a copy of the results of this research, should I wish to do so, and that these results will be provided for me
- I allow video-taping of my child's sleep behaviour to be completed by the researcher and understand that this videotape will be used for data gathering purposes only. I also understand that I have the right to request that video footage is destroyed at any stage.
- I consent to others, listed below, being involved in the implementation of the intervention

Name: _____

Date: _____

Signature: _____

Others I consent to implementing intervention:

Name: _____

Name: _____

Name: _____

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

Please return this form to XXX.

Appendix E: Child Audiovisual Recording Consent Form



An Investigation into the Effectiveness of Treatments for Sleep Disturbance in Children with rare genetic neurodevelopmental disorders (RGND)

VIDEO/ ACTIGRAPH RECORDING CONSENT FORM

We would like to make video/actigraph recordings of your sleep to help gain information for our project.

Video recordings are often used to record sleep because of the detailed information they give. We use a special video which works in the dark.

An actigraph is a watch-like device which measures movement. It gives us information about your sleep pattern, like how much time each night you spend in different kinds of sleep.

Video/actigraph recordings will help us better understand your sleep difficulties and show us any changes in your sleep over time.

We will only record you with your permission and will always let you know when we are recording.

You can ask us to stop recording or to delete footage at any time for any reason.

Only people involved in the project can view the recordings.

The recordings will be deleted as soon as the information needed has been taken off them.

If you agree to video/actigraph recordings being made please sign below:

Signed:

Date:

Appendix F: Parent Audiovisual Recording Consent Form



An Investigation into the Effects of Treatments for Sleep Disturbance in Children with Rare Genetic Neurodevelopmental Disorders (RGND)

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 your 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 be 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 as soon as required video footage has been coded

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

Signed:

Date:

Appendix G: 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
1 st Night time awakening	Time & Duration of awakening	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
~~~~~								
2 nd 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: