

Using Portable Electronic Devices as part of a Functional Behavioural  
Assessment-Informed Intervention for Children on the Autism Spectrum with  
Sleep Disturbance

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## **Abstract**

Sleep disturbances represent a significant problem for many children on the autism spectrum and their families. Without intervention these sleep problems are unlikely to resolve and have been associated with a wide range of impacts on both child and family. Many factors have been recognised that can increase the complexity of delivering sleep interventions to families with children on the autism spectrum. It is therefore essential that interventions acknowledge these complexities and are tailored to meet the needs and values of individual families. This study investigated the feasibility of using a portable electronic device (PED) as part of a behavioural sleep intervention, informed by a Functional Behavioural Assessment (FBA). Outcomes were evaluated using four single-case studies, with AB designs, with children on the autism spectrum aged three to nine years (two female, two male). The PED intervention was perceived well by parents and while not being able to completely resolve sleep problems, may represent a useful and manageable steppingstone for families in facilitating sleep behaviour change, while reducing child resistance and distress. The findings also supported the use of a FBA-informed behavioural intervention to improve sleep in children on the autism spectrum. While complexity factors appeared to impact treatment adherence and outcomes in this study, the results also highlighted that improvements to sleep can be achieved, despite the presence of complexity.

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## **List of Abbreviations**

APA	American Psychiatric Association
ADHD	Attention-deficit/hyperactivity disorder
ASD	Autism spectrum disorder
CBCL	Child Behaviour Checklist
CSHQ	Children's Sleep Habits Questionnaire
CCs	Curtain Calls
DASS-21	Depression Anxiety and Stress Scale-21
DSM	Diagnostic and Statistical Manual of Mental Disorders
DDU	Duration of device use
FBA	Functional behaviour assessment
GARS-3	Gilliam Autism Rating Scale – Third Edition
IOA	Interobserver agreement
MASC-2	Multidimensional Anxiety Scale for Children 2 <sup>nd</sup> Edition
NZ	New Zealand
NWs	Night wakings
PSD	Paediatric sleep disturbance
PBM	Percentage below the median
PSQI	Pittsburgh Sleep Quality Index
PED	Portable electronic device
PERB	Post Extinction Response Burst
QABF	Questions about behavioural function
RQI	Relationship Quality Index
SATT	Sleep Assessment and Treatment Tool

SOD	Sleep onset delay
SOL	Sleep onset latency
TARF-R	Treatment Acceptability Rating Form-Revised
TD	Typically developing
VSM	Video self-modelling
VSG	Videosomnography
Vineland-3	Vineland Adaptive Behaviour Scales, Third Edition

## Chapter 1. Introduction

In this chapter Autism Spectrum Disorder (ASD) is introduced and common sleep patterns and problems are explored, with a focus on the use of portable electronic devices (PEDs). Potential causes and impacts of sleep problems in children on the autism spectrum are discussed, along with common interventions for childhood sleep disturbance. A discussion of complexity issues with reference to child and family factors and the potential implications for sleep interventions is also explored.

### Autism Spectrum Disorder (ASD)

Autism Spectrum Disorder is defined below along with a summary regarding the prevalence and etiology of this disorder.

**Definition.** Autism Spectrum Disorder (ASD) is a pervasive disorder characterised by difficulties in two domains, namely social communication and interaction, and restricted or repetitive patterns of thought or behaviour. ASD is a neurodevelopmental disorder including multiple sub-types and a wide range or ‘spectrum’ of symptoms and levels of impairment (Seltzer, Shattuck, Abbeduto & Greenberg, 2004). Factors that contribute to the complex and heterogeneous nature of ASD include significant variation in etiology, phenotype and outcome in addition to a high level of comorbidity with both medical and mental health conditions (Seltzer et al., 2004).

**Prevalence and etiology.** Prevalence estimates have varied by country with approximately one in 43 children and adolescents estimated to have a diagnosis of ASD in the United State of America (Xu, Strathearn, Liu, & Bao, 2018) and one in 63 children in the United Kingdom (Baron-Cohen et al., 2009). There is no formal prevalence data available in New Zealand, however it is estimated that approximately one in 100 people have a diagnosis of ASD (NZ Ministries of Health and Education, 2016). The etiology of ASD is thought to be

the result of a combination of biological, genetic, and environmental factors, however, it remains largely unclear. ASD is characterised by high levels of complexity and heterogeneity and no single factor has been identified that explains the presence of ASD (Fakhoury, 2015).

### **Comorbidity and Family Impacts in Autism Spectrum Disorder**

ASD is well recognised as a particularly complex disorder. In addition to the core impairments in the areas of restricted interests and social interaction, children on the autism spectrum often experience comorbid physical, behavioural and psychological conditions. For example, recent research has found that approximately 70% of children on the autism spectrum have at least one comorbid psychological disorder (Gjevik, Eldevik, Fjaeran-Granum & Sponheim, 2010; Leyfer et al, 2006 & Simonoff et al, 2008, as cited in Vriend, Corkum, Moon & Smith, 2011). Additional problems for children on the autism spectrum can include anxiety, eating and toileting problems, sensory issues, sleep disturbance, temper tantrums, aggression, self-injury and hyperactivity (Maskey, Warnell, Parr, Le Couteur & McConachie, 2013).

These challenges present a number of difficulties for families, with studies consistently finding that parents of children on the autism spectrum experience higher levels of stress, more marital discord and more mental health difficulties when compared to parents managing children with other conditions including Down syndrome or cystic fibrosis (Bouma & Schweitzer, 1990; Fishman et al 1989 & Wolf et al, 1989, as cited in Seltzer et al., 2004). Studies have also found that parents of children on the autism spectrum have higher rates of divorce compared to parents of typically developing (TD) children (25% vs 14%; Hartley et al., 2010). In comparison to families with children with other developmental disorders, families with a child on the autism spectrum have also been found to experience more negative effects on wellbeing, social experiences and physical and mental health

(Eisenhower, Baker & Blacher, 2005; Lecavalier, Leone & Wiltz, 2006; McStay, Dissanayake, Scheeran, Koot & Begeer, 2014).

### **Sleep Disturbance in Autism Spectrum Disorder**

Sleep disturbance is one of the most commonly occurring and burdensome challenges facing families of children on the autism spectrum (Elrod et al., 2016; Goldman, Richdale, Clemons & Malow, 2012; Jin, Hanely & Beaulieu, 2013; Malow et al., 2016; Richdale & Scheck, 2009). Children on the autism spectrum are at an increased risk of sleep disturbance, with the presence of a clinical sleep problem estimated to occur in between 31-74% of children (Elrod et al., 2016; Goldman et al., 2012; Malow et al., 2016), which is markedly higher compared with TD children (9-50%) (Goldman et al., 2012; Kotogal & Broomall, 2012). Wiggs and Stores (1996) also reported that when compared with other groups of children with intellectual disability, that children on the autism spectrum exhibited the greatest frequency of severe sleep problems.

### **Definition of Paediatric Sleep Problems**

According to the International Classification of Sleep Disorders: Diagnostic and Coding Manual (American Academy of Sleep Medicine, 2005), sleep problems can be grouped into two main categories: parasomnias and dysomnias. Parasomnias include disorders that interfere with sleep such as bruxism, sleep walking, sleep talking, night terrors and restless leg syndrome. These disorders involve difficulties transitioning between sleep-stages, arousal and partial arousal. Dysomnias, on the other hand, involve difficulties initiating or maintaining sleep and/or excessive daytime sleepiness. This study will focus on sleep problems associated with dysomnias and therefore definitions are provided below.

**Bedtime resistance.** Characterised by child non-compliance following parental requests to prepare for bed and includes a range of behaviours associated with stalling, resistance and refusal to go to and stay in bed.

**Sleep onset delay (SOD).** Once in bed, sleep onset delay is characterised by a delay in falling asleep. Other terms that describe this difficulty include ‘sleep onset latency’ and ‘sleep refusal’. The difference between bedtime and time of sleep onset is calculated to derive sleep onset delay.

**Night awakening.** This involves waking after initial sleep onset and prior to morning awakening. Night waking can increase the likelihood of bed-sharing behaviour when the child seeks comfort or attention to fall asleep again (Cotton & Richdale, 2006).

**Bed-sharing.** Bed-sharing (also sometimes termed co-sleeping) is characterised by a parent or other family member sleeping alongside the child. For bed-sharing to be defined as a problem it must be negatively impacting the family, rather than being a conscious parental choice that can be common practice in many cultures (Carr, 2006; Owens, 2004).

**Early morning waking.** This involves waking before a developmentally appropriate and acceptable wake time in the morning, accompanied by an inability to return to sleep. An important role is played by social norms and expectations in determining what an acceptable wake time is (Arber, Meadows & Venn, 2012).

Research has found that the most common sleep problems for children on the autism spectrum are bedtime resistance and difficulties with sleep onset and maintenance, which includes long sleep onset delay, short total sleep duration, night and early morning waking (Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen & Hansen, 2008; Vriend et al., 2011). If left untreated the occurrence of sleep issues may actually increase and persist through adolescence with different types of sleep problems experienced throughout the age span

(Goldman et al., 2012; Hodge, Carollo, Lewin, Hoffman & Sweeney, 2014; Sivertsen, Posserud, Gillberg, Lundervold & Hysing, 2012).

A general consensus exists within the current literature that a complex interaction between behavioural, biological and environmental factors is responsible for the amplified level of sleep disturbance in children on the autism spectrum (Brown et al., 2014 & Richdale, 2013). Sleep problems may also be an inherent feature of ASD. For example, social communication skill deficits may result in children not understanding behavioural expectations at bedtime, a child 's ability to move from daytime to night-time might be impacted by hyper-reactivity and transition difficulties and repetitive behaviours may require adherence to a strict bedtime routine (Mazzone, Postorino, Siracusano, Riccioni & Curatolo, 2018; Veatch, Maxwell-Horn & Malow, 2015). A variety of causes related to the increased risk of sleep disturbance in ASD have been explored including medical conditions (e.g., gastrointestinal disturbance, epilepsy), neurotransmitter and hormone abnormalities (e.g., melatonin), medications (e.g., stimulants, stimulating antidepressants) and psychiatric conditions (e.g., anxiety, resistance to transitions) (Reynolds & Malow, 2011). Behavioural causes have received less attention, but research has identified that establishing consistent bedtime behaviours and routines may be impeded by the core behavioural deficits and excesses associated with the autism spectrum. For example, children on the autism spectrum may have difficulty transitioning to sleep following a preferred or stimulating activity or may struggle with emotional regulation (e.g., ability to calm self) (Malow et al., 2014). The core communication deficits present in ASD may also result in the child having difficulty understanding their parent's expectations related to going to bed and falling asleep. It may also be particularly challenging for parents of children on the autism spectrum who are juggling multiple other priorities and stressors to effectively convey sleep expectations,

modify the environment and change their own behaviour to promote sleep (Malow et al., 2014).

### **Impacts of Sleep Disturbance in ASD**

Persistent sleep disturbance in children on the autism spectrum has been found to significantly impact both child and parental functioning, wellbeing and overall quality of life (Deliens, Leproult, Schmitz, Destrebecqz & Peigneux, 2015; Jin et al., 2013; Kotagal & Broomall, 2012). The presence and impacts of this sleep disturbance can represent an enduring issue for families as research suggests that without treatment, sleep problems in children on the autism spectrum are unlikely to resolve (Goldman et al., 2012; Sivertsen et al., 2012). When compared to TD children, research has found lower rates of remission in sleep disturbance for children on the autism spectrum, with their difficulties often persisting through adolescence into adulthood if left untreated (Goldman et al., 2012; Hodge et al., 2014; Sivertsen et al., 2012). Sivertsen et al. (2012), found that children on the autism spectrum with sleep disturbance had remission rates of only 8% compared to 52% for children without ASD.

**Child impacts.** Sleep disturbance in children has been associated with a wide range of adverse effects on the child, including cognitive deficits (i.e., attention and memory problems), lower levels of adaptive behaviour, poor academic performance, depressed mood, irritability and physical impacts such as impaired growth and immune function (Pollmacher, Mullington, Korth & Hinze-Selch, 1995; Sadeh, Gruber & Raviv, 2002, as cited by Richdale & Wiggs 2005; Taylor, Schreck & Mulick, 2012). Children with or without ASD who have sleep problems have been found to exhibit more challenging behaviours compared with those without sleep disturbance (Allik, Larsson & Smedje, 2006; Sadeh, 2007).

For children on the autism spectrum, sleep disturbance has also been found to have significant adverse impacts on these children and their families by increasing the frequency and severity of ASD symptoms, including reduced nonverbal intelligence, communication skills, motor skills and adaptive behaviour (Hollway, Aman & Butter, 2013; Mayes & Calhoun 2009; Taylor et al., 2012). Sleep problems may also increase the rates of restricted and repetitive behaviours including preservative interests (Richdale, Francis, Gavidia-Payne & Cotton, 2000). Greater ASD symptomology also appears to increase the risk of sleep problems, which suggests that the relationship between autism severity and sleep is bidirectional (Deliens et al., 2015).

Research has found that children on the autism with sleep disturbance experience more externalising and internalising daytime behaviours including lower adaptive functioning, heightened aggression, hyperactivity, self-injury, social skills deficits and lower overall health-related quality of life, compared with children on the autism spectrum who do not experience sleep problems (Fadini et al., 2015; Goldman et al., 2011; Herrmann, 2016; Maski & Kothare, 2013). The severity of daytime problem behaviour has also been linked to the severity of the sleep problem, and the total number of night time sleep hours has been found to be a significant predictor of the presence and severity of core autistic features, such as impairment in social interaction (Schreck, Mulick & Smith, 2004; Sikora, Johnson, Clemons & Katz, 2012). In a multivariate analysis, Mazurek and Sohl (2016) found that sleep problems accounted for between 22 and 32% of the variance in behaviour problems, indicating that sleep disturbance is associated with behavioural dysregulation among children on the autism spectrum.

It is important to note that the impact of sleep disturbance on daytime dysfunction may be especially taxing for children on the autism spectrum, given that they already have difficulties in social communication and interaction (Krakowiak et al., 2008). This may have

a compounding effect on the child and their family, with sleep disturbance in children on the autism spectrum linked to decreased emotional and psychosocial wellbeing and detrimental impacts on family functioning (Engelhardt et al., 2013; Jin et al., 2013; Kuhlthau et al., 2010; Taylor et al., 2012). Poor sleep has also been found to predict anxiety difficulties in both TD children and children on the autism spectrum (May, Cornish, Conduit, Rajaratnam & Rinehart, 2015).

**Family impacts.** For most children, their sleep disturbance can also adversely impact the sleep patterns and overall functioning of the family unit they live with (Lyons, Leon, Phelps, & Dunleavy, 2010). Childhood sleep disturbance has been associated with marital discord, parenting difficulties and general family stress (Krakowiak et al., 2008; Richdale et al., 2000). For children on the autism spectrum, a strong relationship has been described in the literature between poor child sleep and reduced family functioning, parental wellbeing and marital relationship quality (Doo & Wing, 2006; Richdale, 2013; Richdale & Schrek, 2009; Wiggs & Stores, 2004). In their 2019 review, Martin, Papadopoulous, Chellew, Rinehart and Sciberros, found that sleep problems in children on the autism spectrum were associated with higher parenting stress and poorer parental mental health. Sleep problems in these children have also been associated with reduced sleep quality of parents and other family members, which likely goes on to negatively impact parental physical and mental health and wellbeing (Chou et al., 2012; Hodge, Hoffman, Sweeney & Riggs, 2013).

**Siblings.** Other children within these families have been found to be more likely to have behaviour and sleep difficulties, if they have a sibling on the autism spectrum and sleep disturbance, compared with siblings of children on the autism spectrum without sleep problems (Schwichtenberg et al., 2013). Chou et al. (2012) found that compared to TD children, children on the autism spectrum had more sleep problems, including insomnia, daytime sleepiness and sleep-wake cycle disorders. Their non-autistic siblings also had a

higher risk of having nightmares, insomnia and sleep-talking, when compared to TD children (Chou et al., 2012).

While no causal mechanism has been identified, findings to date identify a connection between sleep disturbance and the multiple child and family impacts discussed above. There has been variability in the literature regarding the strength of these connections. In their 2019 systematic review looking at the association between child sleep problems, parenting stress and parent mental health for children on the autism spectrum, Martin et al, highlighted that the strength of these associations was varied, with effect sizes ranging from small to large. This review also highlighted that the findings indicated a connection but not a causal relationship between child sleep problems in ASD and family impacts.

There is also evidence to suggest that the relationships between child sleep and child factors such as mental health and behaviour are complex and potentially bidirectional (Kelly & El-Sheikh, 2014; Shanahan, Copeland, Angold, Bondy & Costello, 2014). For example, a study by Hollway et al. (2013), explored 45 potential cognitive, behavioural, emotional and physiological risk markers for sleep disturbance in children on the autism spectrum. While they found that several predictors were associated with sleep disturbance, the amount of variance in sleep outcomes that was explained by the predictor variables in this study was modest ( $R^2$  from .104 to .201). Future research is required to clarify the directionality of associations. However, despite the limitations of the correlational findings to date, knowledge of the challenges and impacts associated with sleep disturbance children on the autism spectrum, is key in adapting sleep interventions to better meet the needs of these families.

There is a growing body of literature examining factors that precipitate and perpetuate sleep disturbance in children on the autism spectrum, along with correlates of sleep disturbance. However, there are still few studies focusing on the treatment of sleep problems

for children on the autism spectrum (Cain and Gradisar, 2010; Vriend, et al., 2011). Given the impact of sleep problems on the quality of life of children and families, there is a compelling need to develop effective sleep intervention programs for these children and their families.

**Sleep in context.** As discussed above, a complex interaction between multiple factors may affect sleep for each individual, however, a behavioural model based on operant behaviour theory can be used to understand sleep problems in children. Operant behaviour theory stipulates that the likelihood of a behaviour reoccurring in a similar environment or context, is influenced by what occurs before the behaviour (the antecedent) and after a behaviour (the consequence) (Blampied, 2013; Skinner, 1953). The antecedents that precede a behaviour, serve as discriminative stimuli that signal a behaviour to occur. The consequences that directly follow the behaviour can either increase the probability of behaviour reoccurring through reinforcement, or decrease the probability of a behaviour reoccurring through punishment (Skinner, 1953).

The behavioural model explains that paediatric sleep disturbance can be shaped and maintained by a lack of appropriate stimulus control of sleep-related behaviours, along with the reinforcement of sleep-interfering behaviours (Blampied & France, 1993). It is also recognised that the process of falling to sleep is able to be learnt, as it is an operant behaviour that is under stimulus control and reinforced by the event of sleep (Blampied & Bootzin, 2013). There is a growing evidence base to support the use of behavioural intervention approaches that modify environmental antecedents and socially-mediated contingencies to treat sleep disturbance in children (Jin et al., 2013; Van Deurs et al., 2019).

## **Electronic Device Use and Sleep Disturbance**

Over recent years electronic devices have become more portable, affordable and multifunctional (Gradisar & Short, 2013). The use of electronic media by children and youth continues to increase, with estimates suggesting that 8-18-year olds spend seven and a half hours per day using electronic devices (Rideout, Foher & Roberts, 2010). Approximately 20% of this time is spent using increasingly lightweight portable electronic devices (PEDs)<sup>1</sup>, including cell phones, tablets and hand-held video games (Rideout et al., 2010). This portability along with the availability of wireless technology has contributed to increased use of devices in bedrooms (Bartel, Gradisar and Williamson, 2015). Notably, this portability carries with it an increased opportunity to impact children's sleep.

Use of PEDs at bedtime is a well-established environmental correlate of sleep disturbance in TD children (Bartel et al., 2015). In a 2015 meta- analysis, Bartel et al, found that computer use was associated with less total sleep and internet, phone, computer and video game use were all associated with later bedtimes. There were, however, mixed findings regarding the impact of PED use on other sleep measures, such as sleep duration and sleep onset latency and when an impact was present the effects were not large (Bartel et al., 2015). A notable limitation of this study was that the content of the PED use was not ascertained and it is possible that devices that had the potential to be interactive, and were being used for passive content such as watching movies which, therefore, had less impact on sleep. This is relevant, as previous research has highlighted device content as a key factor relating to sleep impacts and has found, overall, that interactive screen time is more detrimental to sleep than passive screen time (Cain & Gradisar 2010).

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<sup>1</sup> To assist the reader, for the remainder of this paper the term Portable Electronic Device (PED) will be used instead of 'media' when referring to any electronic media presented on a portable device including cell phones, tablets (eg iPads and Android), hand-held video games and portable DVD players. This will include discussions referencing studies where other terms such as 'media' may have been used by the original author.

In the literature for TD children, the potential mechanisms through which use of PEDs at bedtime may affect sleep have been identified as suppression of melatonin resulting from exposure to screen light (Cajochen et al., 2011; Wood, Rea, Plitnick & Figueiro, 2013) and the increased cognitive and physiological arousal resulting from media use (Van den Bulck, 2004), which may lead to a heightened arousal state at bedtime (Mazurek, Engelhardt, Hilgard & Sohl, 2016). The use of PEDs may also engage the user so they become apathetic or distracted, displacing bedtime (Bartel et al., 2015). Therefore, not only does the use of PEDs arouse rather than help to instigate the consummatory response of behavioural quietude before sleep, but it may also strongly (because of immediacy) reinforce sleep competing behaviour, leading to displacement of bedtime.

This displacement of bedtime can be further understood by looking at behaviour chains. Getting to sleep involves a behaviour chain consisting of the full bed preparation sequence prior to the final response of falling asleep. Establishing a regular, consistent bedtime routine ensures that this chain of behaviour is under strong stimulus control, making it more likely that the chain will be followed through to completion terminating with falling asleep (Blampied, 2013). The use of PEDs may represent an alternative to the bedtime routine with immediate and attractive reinforcement. Engaging in the alternative choice of device use, can result in the individual switching to another chain of behaviour which disrupts the bed preparation chain, thereby impairing bedtime compliance and sleep onset (Blampied, 2013).

Most of the research to date regarding PED use and sleep in children has involved cross-sectional methodologies and therefore causation has not been determined. The relationship between technology use and sleep is likely to be very complex, with a variety of factors potentially impacting both technology use and sleep in children. The relationship between child PED use and sleep needs to be considered within a wider family context,

where there are a number of factors that potentially interact to influence this relationship, including parent use of PED, the degree to which parents are permissive about child PED use, parent mental health, child mental health and child preference for using PEDs when struggling to sleep. Further research is therefore required, to define a framework in which to understand the complexity of the relationship between PED use and sleep and disentangle the contribution of factors such as timing, duration of use and content (Gradisar & Short, 2013).

### **Portable Electronic Device Use and Sleep in Autism Spectrum Disorder**

PED use is a particularly relevant issue to consider for children and families affected by ASD, as studies have found that children on the autism spectrum demonstrate a preference for PEDs, spending more time than children without ASD playing video games and watching television (Mazurek & Engelhardt, 2013; Mazurek & Wenstrup, 2013).

Children and youth on the autism spectrum can also find it more difficult to disengage from PEDs and have higher rates of problematic video game use (Mazurek & Engelhardt, 2013; Mazurek & Wenstrup, 2013; Nally, Houton & Ralph, 2000). Contrary to other findings, where gender, demographic and family income factors are usually predictive of video game use, Mazurek, Shattuck and Cooper (2012), found that compared to three other disability groups, the use of video games among youth on the autism spectrum is highly prevalent and largely independent of sociodemographic or individual factors.

There appears to be evidence that PED use may increase the risk of sleep problems for children on the autism spectrum (Mazurek et al., 2016), however, there is a paucity of research in this area. Some findings indicate that sleep for such children was significantly more affected by PED use than for TD children (Engelhardt et al., 2013). Mazurek et al. (2016), is the only study identified to date that specifically examined patterns of night-time PED use in children on the autism spectrum. This study found, using a parent report

questionnaire, that when PED use (TV, video games and computers) was part of the bedtime routine, children on the autism spectrum had significantly longer sleep onset latency (SOD = 39.8 minutes) when compared to similar children on the autism spectrum where PED use did not form part of the bedtime routine (SOD = 16 minutes). However, these findings are correlational, and the authors were not able to make causal conclusions.

Although the reasons for differences between children with and without ASD remain unclear, possible hypotheses specifically relevant to ASD relate to the finding that children on the spectrum have more trouble disengaging from activities, including PED, and that this can lead to increased bedtime resistance and ultimately delayed sleep onset (Mazurek & Engelhardt, 2013). This hypothesis highlights behaviours that are related to ASD, such as difficulty transitioning from evening activities to bedtime routines and reluctance to exchange preferred activities (for example using PEDs), for others that may be more conducive to sleep such as reading a book or listening to a sleep story (Veatch et al., 2015). Additional work is required to better understand the process through which PED use affects sleep in individuals on the autism spectrum.

While there appears to be evidence of an adverse impact of PED use and sleep for children on the autism spectrum, there is also clinical evidence that use of PED can provide a calming influence for these children (Veatch et al., 2015). Other studies have found that parents of children on the autism spectrum use television to manage behaviour, including distraction and minimisation of outbursts and tantrums which may be particularly beneficial for families at bedtime (Mazurek et al., 2016; Nally et al., 2000). Given that children on the autism spectrum are at increased risk of sleep issues and behaviour dysregulation, it makes sense that parents of children on the autism spectrum might be even more likely to utilise PEDs to encourage a calm bedtime transition as part of their nightly routine (Mazurek et al., 2016).

There is also the possibility of reverse causation, where children and adolescents who have trouble sleeping use PEDs as a sleep aid or to fill in time when they are unable to sleep, and therefore report more use of PEDs than their peers who have no trouble sleeping. For example, a study involving Belgian adolescents found that 15% of girls and 28% of boys used video games as a ‘sleep aid’ rather than lying awake in bed, possibly with worries from the day and of the day to come (Eggermonet & Van den Bulck, 2006; Gregory, Noone, Eley & Harvey, 2010, as cited by Gradisar & Short, 2013). Given the presence of such complexity, a one-size-fits all solution is unlikely to be successful and therefore, an individualised functional behavioural assessment is needed to understand and intervene in any instance of paediatric sleep disturbance. The use of functional behavioural assessment in interventions is discussed further below.

### **Interventions for Sleep Disorders in Children**

There are generally three categories of interventions for the treatment of sleep disorders among children (Herrmann, 2016). The first includes various forms of complementary therapies such as weighted blankets and massage. Due to the lack of empirical evidence regarding the effectiveness of such procedures to date, they will not be discussed further (Malow et al., 2012). The second category involves pharmacological interventions, the most widely studied of which is supplemental melatonin, which has been shown to be effective in treating sleep disorders among children on the autism spectrum. In their 2011 review and meta-analysis, Rossignol & Frye, found large effect sizes for significant improvement using melatonin in two key sleep parameters compared to baseline: sleep duration (Hedge’s  $g = 1.97$ ; Glass’s  $\Delta = 1.54$ ) and sleep onset latency (Hedge’s  $g = 2.42$ ; Glasses  $\Delta = 2.18$ ). The focus of the current study, however, is on the third category, namely, behavioural interventions. Research has demonstrated that behavioural interventions are highly effective in treating sleep disturbance in TD children and that such interventions

can also be effective in treating sleep problems in children on the autism spectrum (McLay et al., 2020; McLay, France, Blampied, Danna & Hunter, 2017; McLay, France, Blampied & Hastie, 2019; Turner & Johnson, 2012; Van Deurs et al., 2019; Vriend et al., 2011). Research to date has also suggested that, even when there is an underlying medical or developmental condition, many common sleep problems often have a behavioural component or are behaviourally maintained (Richdale & Wiggs, 2005).

Behavioural interventions as an alternative to pharmacological interventions can be advantageous as they typically have fewer side effects, are generally more socially acceptable and have the potential to result in longer lasting success (McLay et al., 2017; McLay et al., 2019; Richdale & Wiggs, 2005; Vriend et al., 2011). Importantly, behavioural interventions teach children the skills to initiate and maintain sleep. Behavioural interventions can also contribute to an increased sense of competency and control for parents, potentially leading to an enhanced parental coping ability (Wolfson, Lacks & Futterman, 1992, as cited by Vriend et al., 2011).

### **Evidenced Based Behavioural Sleep Interventions for Children on the Autism Spectrum**

While there is a strong base of literature outlining the efficacy of behavioural interventions for the treatment of sleep disturbance in TD children, there is a paucity of research evaluating these approaches specifically for children on the autism spectrum. Of the research that has been completed, the studies often involve small samples and primarily use single subject designs (Turner & Johnson, 2012; Vriend et al., 2011). While this is a valid experimental approach, there are a lack of meta-analytic reviews synthesizing these single subject design studies, contributing to a lack of understanding regarding the overall effect-size for such interventions or the influence of moderating factors. One recent quantitative review of 18 studies, found that multi-component behavioural interventions were effective in

treating sleep disturbance in children on the autism spectrum, with moderate effect sizes (Carnett, Hansen, McLay, Neely & Lang, 2020). Due to the variation in target behaviours and treatment components, the authors were unable to make direct comparisons and identify best practices for particular sleep problems. In their 2011 review, Vriend et al, reviewed 15 studies including one Randomised Controlled Trial (RCT) that involved behavioural interventions for treatment of sleep problems in children on the autism spectrum, using the Chambless and Hollon (1998), criteria for treatment efficacy. Three categories of treatment efficacy are described by Chambless and Hollon (1998): well established, probably efficacious, and possibly efficacious. Multiple studies that compare the treatment to another treatment or wait-list control are required to be classified as a well-established or probably efficacious treatment.

Vriend et al. (2011), evaluated interventions based on whether they could meet the Chambless and Hollon (1998) criteria for a possibly efficacious intervention. The criteria included whether an intervention had proved beneficial to at least three participants in research by a single group, for which there was no conflicting evidence. The possibly efficacious criteria also included a requirement of an adequate number and description of participants, along with methodological rigour and replicability. Vriend et al. (2011), interpreted methodological rigour as increasing confidence that effects are due to treatment and not the expectation of change or consequences of receiving attention, by requiring studies to have a means of controlling for non-treatment effects (e.g., control-group design, A–B–A–B design and multiple baseline design).

Vriend et al. (2011), found that only two behavioural interventions namely standard extinction and scheduled awakenings, met criteria for possibly efficacious interventions for treating sleep problems in children on the autism spectrum. The review acknowledged the relatively few published studies examining behavioural treatment of sleep problems in

children on the autism spectrum compared with TD children. They also noted that positive outcomes had been identified for additional intervention types, but that additional research and study replication was required before these interventions could meet the Chambless and Hollen (1998), criteria of being well-established or probably efficacious (Vriend et al., 2011).

A second review (Turner & Johnson, 2012) was undertaken to determine the efficacy of behavioural interventions to address sleep disturbance in young children on the autism spectrum and therefore only reviewed research literature including children younger than seven years of age. This review utilised the National Autism Centre (NAC) guidelines for evidence strength. They found antecedent approaches such as stimulus fading, scheduled awakenings and bedtime fading met criteria for providing “emerging evidence” of treatment effectiveness. Several consequence-based interventions utilising standard and graduated extinction also fell into this category. Their conclusion was similar to Vriend et al. (2011), in that behavioural interventions represent a promising intervention for treating sleep disturbance in children on the autism spectrum, but noted that further quality well-controlled studies are required to establish an evidence base (Turner & Johnson, 2012). Studies with larger ASD samples have demonstrated positive results, such as Reed et al. (2009) which used a multi-component behavioural intervention to treat sleep disturbance in 20 children on the autism spectrum. Study outcomes demonstrated reduced sleep onset delay, decreased bedtime disturbances and improvement in daytime behaviour. However, the study design did not allow for the effectiveness of specific components to be tested and therefore cannot be used to establish the efficacy of each intervention (Reed et al., 2009).

The following section will briefly outline the most commonly used evidenced-based behavioural interventions for the treatment of childhood sleep disorders in the context of evidence available for children on the autism spectrum.

## **Antecedent-Based Procedures**

Antecedent-based procedures are based on operant behaviour theory and involve stimulus control in which environmental modifications are used to change the conditions in the setting that prompt a child to engage in a behaviour. The antecedent stimulus serves to prime an understanding that a behaviour that occurs in its presence will be reinforced.

Antecedent based procedures can also be used in behaviour chains where each behaviour establishes the occasion for the next part of a routine. Antecedent-based procedures such as good sleep hygiene, bedtime routine, bedtime fading, stimulus fading and visual supports have been used effectively with children on the autism spectrum to ameliorate night wakings, delayed sleep onset and night terrors (Turner & Johnson, 2012).

**Sleep hygiene and bedtime routines.** Sleep hygiene can be defined as “a set of sleep-related behaviours that expose persons to activities and cues that prepare them for and promote appropriately timed and effective sleep” (Jan et al., 2008, p1344). Examples of appropriate cues and behaviours include environmental cues such as a dark, quiet bedroom; physiologic cues such as appropriate timing of meals and avoidance of overstimulating activities close to bed and scheduling regular sleep and wake times. Removal of incompatible sleep behaviours that are maintained by reinforcement such as television or tablet use at night can also be characterised as a sleep hygiene interventions (Cortesi, Giannotti, Ivanenko & Johnson, 2010; Deliens et al., 2015; Gradisar and Short, 2013). An important part of sleep hygiene is a positive bedtime routine which should include a series of sleep conducive and low stimulation activities that are relaxing for the child (Kodak & Piazza, 2008).

Children on the autism spectrum typically respond well to routines and this has been incorporated into sleep interventions, through improvements to sleep hygiene (Kodak & Piazza, 2008; Vriend et al., 2011). However, while addressing sleep hygiene is essential in

ensuring that other, more complex behavioural sleep interventions are successful, sleep hygiene changes alone are unlikely to completely resolve sleep problems (Johnson, Giannotti & Cortesi, 2009; Vriend et al., 2011). As families with children on the autism spectrum often have unique needs, sleep hygiene practices and routines need to be adapted for each individual child and family (Jan et al., 2008; Vriend et al., 2011). Jan et al. (2008), also recognised that changes to sleep hygiene and bedtime routines can often be more challenging to implement for children with developmental disabilities, compared with TD children.

**Bedtime fading, sleep restriction and chronotherapy.** In a faded bedtime intervention, the first step is to identify the time at which a child will naturally fall asleep within 15 minutes of being put to bed. Following this the parents are then asked to put their child to bed at this time. Once the child can fall asleep with little resistance at this time the bedtime is set slightly earlier and earlier each night until a more desired and age appropriate bedtime is achieved. For this to work properly the child is also woken each morning at the same time and not permitted to nap during the day (Vriend et al., 2011).

When implementing sleep restriction, parents reduce the time a child is in bed so that they are only in bed for 90% of the total amount of time they usually sleep (Turner & Johnson, 2012). The child is removed from their bed and engaged in non-stimulating activity if they do not fall asleep within 15 minutes. Their bedtime is gradually brought forward once the child is falling asleep within a short timeframe, until the desired bedtime is reached (Vriend et al., 2011).

Based on three studies, Turner and Johnson (2012), found some evidence for the effectiveness of bedtime fading and sleep restriction interventions in reducing target sleep problems for children on the autism spectrum (Christodulu & Durand 2004, Durand & Christodulu 2004 & Piazza, Fisher & Sherer, 1997, as cited by Turner and Johnson, 2012).

Chronotherapy involves initially putting children to bed at a time which is likely to result in rapid sleep onset. The child's bedtime is delayed by 2 hours each subsequent night, until the child is able to fall asleep quickly at an appropriate time. The child's regular morning wake schedule is maintained during the intervention (Kodak & Piazza, 2008; Vriend et al., 2011). There is limited research available with only one study using chronotherapy as sleep intervention for a child on the autism spectrum. The intervention was undertaken in an in-patient unit and decreased sleep delay and night wakings and increased total sleep duration (Piazza, Hagopian, Hughes & Fisher, 1998).

**Stimulus fading.** Stimulus fading is most commonly used to reduce bed-sharing and involves the parent gradually increasing the distance between themselves and the child to increase the child's ability to fall asleep without parental presence. Only two of the studies reviewed in Vriend et al. (2011), incorporated stimulus fading. In a single case study Howlin (1984), found that stimulus fading resulted in an overall decrease in sleep onset delay, bed-sharing and night wakings. Reed et al. (2009), used stimulus fading as a part of a multicomponent intervention, which was successful in reducing bedtime disturbances and improving daytime behaviour, however the multicomponent nature of the study makes it difficult to isolate the effectiveness of the stimulus fading component.

**Scheduled awakenings.** Scheduled awakening involves the parent awakening their child before the time of the child's previous spontaneous night waking. The awakenings are faded over time until the interval of uninterrupted sleep for the child is prolonged. There has been one single-case multiple baseline design study that utilised scheduled awakenings to successfully reduce the frequency of night terrors and increase total asleep time for three children on the autism spectrum (Durrand, 2002). Due to the robust methodology used in this study, Vriend et al. (2011), classified scheduled awakenings as a possibly efficacious

intervention for children on the autism spectrum. Other antecedent based procedures including visual supports and social stories are covered later in the chapter.

### **Consequence-Based Procedures**

**Extinction.** Extinction interventions are used when sleep-interfering behaviour is reinforced by escape from an undesired event, such as going to bed, or access to attention or tangibles (eg preferred items, food, devices) (Didden et al., 2014). The goal of extinction is to remove the reinforcement to reduce or extinguish the sleep-interfering behaviour. Standard extinction is the most widely studied and empirically supported behavioural strategy and in cases where parental attention is reinforcing disruptive behaviour, involves the parent withholding reinforcement by ignoring all bedtime disruptions after placing the child in bed, until morning (Herrmann, 2016). It has been used successfully for children on the autism spectrum in reducing sleep problems including bed-sharing, falling asleep alone and night waking (Vriend et al., 2011; Weiskop, Richdale & Mathews, 2005). Implementing standard extinction, however, can often result in a temporary but sometimes dramatic increase in negative behaviours referred to as a Post Extinction Response Burst (PERB). This increase in unwanted behaviours can create a challenging and undesirable situation that can cause both the parents and child distress (France & Blampied, 2005; Kodak & Piazza, 2008). Parents have also reported concerns that leaving their child to cry without comforting them may be causing the child psychological trauma (Didden, Curfs, Sikkema & Moor, 1998, as cited by Turner & Johnson, 2012). It may therefore become difficult for parents to continue with the intervention and an increase in negative behaviour, child distress and parental stress may be magnified for parents of children on the autism spectrum, who are already dealing with an increased level of stress (France & Blampied, 2005).

**Graduated extinction.** Graduated extinction is a modification to standard extinction which involves the parent checking on the child on a regular predetermined schedule (e.g., every 5 minutes), which is not contingent on the child's behaviour. The interval between checking is then gradually increased over time (Turner and Johnson, 2012; Wiggs and France, 2000). This approach can be considered more socially acceptable to parents as they are able to continue checking on their child, while still gradually reducing the attention that the child receives. There is some evidence of positive improvements using standard extinction techniques, as part of a multi-component sleep intervention for children on the autism spectrum (Moore, 2004; Knight & Johnson, 2014). More methodologically rigorous studies are required, before graduated extinction can be rated as a possibly efficacious treatment for children on the autism spectrum (Vriend et al, 2011).

### **Multimodal interventions**

The majority of studies to date have focused on multimodal treatment programs utilising various combinations of behavioural interventions. Multicomponent behavioural interventions have been found to be effective for treating sleep disturbance in children on the autism spectrum (Cuomo et al., 2017; Knight & Johnson, 2014; McLay et al., 2020; Moon et al., 2010; Montgomery, Stores & Wiggs, 2004; Moore 2004; Reed et al., 2009; Rigney et al., 2018; Vriend et al., 2011; Yu et al., 2015). Treatment components often included in multimodal intervention programs include, improvements to sleep hygiene practices, stimulus fading, extinction-based procedures, visual supports, parent education and reward systems (Montgomery et al., 2004; Van Deurs et al., 2019, Vriend et al., 2011).

### **Functional Behavioural Assessment in Interventions**

Functional Behavioural Assessment (FBA) is a process used to understand the nature of a child's sleep problems by identifying the specific factors that maintain each child's sleep

difficulties. The FBA is focused on identifying the function, or purpose of the target sleep related behaviour for each child, by examining the behaviour within the context of the environment. FBA assists in customising a treatment plan for an individual by focusing on the specific controlling variables that evoke or maintain the target behaviour (antecedents and consequences) which assists in determining the function of their behaviour. When used in sleep treatment, FBA informs the modification of the antecedents and consequences that are maintaining sleep problems for the child, with some common examples including; parental presence during sleep onset, lack of sleep hygiene and routines and use of a tangible item which can be stimulating, such as PED and/or self-stimulating behaviours (Blampied, 2013).

FBA also considers unique family circumstances in the development of an individualised treatment plan, which has been found to enhance the effectiveness of a behavioural intervention and increase the likelihood of a positive treatment outcome (Jin et al., 2013). Research also indicates that parents prefer tailored interventions which have been informed by a thorough assessment of their unique situation, which is likely to increase their level of engagement in the intervention (Beresford, Stuttard, Clarke & Maddison, 2016). Tailoring the approach for each child and family increases the effectiveness of the resulting intervention when compared to generic treatments (Blampied, 2013; Jin et al., 2013). A lack of success in sleep interventions has been associated with approaches that target behavioural topographies that appear similar across cases, rather than identifying and addressing the function of the sleep-interfering behaviour for each individual (Brown & Piazza, 1999; Kodak & Piazza, 2008). For example, leaving the bedroom after being put to bed may be functionally maintained by access to tangibles for one child, and social attention for another, which indicates the need for different interventions (Kodak & Piazza, 2008). Due to the heterogeneity present in ASD, the factors that may be rewarding or punishing for one child might be expected to be quite different to those of another child, suggesting that an

individualised approach such as FBA would be more effective in ameliorating sleep difficulties (Richdale & Wiggs 2005). There is a growing evidence base to support the use of FBA-informed sleep interventions in treating sleep disturbance in children on the autism spectrum (Jin et al., 2013; McLay et al., 2017, 2019; Van Deurs et al., 2019).

### **Promising Supplementary Intervention Tools for Children on the Autism Spectrum**

Research has found that multimodal interventions can be effective in improving sleep for children on the autism spectrum (Cuomo et al., 2017; Knight & Johnson, 2014; McLay et al., 2020; Montgomery, Stores & Wiggs, 2004; Moon et al., 2010; Moore, 2004; Reed et al., 2009; Rigney et al., 2018; Vriend et al., 2011; Yu et al., 2015). It is therefore valuable to look at specific tools that have been identified as useful in supplementing interventions for children on the autism spectrum. The intervention tools discussed below require further empirical investigation but have been found to hold promise for treating sleep disturbance in children on the autism spectrum.

**Social stories.** Research has identified that children on the autism spectrum may have difficulty understanding what is expected from them at bedtime and increasing this understanding therefore represents a potential target area for intervention (Kotagal & Broomall, 2012; Veatch et al., 2015). A social story is a visual support, that can be used to describe cues in a specific situation and appropriate behavioural responses, expected from children including bedtime and throughout the night. Developed by Gray and Garand (1993), social stories are an example of an antecedent strategy that combine pictures and text geared toward the visual learning strengths of children on the autism spectrum. Social stories were developed to assist in changes in routines, teaching new academic and social skills and to explain the reasons for other's behaviour (Gray & Garand, 1993). Numerous studies have shown that children on the autism

spectrum learn most effectively through visual means and the literature has consistently demonstrated that social stories can be used to help these children reduce challenging behaviours, introduce new behaviours and routines and teach academic skills (Kuoch & Mirenda, 2003; Scattone, Tingstrom & Wilcynski, 2006; Smith, 2001).

In addition to being a visual support, social stories offer explicit information, are situation specific and can be personalised, and have short learning intervals which also tap into the learning strengths of children on the autism spectrum (Quirnbach, Lincoln, Feinberg-Gizzo, Ingersoll & Andrews, 2008; Smith, 2001). Social stories also represent a tangible intervention tool that can serve as a reminder for parents to ensure consistency in a routine and enhance their skills and understanding regarding effective communication with their child (Moore, 2004).

Moore (2004), is the only study to examine the effect of social stories on sleep problems in children on the autism spectrum and used this technique to illustrate the child's bedtime routine and consequences. The study reported positive effects, including improved sleep onset delay, elimination of bed-sharing and a reduction in disruptive behaviour at bedtime (Moore, 2004). However, the social stories in this study were paired with a graduated extinction strategy, which makes it difficult to isolate the effectiveness of the social story technique. There is a need for further research looking at the effectiveness of social stories, to address sleep disturbance in children on the autism spectrum and it would be beneficial to include a comparison against regular bedtime stories (Moore, 2004).

Research to date indicates that social stories cannot be considered an efficacious evidence-based procedure as social stories alone have not produced substantial and enduring changes (Test, Richter, Knight & Spooner, 2011). However, social stories have been used as

part of multicomponent sleep interventions which have been effective in reducing sleep onset delay and duration of NWs and eliminating bed-sharing (McLay et al., 2017; McLay et al., 2019; Souders et al., 2017). Social stories are generally considered to be a useful and socially valid tool to complement more complex behavioural sleep interventions for children on the autism spectrum (Moore, 2004; Test et al., 2011).

**Other visual schedules and cues.** As noted above, children on the autism spectrum learn best through visual means (Ganz, Earles-Vollrath & Cook, 2011; Simpson, Myles & Ganz, 2008). Visually based schedules or routines, therefore, have been used with children on the autism spectrum in a variety of domains to increase work completion, facilitate play and reduce problem behaviour (Ganz et al, 2011). Technology is also increasingly being used to provide visual supports. For example, video modelling using the self or others has been used to improve daily living skills (Keen, Brannigan, Cuskelly, 2007), reduce problem behaviours (Buggey, 2005) and improve play skills (Hine & Wolery, 2006). The Groclock™ is a digital clock that is designed to assist children in identifying whether it is awake time or sleep time. The clock has a bright screen that displays a sun to indicate wake time or a star to indicate sleep time which can be programmed by parents or clinicians. The Groclock™ has also been used to support behavioural interventions in a growing number of studies (McLay et al., 2019; Van Deurs et al., 2019).

As part of a behavioural sleep intervention Weiskop, Mathew and Richdale (2001), utilised visual supports and presented the bedtime routine for a five-year-old boy with Autism, pictorially on a chart. The programme which also included reinforcement, partner support strategies and extinction procedures was successful in eliminating night settling difficulties, night waking and bed-sharing.

While further research is required before visual supports such as social stories can be classified as an evidenced based strategy for sleep intervention, they have been recognised as a useful complementary tool as part of more comprehensive interventions which help to aid the child's comprehension of an intervention (Test et al., 2011).

### **Complexity in ASD and Intervention**

ASD is a particularly complex disorder and can therefore present many challenges in conducting research into effective sleep interventions (Vriend et al., 2011). These challenges include the highly variable expression of ASD in terms of symptomology, level of adaptive functioning and cognitive ability (Vriend et al., 2011). For example, in children with autism, language and cognitive abilities can vary greatly (APA, 2000), thus, while children on the autism spectrum who are high functioning may need only verbal instructions to outline contingencies and desired behaviours, nonverbal children on the autism spectrum may require visual-based supports or even physical prompting (Turner & Johnson, 2012).

Other challenges of complexity involve the high rate of co-occurring health-related, psychological and behavioural problems commonly associated with the autism spectrum (Moore, Evans, Hanvey & Johnson, 2017). Among the most common psychological co-occurring conditions are anxiety and attention-deficit hyperactivity disorders (ADHD). This requires important consideration as these disorders are also associated with sleep difficulties (Hollway et al., 2013; Mayes & Calhoun, 2009; Mazurek & Petroski, 2014). Manohar, Kuppili, Kandasamy, Chandrasekaran and Rajkumar (2018), found that for children on the autism spectrum, the severity of comorbid ADHD also predicted parenting stress and coping which, in turn, impacted parents' ability to engage in interventions.

In their 2017 review, Muskens, Velders and Staal found the presence of medical disorders in children and adolescents on the autism spectrum was widespread: children on the

autism spectrum were found to have significantly higher rates of gastro-intestinal symptoms (Valicenti-McDermott et al., 2006), allergies and autoimmune diseases (Zebro et al., 2015) and epilepsy (Mouridsen, Rich & Isager, 2013). There is evidence that some of these medical comorbidities, for example, seizure activities / epilepsy, asthma, gastrointestinal problems and acid reflux, could also negatively impact sleep (Hollway et al., 2013; Liu, Hubbard, Fabes & Adam, 2006; Mannion, Leader & Healy, 2013; Moore et al., 2017).

Problematic challenging behaviours are also common, with the majority of children on the autism spectrum possessing at least one challenging behaviour (Hattier, Matson, Belva & Horovitz, 2011). These behaviours include aggression, self-injury, hyperactivity, hypoactivity, property destruction, disruptions/tantrums and stereotypies (Gillberg & Billstedt, 2000; Hattier et al., 2011). Research has found a greater occurrence of problem behaviours in children on the autism spectrum compared with TD children, which can exacerbate functional impairments and increase the level of complexity present in these families (Hattier et al., 2011).

There are also a range of family related factors that have been found to complicate the trajectory of ASD and associated child and family problems (Fountain, Winter, & Bearman, 2012; Lucia & Breslau, 2006; Midouhas, Yogaratnam, Flouri, & Charman, 2013). The families of children on the autism spectrum often experience increased stress, family dysfunction, maternal psychopathology, relationship conflict and marital discord, along with disrupted sleep in siblings and parents (Lopez-Wagner, Hoffman, Sweeney, Hodge, & Gilliam, 2008; Midouhas et al., 2013; Polimeni, Richdale & Francis, 2005; Wiggs & Stores, 2004). Individual parent factors such as maternal overprotection (assessed using the Parental Bonding Instrument; Cox, Enns & Clara, 2000) and inability in keeping sleep schedules have also been associated with the sleep problems of children on the autism spectrum and their siblings (Chou et al., 2012). Waddington et al. (2020), also found that greater maternal autism

traits and anxiety were associated with and predicted increased sleep disturbance in children on the autism spectrum.

The cost of raising a child on the autism spectrum has been estimated at approximately three to five million dollars more than a typically developing child (Lord & Bishop, 2010, as cited by Karst & Van Hecke, 2012). Factors such as a lower socioeconomic (SES) environment may also add complexity and impact sleep conditions for a child, with lower family income found to be associated with sleep disturbance in children on the autism spectrum (Waddington et al., 2020). Gabriels, Hill, Pierce, Rogers & Webner (2001), also found that low SES was a significant predictor of treatment outcome for children on the autism spectrum.

The additional time and care requirements for families of children on the autism spectrum has also been found to impact the primary caregiver's ability to return to work and parental opportunities for self-care. Seltzer et al. (2001), found that mothers of children with developmental disabilities worked eight weeks less per year, compared to mothers of children with other mental health difficulties. Compared to mothers of TD children, mothers of adolescents on the autism spectrum were found to devote more time to providing childcare and completing household work and less time participating in leisure activities (Smith et al., 2010a, 2010b). A reduced ability to work and allocate time for self-care increases financial stressors on families and may also diminish parents' opportunities for both social and emotional support, which may impact on their ability to participate in a sleep intervention.

In 2016, Bersford et al, conducted a qualitative analysis of parental responses to sleep management interventions (SMI) for children with neurodevelopmental disabilities. Parents of children with a diagnosis of ASD believed that the inherent characteristics of ASD was a

barrier to successfully implementing a sleep intervention. Parents also highlighted a lack of options regarding the sleeping environment (e.g., a child needing to share a room with a sibling) as a significant barrier. Parents also reported that school holidays and even weekends could impact their ability to implement a new sleep routine (Beresford et al., 2016).

It is therefore important to consider the relationship between child and family characteristics and sleep for children on the autism spectrum, as these factors may add complexity and have significant implications for the delivery of a sleep intervention (Waddington et al., 2020).

### **Acknowledgement of Complexity in Developing Interventions**

While the presence of complexity relating to children on the autism spectrum and their families has been recognised, there is a lack of research investigating how such complexities influence treatment outcomes for children on the autism spectrum (Howlin, Magiati & Charman, 2009). Reliable predictors of success or failure, that would assist clinicians in recognising which specific child and familial factors need to be addressed in treatment planning to minimise non-responders, have not yet been identified. While there is a growing research base which looks at the impact of sleep disturbance in children on the autism spectrum on the child and family unit, there is a paucity of research involving behavioural sleep treatment which has looked at the factors associated with the child and family unit and the effects of these elements on sleep and intervention success for children on the autism spectrum (Kazdin, 1995).

Kazdin and Whitely (2006), is the only known study that focused on using complexity to predict treatment outcomes in children with oppositional defiant disorder or conduct disorder. They referred to the following complexity hypothesis: ‘more severely disturbed or complex cases are not likely or are less likely to respond to Evidence-Based Treatments

(EBTs) and may make the results of controlled trials less generalizable to clinical work' (Kazdin & Whitely, 2006 p456). Multifaceted case complexity was considered across four domains including the severity of child dysfunction, socioeconomic disadvantage, parent and family function and barriers encountered during treatment. They found that case complexity or comorbidity did not necessarily influence treatment outcomes (Kazdin & Whitely, 2006). Representing some support for the complexity hypothesis, 'parents perceived barriers associated with treatment participation', was significantly associated with therapeutic change, with children from families perceiving barriers to treatment changing significantly less than families that perceived fewer barriers. However, it should be noted that the group with greater barriers to treatment still showed a large effect size for therapeutic changes. (Kazdin & Whitely, 2006). Identifying and addressing perceived barriers to treatment for each family would be valuable in enhancing the social validity of a proposed intervention, which would help in promoting treatment adherence and therefore better outcomes (Turner & Johnson, 2012).

In the context of ASD and sleep, a Master's thesis (Chow, 2019), is the first study to look at whether three components of family complexity, namely parental relationship, maternal wellbeing and paternal wellbeing could predict sleep treatment outcomes in children on the autism spectrum. The study found that parental relationship quality was the only predictor of sleep treatment outcomes and only at the short term follow up phase (i.e., 4 to 6 weeks post-treatment). This is consistent with some previous research that has referred to marital problems as a factor which might make sleep problems more difficult to resolve (Carpenter, 1990 & Jones & Verduyn, 1983 as cited by Mindell, Kuhn, Lewin, Meltzer & Scadeh, 2006). The mechanism linking child sleep and marital relationship, however, requires further investigation.

Completed at around the same time, another Master's thesis study (Ng, 2019), endeavoured to further understand the relationship between child complexity variables and treatment outcomes in children on the autism spectrum with sleep problems. The study found that a clinical complexity dimension comprised of behaviour problems and sleep problems was a significant predictor of change in a child's Sleep Problem Severity score (SPS), pre and post treatment. However, the children with more severe SPS had larger changes in SPS consistent with Remington et al. (2007), who found that children on the autism spectrum who had the best response to behavioural interventions had more behaviour problems pre-treatment (i.e., a severity by treatment interaction). While adding to the limited literature investigating the child characteristics that may impact upon response to behavioural sleep interventions in children on the autism spectrum, further research is still required to develop a greater understanding of the multiple potential factors that may impact treatment responses (Ng, 2012).

Due to the very nature of sleep it is most often the parent who is present within the natural home setting, who will need to implement a sleep intervention. However, to date it is a limitation of the majority of research in this area that studies have not collaborated with the family in developing the sleep intervention or even considered the importance of designing a sleep intervention that was a good contextual fit with that particular family (Turner & Johnson, 2012). Family context can also become particularly important when considering cultural diversity and the impact this may have on acceptable sleep practices and defining sleep problems (Turner & Johnson, 2012).

Maglione, Gans, Das, Timbie and Kasari (2012), utilised the findings of a systematic review to create treatment guidelines for nonmedical interventions for children on the autism spectrum. They found inconsistency in the success of treatment programs which suggested that interventions need to build in flexibility and be individualised to the strengths and

challenges of each child. It was also recognised that while families play a key role in intervention, they can be greatly impacted by the demands of caring for children on the autism spectrum and can experience burnout and decreased emotional strength when extending treatment programs into the home setting (McConachie & Diggie, 2007, as cited by Maglione et al., 2012). The guidelines therefore recommend that treatment programs must address the concerns of the family and provide opportunities for their active participation, and highlighted that progress of individuals on the autism spectrum may be impeded if family needs are not adequately addressed (Maglione et al., 2012). To optimise treatment outcomes the development of intervention goals and strategies should therefore be informed by both child and family characteristics (Vivanti, Prior, Williams & Dissanayake, 2014).

In terms of the clinical application of sleep interventions, it is therefore important to consider the presence of such complexities and the unique needs of children on the autism spectrum and their families. Tailoring interventions should consider not only the varied characteristics of the child but also the competencies and preferences of the parents and family to assist in shifting perceived barriers to treatment (Vriend et al., 2011). In order to increase treatment effectiveness and minimise cases of non-responders, it is important to have an awareness and understanding of what specific child and familial complexities need to be considered and addressed in treatment planning (Vriend et al., 2011; Webster-Stratton, 1985). It is also important that alongside identifying effective treatments, we understand the social validity of sleep interventions for these families and the parental experiences relating to the end to end intervention process that may relate to the therapeutic responses from the child (Elliot, 1988). Given that achieving a positive outcome relies on parental engagement with and adherence to the intervention (Beresford et al., 2016), it is important to address these complexities during intervention using the guided participation model, developed by Sanders and Burke (2014). The model aims to create a conducive environment to support change and

encourage parent participation and willingness during the intervention, using four key process areas: (1) practitioner assumptions, beliefs and behaviours; (2) introducing change; (3) supporting change; and (4) preventing and managing resistance (Sanders and Burke, 2014).

### **Potential for using PEDs in Sleep Interventions for Children on the Autism Spectrum**

Despite the evidence that has shown the effectiveness of behavioural interventions which aim to minimise aversive interaction, such as the parental presence procedure (France & Blampied, 2005), families may not be willing or able follow this path. This possibly due to the increased complexity of the household and the impact on parental stress, coping and the ability to follow through on intervention requirements. There is therefore a need to identify effective interventions which can be tailored to the individual strengths of the child involved and also to the preferences and capacity of the family.

Research has found that children on the autism spectrum demonstrate a preference for PEDs over other leisure activities and spend significantly more of their free time on non-social media compared to other disability groups (Mazurek et al., 2012; Shane & Albert, 2008). Children on the autism spectrum also demonstrate a preference for visual learning facilitated by PEDs, with higher levels of attention and learning rates compared with teacher-only scenarios (Shane & Albert, 2008). Presenting instruction using PEDs has also been found to decrease behaviour problems, and increase attention, motivation and learning compared to live personal instruction for children on the autism spectrum (Goldsmith & LeBlanc, 2004). A growing number of studies have successfully used PEDs to facilitate learning of skills and appropriate behaviours in a range of settings (Flores, Faciane, Edwards, Topley & Dowling, 2014; Goldsmith & LeBlanc, 2004; Kuoch & Mirenda, 2013).

As previously mentioned, there is also some clinical evidence that the use of PEDs may help to manage behaviour and have a calming influence before bed for children on the

autism spectrum (Veatch et al., 2015; Nally et al., 2000). There is also evidence that PEDs are increasingly a common household item and that some families are using such devices to encourage a calm bedtime transition as part of their nightly routine (Mazurek et al., 2016).

Researchers are also increasingly recognising the importance of developing treatment interventions that take advantage of this preference for PEDs, however, there are no studies to date which have utilised PEDs such as tablets, as part of a sleep intervention for children on the autism spectrum. Since effectively treating sleep disturbance is likely to improve well-being and overall quality of life for children on the autism spectrum and their parents, it is essential that research continue to investigate different treatment approaches that are practical, relevant and beneficial for these families. Using a PED, such as a tablet, as part of a behavioural sleep intervention may offer a path of least resistance to implementing changes around bedtime and sleep.

As previously mentioned, social stories have been successfully used to teach children on the autism spectrum new behaviours and routines and reduce challenging behaviours (Kuo & Mirenda, 2003; Scattone et al, 2006; Smith, 2001). While traditionally a paper-based intervention, social stories have also been successfully presented in electronic format (Flores et al., 2014; Hagiwara & Myles 1999 as cited by Goldsmith & LeBlanc 2004). Recognising that children on the autism spectrum can be captivated by technology and movies, Gray (2011) developed video social stories ('Storymovies'), using a combination of children acting out appropriate behavioural responses along with supporting text.

The findings of Bandura (1977) that modelling or observational learning can have a considerable impact on skill acquisition, behaviour and development can in part explain why video may be an effective method for presenting a social story. Video is also an economical, readily available and user friendly technology that can be used to model appropriate

behaviour in an engaging way and has been used successfully to teach daily living skills and routines to children on the autism spectrum (Reichow & Volkmar, 2010; Shipley-Benamou, Lutzker & Taubman, 2002). One study also compared in-vivo modelling to video modelling and found that video modelling was more effective in teaching developmental skills to children on the autism spectrum (Charlop-Christy, Le and Freeman, 2000). Kroeger, Schultz and Newsom (2007), also successfully utilised a video-modelling format to increase social skills in children on the autism spectrum aged 4-6 years.

Video technology also provides a vehicle for self-modelling which involves the construction of a story for an individual child that allows the individual to observe themselves successfully performing a behaviour in the appropriate context (Dorwick, 1999). Self-modelling also attends to important features of Bandura's Social Learning theory, encouraging the required attention as a child is most likely to attend to a model similar to themselves, as well as promoting self-efficacy for the child who is able to observe their own success at performing the correct behaviour (Bandura, 1977; Bellini & Akullian, 2007). Flores et al. (2014), successfully used a video social story intervention using an Apple iPad to improve classroom behaviour for 11 children between 3-11 years on the autism spectrum. Researchers were able to create the video social story on this device without difficulty and found the portability of the device beneficial, which highlights the suitability of such devices for use in a range of different household settings.

### **Study Rationale**

Given the significant impacts of child sleep disturbance, further research to identify appropriate interventions that may improve sleep for children on the autism spectrum is an urgent priority for many children and their families. The target of any treatment intervention is to ultimately improve child and family quality of life. Given the reliance on parents to be

supportive of and capable of implementing the intervention, just simply removing access to PEDs which may impact sleep at bedtime, is not always feasible and unlikely to be successful. Parents are also unlikely to use techniques that are too burdensome and do not fit with the family's values (Koegel, Koegel and Brookman, 2003). An intervention that might help families is one that acknowledges the preference of children on the autism spectrum for PEDs and the potential calming benefits such devices can provide to families around bedtime and which focuses on selecting content for these devices that is sleep-facilitative rather than sleep interfering.

### **Research Questions**

The aim of the current study was to add to the current literature relating to sleep interventions for children on the autism spectrum, by exploring the utility of interventions that use PEDs to treat sleep disturbance in children on the autism spectrum. In addition, the study aims to enhance our understanding of the function of PED use for children and parents at bedtime. The interventions focused on regulating the content available on the PED, by replacing the content typically viewed by the child with content that was less stimulating and reinforcing than the child's preferred device content. To ensure that a high level of ethical responsibility was maintained, an additional FBA based intervention phase was made available for those children who did not respond adequately to the primary device intervention focus. The current study was driven by the following key research questions:

1. What is the effect of using an age and developmentally appropriate bedtime story presented on a PED to improve sleep in children on the autism spectrum?
2. What is the effect of using a Video Self-Modelled Social Sleep Story presented on a PED, to improve sleep in children on the autism spectrum?
3. What are the parent's perceptions of the treatment approach using PEDs?

4. What is the effect of a FBA-informed individualised intervention to improve sleep in children on the autism spectrum?
5. What is the effect of improving child sleep on parent and child wellbeing?

## **Chapter 2. Method**

### **The Sleep Research Team**

The studies reported within the researcher's thesis were part of a larger research project focused on sleep disturbance in children on the autism spectrum led by A/Prof. Laurie McLay, A/Prof/ Karyn France, and Prof. Neville Blampied.

### **Ethics and Participant Consent**

Prior to participant recruitment the study received ethical approval from the University of Canterbury Human Ethics Committee (HEC 2018/47; Appendix A). All parents were provided with written information regarding the study and provided signed consent and where possible the child who was participating was given information and also gave their assent. The parent and child information, consent and assent forms are attached in Appendixes B, C, D, E and F. Consent regarding the recording of children's sleep using video was also obtained from families and this form is attached in Appendix G.

### **Research Design**

Four single-case studies are presented, permitting any replication of treatment effects over cases to be examined. Each case constituted an A-B (i.e., baseline, treatment phase) design, where the B (treatment) phase contained a sequence of individualized treatments devised for each case following careful functional behavioural assessment. Through repeated gathering of core dependent variables within and across different phases, single-case designs enable inferences about individuals to be drawn (Blampied, 2013). AB designs permit only limited inferences to be drawn (Hersen & Barlow, 1976), but in the context of this study, were deemed clinically necessary because of the exploratory nature of the investigation of the role of PED use in the maintenance and remediation of sleep problems in highly complex

families with children with ASD. The research is, therefore, properly regarded as an exploratory study rather than controlled research.

## **Participants**

While all four of the children participating in the study, shared an ASD diagnosis and sleep interfering PED use, they varied in regard to their ASD symptomatology, presence of child and family complexities, co-occurring sleep problems and factors contributing to the maintenance of their sleep problems.

**Recruitment.** Participants were recruited for the current study as part of the recruitment process in place for a larger research project running out of the Pukemanu Centre Clinic at the University of Canterbury, focused on sleep disturbance in children on the autism spectrum. Participants were referred by organisations within New Zealand who provide services for children on the autism spectrum and their families. Participants were also sourced via professional contacts and an Autism New Zealand social media group which allowed interested families to self-refer.

**Screening and confidentiality.** An initial screening phone call was undertaken with prospective families to ensure that they were suitable for inclusion in the study. Prior to asking screening questions the researcher conveyed basic information regarding the study including the study aim, summary of procedures and confidentiality. Appendix H contains an outline of screening questions.

**Inclusion/exclusion criteria.** Children were included in this study if they met the following eligibility criteria: (a) aged between three and 11 years, (b) had received or were undergoing assessment for ASD, as verified by a paediatrician, psychiatrist or registered psychologist (c) had a parent-reported sleep problem, including delayed sleep onset (more than 15-20 minutes), frequent and/or extended waking at night (when a child wakes for more

than 5 minutes this is considered 1 night waking), daytime sleepiness and/or problematic bed-sharing, (d) PED use (e.g. tablets, cell phones, television) was interfering with sleep. Children were excluded from the study if they had a physical or medical condition that caused or exacerbated their sleep problems or that impacted their parent's ability to follow procedures recommended as part of the study.

**Participant characteristics.** The participants were three girls and two boys aged between three years and six months and nine years two months. Pseudonyms have been assigned for each participant to maintain confidentiality. Two participants had received a diagnosis of ASD prior to commencement of the study and the other two participants received a diagnosis of ASD during the course of the intervention. At commencement of the study, one child was taking Phenergan and two children were taking melatonin to assist with sleep, as prescribed by their Paediatrician or General Practitioner. Melatonin has been associated with improvements in sleep for children with autism (Rossignol & Frye, 2011). Phenergan, an antihistamine, is also commonly used to treat difficulties in sleep initiation in children (Heussler et al., 2013). The policy of the sleep research team was to complete interventions without altering the prescribed drug regime, unless it was a priority of the family to phase out medication. The prescribing physician was consulted as part of the process of phasing out medication. A summary of participant characteristics as at the time of recruitment can be found in Table 1 (pseudonyms have been used in place of real names to protect participant privacy).

Table 1

*Summary of Participant Characteristics*

Child name	Age (Y-M)	Gender	Diagnosis	Sleep difficulties	Sleep related medication
Anna	9-2	Female	ASD ADHD	Delayed Sleep Onset Device Use Co-sleeping	Melatonin
Jane	5-6	Female	ASD (anticipated)	Delayed Sleep Onset Device Use	Inconsistent Melatonin
Max	6-4	Male	ASD	Delayed Sleep Onset Night Waking Device Use	Phenergan
Colin	3-6	Male	ASD	Delayed Sleep Onset Night Waking Co-Sleeping	Melatonin

**Setting.** The sleep interventions were implemented by the parents in the family home in their child’s natural sleep environment. Two out of the four participants resided outside Christchurch and, depending on the location of the family, clinical interviews and meetings between the research team and the families, were conducted in the Pukemanu Centre Clinic at the University of Canterbury, within the family home or via Skype. The researcher kept in regular contact with participants in the study either via home visits, phone, text, email or Skype.

**General Materials**

As each child had a unique set of sleep problems, the intervention approach and supporting materials were tailored for each individual child, however, some materials outlined below were used across multiple participants.

**Video equipment.** Video recordings were used to gather reliability information on child and parent behaviour and to facilitate a more precise assessment of the child’s sleep

interfering behaviours. Recordings were also used to validate the reliability of parent recorded sleep diaries and provide inter-observer agreement data. Cameras were discreetly positioned in the child's bedroom and were turned on when the child was bid goodnight and turned off when the child woke in the morning. The video data was recorded onto an internal hard drive and later uploaded onto a USB drive either by families or by the researcher for viewing and coding by the research team.

**Sleep diaries.** Within research and clinical contexts, the primary source of information regarding children's sleep is parental report (Blampied, 2013). In this study parents completed paper-based sleep diaries throughout each phase of the study, which provided unique information, including parental responses to sleep-related problem behaviours (Wiggs & Stores, 2004). An example of the sleep diary used in this study is provide in Appendix I.

**Android tablet.** In the case where families did not have access to an appropriate PED to allow participation in the study, the research team supplied the child with a basic Android Tablet, to be used for the purposes of the study.

**Social stories.** An individualised social story was developed based on the goals of intervention, treatment plans and routines for each child included in the study. The researcher guided parents on what photos to collect for each story prior to intervention and the social stories were developed in accordance with Gray's (2010) recommendations. For Colin and Max, a paper-based social story was used as part of their new bedtime routine to aid their understanding of what was expected of them at bedtime and during the night. The social story for the older participants (Anna and Jane) was electronic and presented on their PED, which they were permitted to take to bed with them as part of the intervention. The social story for Anna and Jane aimed to increase their understanding of what was expected of them at

bedtime and provided some further psychoeducation about why sleep is important. The social story was presented on their PED as part of an intervention component aimed at changing the content on the device from sleep interfering to sleep conducive (this rationale is discussed further under Anna and Jane's intervention phase below).

### **Dependent Variables**

For each individual child, dependent variables were determined based on their presenting problems and family goals. A description of the common dependent variables is provided below:

**Curtain calls.** Curtain call frequency was counted per night. Curtain calls were defined as instances where the child got out of bed to seek parental attention or access to preferred items or where the child remained in bed but called out and made requests (e.g., for access to food, drinks, toys or parental attention).

**Sleep onset delay (SOD).** SOD was defined as the time (in minutes) that elapsed between when the child was initially bid good night and the time, they fell asleep.

**Night waking.** The definition of night waking involved an arousal in which the child was unable to reinitiate sleep on their own (Knight & Johnson, 2014). This included any waking during the night, which was before a time in the morning that the family deemed it appropriate for the child to awaken and start the day. The frequency and summed duration (minutes) of wakings per night was recorded from the sleep diary.

**Bed-sharing.** Bed-sharing (also sometimes termed co-sleeping) was defined as the child lying in the same bed (the child's or another individual's bed) for any period of the night with another individual (e.g. parent, sibling, caregiver). This included during the original sleep onset, following a night waking or for the entire duration of the night.

## **Procedure/Study Phases**

Individual case variations will be described under each case study, but the following section describes the five general phases used across cases. Parents completed sleep diaries for each night across all phases and where possible, video recordings were also obtained.

**Phase 1: Assessment.** Upon obtaining the necessary consents, the assessment phase primarily consisted of a detailed Functional Behavioural Assessment (FBA) to inform the conceptualisation of each case. The FBA process was conducted using a combination of an initial semi-structured parent interview, completion of the Sleep Assessment Treatment Tool (SATT; Hanley, 2005), analysis of sleep diaries and video recordings and in some cases completion of the Questions About Behavioural Function (QABF; Matson & Vollmer, 1995).

The clinical interview with parents was conducted at the clinic via skype or by phone, as determined by family circumstances. It took approximately 60-90 minutes and was conducted by an intern psychologist under the supervision of a registered psychologist. To guide questions in the clinical interview, the SATT, an open-ended functional assessment interview was used to identify environmental variables and other factors contributing to the child's sleep disturbance. Other relevant child and family factors were discussed that were pertinent to the study and/or the implementation of the intervention. To further assist in establishing the function of a target behaviour, the QABF, a brief functional assessment checklist, was also completed following the clinical interview. Sleep diaries and video recordings were also analysed to provide further information regarding the child's sleep problems.

The information gathered was then synthesised to complete a functional behavioural assessment and case conceptualization for each child, which involved identifying environmental factors, antecedents and consequences that might be maintaining sleep

difficulties and the possible function of key behaviours (Blampied, 2013). The results of the FBA were used to inform the development of comprehensive, individualised interventions for each child.

**Phase 2: Baseline data collection.** Participants were randomly assigned an initial baseline period of seven, 10 or 14 days, but this could be extended if an unstable sleep pattern or unanticipated change in circumstances or illness was encountered. During the baseline phase, parents were asked to respond to their child in the typical manner and to avoid making any changes.

**Phase 3: Intervention.** The individualised, FBA-informed interventions for each participant consisted of multiple components and were parent-implemented under guidance of the researcher. A variety of factors including antecedents and consequences that were hypothesised to be interfering with each participant's sleep were revealed as part of the FBA process. Intervention components were selected to address the sleep interfering variables for each participant, establish healthy sleep habits and sleep hygiene and to promote compliance with the treatment plan.

The author (or sometimes another member of the team) were in regular contact with parents and provided feedback on treatment implementation, troubleshooting of issues and encouragement. As part of this regular contact, primarily via phone, treatment fidelity was also monitored by recording the level of parental compliance with the treatment plan. The intervention phase continued until a significant reduction or elimination of sleep disturbance for the participant was consistently achieved across a 10-14-day period. In one case cessation of the intervention was initiated by research / therapy team after consistent parental deviation

from treatment plans.<sup>2</sup> As intervention plans were individualised, the intervention phase will be discussed in detail under each participant below.

**Phase 4: Maintenance.** The purpose of this phase was to give families an opportunity to consolidate new skills and behaviours learnt during the intervention into their everyday lives (Blampied, 2013). The research / therapy team had no contact with participants during the maintenance phase and therefore parents were responsible for problem solving any issues that may have arisen during this phase. The post-intervention psychometrics were administered, and a post-treatment interview was conducted as part of this phase.

**Phase 5: Follow up.** Follow-up data was collected for seven days, 12 weeks post-intervention, and was only collected for families that had completed intervention. Due to practical problems this period was longer than seven days for some families. The purpose of follow-up was to measure the maintenance of treatment effects over time.

### **Case Study Participants**

**Colin.** Colin was a three year, six-month-old boy who had been diagnosed with Global Developmental Delay by a paediatrician and was scheduled to be assessed for ASD. Colin showed traits of ASD including communication delay and restrictive and repetitive behaviours. Colin communicated using one- or two-word utterances and received an age-equivalent score of one year, two months and one year, one month on the receptive and expressive subdomains of the Vineland-3 (Sparrow, Cicchetti & Saulnier, 2016) respectively. Colin lived at home with his parents and older sibling (17yrs). Colin's parents suspected that his sibling may share some ASD characteristics and sleep difficulties He had previously been

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<sup>2</sup> The decision was made by the research/ therapy team to cease intervention for this participant as the parental deviation from the treatment plan was resulting in the maintenance of sleep interfering behaviours through both positive and negative reinforcement. The parents agreed with the decision and reported that they were unable to follow through on the limits required to extinguish the child's sleep interfering behaviours.

prescribed melatonin, but his parents felt that this was ineffective and had discontinued using it. Colin attended a local preschool three days a week.

His parents reported that Colin had experienced sleep issues since birth. He was not able to fall asleep without a parent being present and had never slept through the night without waking. Colin's parents had tried a number of things to improve his sleep including white noise, a Lulla Doll (a soft doll that simulates breathing and a heart rate), leaving a lamp on in his room and a programme that involved modifications to an extinction-based procedure, run by a family support service.

As had reportedly been the case throughout his life, Colin had some physical health issues at the time of the study. During the study Colin had a fall resulting in a head injury (not the first he had received), which required medical attention and the suspension of the study for 44 days between the end of baseline and the start of intervention. During the study Colin also had multiple bouts of viral illness resulting in intervention ceasing temporarily.

Colin's sleep environment during the study was vulnerable to noise and disruption from family activities and his family decided to withdraw from the study after a marital separation, in circumstances that were likely to have been stressful for Colin.

***Presenting sleep issues.*** Colin's parents were concerned regarding his inability to sleep independently in his own bed and his reliance on PEDs or parental presence to initiate sleep onset. They were also concerned about the frequency of night waking episodes which required parental attention and on the majority of nights, bed-sharing to return Colin to sleep. Colin would typically fall asleep on the couch each night while watching the television or something on his iPad device, with his parents in the room. After being asleep for 20-30 minutes his parents would carry him and put him in his own bed for the night. During night wakings Colin did not use PEDs, but most often required continued parental presence and

sometimes also a drink of milk to reinitiate sleep. After two or three night wakings, Colin's parents typically moved him into their bed in an attempt to get some sleep. If he woke again but someone was present, he would resume sleep quickly.

Colin's parent's goals were for him to 1) settle to sleep independently in his own bed during sleep onset, 2) remain in his own bed for the duration of the night without requiring parental attention or bed-sharing, and 3) eliminate PED use during sleep onset period.

**Baseline.** Colin's baseline phase ran for 41 days, which was longer expected, as data was not recorded for approximately a week over the Christmas period, so baseline was continued for a further 23 days post-Christmas, to ensure a stable pattern of data was captured.

**Intervention Phase 1.** Colin's primary sleep problems were extended SOD and night waking. Factors incorporated in Colin's case conceptualization of his sleep disturbance included inappropriate sleep dependencies such as iPad use and parental presence and a lack of discriminative stimuli for bedtime. Access to an iPad containing stimulating content at bedtime also represented an inappropriate a sleep interfering behaviour and Colin's natural melatonin secretion may have also been impacted by exposure to bright light on this device. Reinforcement contingencies implicated in maintaining sleep disturbance for Colin included access to tangibles such as milk and PEDs, social attention from parents and escape from falling asleep alone.

Phase one of treatment involved the introduction of a bedtime routine, social story, restriction of content available on Colin's iPad at bedtime, along with withdrawal of parental presence during sleep onset. Treatment components were implemented simultaneously and are discussed below:

***Social story.*** A paper based social story was developed for Colin to reflect changes in his bedtime routine and to help him understand the expectations around sleep. The social story utilised pictures of Colin getting ready for bed and sleeping the entire night in his own bed, without parental presence. Steps included Colin having a bath, putting on his pyjamas, brushing his teeth, watching preferred iPad content before bed in the lounge room, an alarm signalling that it is time for bed, Colin taking his iPad to bed with restricted content to watch until he felt sleepy and sleeping in his bed during the night. The social story was shown to Colin before his preferred iPad time every night.

***Introduction of portable electronic device at bedtime.*** As Colin had been falling asleep every night in the lounge, the first priority for his family was for him to initiate sleep onset in his own bed. To assist with this transition Colin was permitted to take his iPad to bed each night. The content which Colin could view on his iPad in bed was restricted to an age appropriate bedtime story which had been selected in consultation with his parents (refer to Appendix J). The story was an audio book with accompanying words and pictures, but with no animation or opportunity for user interaction. The rationale involved selecting device content that was less stimulating and reinforcing and therefore more sleep conducive, rather than Colin's preferred animated and interactive content which was considered more sleep interfering. Colin was permitted to listen to the bedtime story as many times as he liked before becoming sleepy and falling asleep. It was hypothesised that allowing Colin access to the iPad at bedtime would encourage a calm, less resistant transition to the change in routine of falling asleep in his own bed without parental presence.

***Withdrawal of parental presence during sleep onset.*** Colin was previously falling asleep in the lounge each night, surrounded by one or more of his family members. It was hypothesised that this provided inconsistent and inappropriate discriminative stimuli for falling asleep. Instead, Colin would be put to bed, with his iPad and his parents would bid

him goodnight and leave the room. If Colin left his room, his parents would return him to bed, remind him that it was time to go to sleep and limit any additional verbal or physical interactions. They then left the room again.

***Procedural modifications.*** The treatment plan was altered towards the end of treatment phase one, after Colin had been unwell. His mother had allowed him to watch preferred content (rather than the restricted content) on his iPad to settle him in bed. As his sleep onset period remained under 30 minutes with the use of the preferred content, the research team decided in consultation with his parents that this could continue as the goal of Colin falling asleep in his own bed in under 30 minutes had been met.

***Intervention Phase 2.*** Intervention phase two was introduced on day 32 and involved the introduction of a procedure to eliminate bed-sharing during the night. Parents were asked to ignore calling out behaviours during the night unless Colin was distressed. Parents would initially settle Colin in his own bed with limited verbal or physical interactions. If at any time Colin became highly distressed, a parent could lie on the bed with Colin until he fell asleep. Overtime the intention was to gradually remove the presence of the parent at night (i.e., settle him by sitting on the floor in his room rather than lying on the bed with him).

***Conclusion of treatment.*** After successfully completing phase one of treatment and making a good start to treatment phase two, Colin's mother decided to withdraw from the study due to the marital separation that had recently occurred.

***Dependent variables.*** A key dependent variable for Colin was duration of device use (DDU) and this was defined as the time elapsed from when Colin was initially bid goodnight with his device in bed, until when he fell asleep. Dependent variables for Colin also included night waking frequency and duration.

**Anna and Jane.** Anna and Jane lived with their parents and two siblings who also had neurodevelopmental and sleep difficulties. The parents both rated their relationship quality as high on the Relationship Quality Index (RQI; Norton, 1983), but their mother noted that she and her partner sometimes had differing opinions as to what was expected of the children at bedtime. Their father preferred the children to be in bed for the night by 7.30pm but their mother believed this might be too early, as the children were getting older and would sometimes remain awake in their rooms hours after being put to bed. Their parents also differed in their thoughts around PED use; their father was quite comfortable with the kids playing on the computer or the tablets for long periods of time but their mother had some concerns regarding the amount of time her children spent using PEDs.

**Anna.** Anna was a nine year, two-month-old girl who had been diagnosed with ASD and Attention Deficit Hyperactivity Disorder (ADHD), by a paediatrician and attended school fulltime. Anna was taking Melatonin at the time of the study along with Concerta, a stimulant medication, to manage her ADHD. Her parents described Anna as being able to communicate effectively, although she would take things quite literally. However, her age equivalency scores were below her chronological age for both receptive (2.1 years) and expressive (4.6 years) language, on the relevant subdomains of the Vineland-3 (Sparrow et al, 2016).

Her mother reported that Anna spent ‘too much time on technology’ but that this helped to calm her down and prevent fights with her siblings. Anna’s parents reported that she required strict conditions to fall asleep and could spend up to an hour arranging her soft toys and pillows into a ‘nest’ that she liked to sleep in. For the duration of the study Anna shared a room with her younger sister Jane (five years) which added some complexity to the intervention.

In terms of physical health, her mother reported that Anna had a history of stomach related complaints and had recently been put on medication to manage reflux symptoms. During the study Anna's mother also raised concerns about Anna's anxiety and the impact this might be having on her sleep. Anna's total score on the Multidimensional Anxiety Scale for Children (MASC-2; March, 2012) of 93 indicated a very high probability of clinically significant anxiety.

***Presenting sleep issues.*** Anna's parents were concerned with her difficulties initiating sleep, reporting that she could be awake until 11pm many times per week and communicated to her parents that she 'can't sleep'. Anna often took a PED or DVD player to bed with her that she used for hours. Other nights she read books in bed for long periods of time before falling asleep. Her parents reported that on some nights Anna could fall asleep very easily, usually when she had been awake late the night before. Anna also reported waking during the night several times per week with the need to defecate, waking her mother for help with this. Her parents reported that on other nights she could wake them up to communicate her trouble getting back to sleep and would sometimes end up bed-sharing.

Anna's parents' goals for her were to 1) be able to fall asleep "faster and easier" (interpreted by the team as within 60 minutes of being put to bed); 2) eliminate device use prior to sleep onset; 3) to remain in her own bed for the duration of the night without requiring parental attention to manage toileting.

**Jane.** Jane was a five year, six-month-old girl whose parents had been seeking an ASD assessment for Jane and thought that a diagnosis of ASD was probable. Her score on the Gilliam Autism Rating Scale (GARS-3; Gilliam, 2013) indicated the probable presence of ASD. Her parents described Jane as being able to communicate effectively, however, her age equivalency scores were below her chronological age for both receptive (2.8 years) and

expressive (4.2 years) language, on the relevant subdomains of the Vineland-3 (Sparrow et al, 2016). Jane was not on any medication at the time of the study, however, would often request some of Anna's melatonin when having trouble sleeping, which her parents believed had helped. Jane attended a local school fulltime and her parents reported some concern that she had not yet started to read or hold a pencil properly.

Her mother reported that like her siblings, Jane spent "too much time on technology" and that although she didn't really need this to keep her calm, it would be unfair if she was unable to use the computer when her siblings were. For the duration of the study Jane shared a room with her older sister Anna which added some additional complexity to the intervention.

***Presenting sleep issues.*** Jane's parents were concerned with Jane's reliance on watching a movie or video game instructional videos on the portable DVD player to fall asleep. They reported that sometimes Jane was in and out of bed, requesting food and other items, especially if Anna was making noise or was unsettled. There were no night waking or bed-sharing issues reported.

Jane's parent's goals for her were to 1) be able to fall asleep without the need for the DVD player; 2) reduce the number of times Jane sought parental attention and other items after being put to bed to zero; and 3) to be fall asleep at an age-appropriate time (set after consultation between the parents and the team as by 8.30pm).

### **Procedures for Anna and Jane.**

***Baseline.*** The baseline phases for Anna and Jane ran for a period of seven days, during which a stable pattern of data emerged. Anna and Jane's parents were told to respond as normal without making any changes during the baseline period.

***Intervention Phase 1.*** The primary sleep problems for Anna and Jane were frequent CCs and delayed SOD. Similar factors were implicated in the case conceptualizations for Anna and Jane's sleep disturbance. Antecedent variables included lack of physiological sleep pressure and lack of discriminative stimuli for bedtime. Access to PEDs containing stimulating content at bedtime also represented an inappropriate sleep dependency and sleep interfering behaviour. Anna and Jane's natural melatonin secretion may have also been impacted by exposure to bright light on their PED at bedtime. Reinforcement contingencies implicated in maintaining sleep disturbance for Anna and Jane included access to tangibles such as food, PEDs, torches, social attention from parents and each other and for Anna escape from her fears associated with bed.

Phase one of treatment involved the introduction of restricted content on a tablet at bedtime. During the baseline phase, Anna and Jane were already taking a PED (handheld video game or DVD player) with them to bed each night. In this phase the settings on the device were adjusted so that each child was only able to view an age-appropriate bedtime story, which was selected in consultation with parents. The story was an audio book with accompanying words and pictures, but with no animation or user interaction. Anna and Jane were permitted to listen to the bedtime story as many times as they liked before becoming sleepy and falling asleep. As described for Colin, the rationale involved selecting device content that was less stimulating and reinforcing and therefore more sleep conducive, rather than Anna and Jane's preferred animated and interactive content which was considered more sleep interfering. It was hypothesised that allowing Anna and Jane access to a tablet at bedtime would encourage a calm, less resistant transition to making the first steps in changing their sleep routines and expectations. Both Anna and Jane were reluctant for the content on their tablets to be restricted, however, this step was crucial as access to preferred content on their devices was hypothesized to be their primary sleep-interfering behaviour.

***Intervention Phase 2.*** Phase two of treatment involved replacing the bedtime story content on the tablet with a video self-modelled (VSM) social story. The VSM was under five minutes long and was intended to help Anna and Jane understand the expectations around bedtime and sleep, along with some educational content on why sleep is important. The video included an introduction of each child and what they enjoy doing, where they live and who they live with and portrayed each child getting ready for bed, sleeping in their own bed all night and the next day having the energy they need to do the things they enjoy. The researcher used a voice over dialogue from the child's perspective to accompany the video.

Anna viewed the VSM once during this phase and declared that it did not help her. From this point on in the intervention Anna elected not to take the tablet with her to bed at all and chose to read instead. Jane opted to watch the VSM for the first four nights of this phase and thereafter also elected not to take the tablet with her to bed, describing the content as 'boring'.

***Intervention Phase 3.*** Treatment in phase three, involved the introduction of a modified bedtime routine to establish appropriate discriminative stimuli for bedtime. Anna and Jane shared a room, and this was often disruptive for sleep onset for both children, so a key part of this phase was introducing staggered bedtimes. The youngest, Jane, was put to bed first and the new routine included a book read by a parent. The intention of this was to provide a sleep-conducive activity which would also fulfil her need for parental attention around bedtime. An hour later, when Jane had been given the opportunity to fall asleep, Anna (the older child) would be put to bed. She would have the opportunity to briefly read her book, with a reading lamp, before going to sleep. Bedtimes were also made later and more age-appropriate to increase physiological sleep pressure and close the gap between the ideal and current timing of sleep onset. These treatment components were implemented simultaneously for both girls. During phase three, both Anna and Jane were able to take the

tablet to bed and view the VSM if they wanted to but had long opted not to do so (as explained above).

***Follow up.*** Follow up was conducted approximately eight weeks post intervention.

***Dependent variables.*** The key dependent variables for both Anna and Jane were the frequency of curtain calls and sleep onset delay.

***Inter observer agreement (IOA).*** IOA could not be coded. A video camera was initially installed in Anna and Jane's bedroom, but was removed at the parents' request, due to worries that one of the children was uncomfortable with it being there and therefore disrupting their sleep.

***Max.*** Max was a six year, four-month-old boy with a diagnosis of ASD. He was nonverbal but had ways of communicating such as bringing someone a cup if he wanted a drink. Max received an age-equivalent score of one year, six months and one year, eight months on the receptive and expressive subdomains of the VABS –II (Sparrow et al., 2016) respectively. Max lived at home with his parents and two siblings and attended a special school five days a week, which he enjoyed.

Max's parents reported that he had sleep problems since birth. They had tried Melatonin and believed this had helped Max to fall asleep but found that he would still wake up during the night. At the time of the study, Max was taking Phenergan which, his parents believed "helped a bit". His siblings (aged eight and four) had no reported sleep issues and were typically developing.

***Presenting sleep issues.*** Max's parents were concerned with his difficulties initiating sleep, reporting that he had trouble relaxing at the end of the day and trying to get to sleep. They reported that Max did not appear tired or able to pick up on sleep cues at bedtime and resisted his bedtime routine, climbing on furniture and vocalising. When neither parent was

in his room when he was settling to sleep, Max would get up and play and delay sleep, so one parent would sit with Max until he fell asleep. Max would typically fall asleep watching a movie on the TV in his room, which his parents believed reduced his sleep onset delay (currently between one and three hours). When he woke in the night, he would take a long time to settle back to sleep, sometimes over an hour. While this happened only once or twice a week, he was sometimes awake for between one and three hours and his parents either lay with him in bed or played a movie for him in his room to get him back to sleep.

Max's parent's goals were for him to 1) settle to sleep independently (without parental presence or TV) in his own bed during sleep onset; 2) greatly reduce his sleep onset delay to under 30 minutes; and 3) Settle back to sleep independently if he woke during the night.

**Baseline.** The baseline phase for Max ran for a period of 21 days, until a stable pattern of data emerged. Max's parents were told to respond as normal without making any changes during the baseline period.

**Intervention.** The treatment phase for Max involved a multimodal program with parental presence including a paper based social story, delayed bedtime, the replacement of device use (movie or TV) with relaxing music at bedtime, a variant of the parental presence procedure (see below), setting a consistent morning wake time and use of a reward (see below).

**Social story.** A paper-based social story was developed for Max to reflect changes in his sleep environment and sleep routine. The social story utilised pictures of Max getting ready for bed, listening to music, sleeping the entire night in his own bed and the reward he would receive in the morning if successful. The social story was read with Max each night as part of his bedtime routine.

***Delayed/faded bedtime.*** To increase sleep pressure and to help Max establish consistent discriminative stimuli for going to bed and going to sleep, his parents were asked to delay his bedtime by one hour to move bedtime closer to his natural sleep onset and promote rapid sleep initiation. This bedtime fading procedure relies on promoting internal cues of sleepiness caused by sleep deprivation and aims to establish sleep as a reinforcer (Delemere & Dounavi, 2018). Once the sleep onset period was reduced to a desirable time period, Max's bedtime was systematically moved forward towards an earlier, more age appropriate time (i.e., bedtime fading).

***TV (device use) replaced with music.*** Instead of Max being able to watch TV in bed at night, the TV was replaced by audio only in the form of either relaxing music or white noise. The goal of this component of treatment was to eventually fade out any device-based stimuli that may be interfering with sleep onset. The music was also dependent on Max being present in his bed.

***Parental presence.*** As Max's father had previously sat on a chair by the bed with his laptop until Max fell asleep, a parental presence program was implemented where his father would sit in a chair by Max's bed without his laptop, and gradually move the chair further away, with the end goal of fading the parent out of the room. If Max left his bed before settling to sleep, Max's father was instructed to immediately turn the music off and guide him back to bed. This was to be done with minimal interaction and once Max was back in bed, the music could be turned back on.

***Consistent morning wake time.*** Max's parents were instructed to wake him at a consistent time each morning, to help him in establishing a clear and consistent sleep-wake pattern. Exposure to natural light was advised to aid in waking.

**Reward.** If Max was successful in sleeping in his bed all night, he would be given access to a reward such as a small chocolate. His parents were advised to give this immediately upon waking and explain to him why he is receiving the reward, using visual aids (eg the social story) and verbal praise.

**Dependent variables.** The key dependent variables for Max were SOD and the frequency and duration of night wakings.

### **Participant Complexity Summary**

The participants in the study exhibited a range of complexity challenges, including health, psychological and behavioural comorbidities, that are commonly recognised as being associated with ASD (Hattier et al., 2011; Moore et al., 2017; Muskens et al., 2017). There were also a range of family factors present that potentially add further complexity, including marital discord, room sharing and sleep disruption in siblings (Fountain, Winter & Bearman, 2012; Lucia & Breslau, 2006; Midouhas et al., 2013). One family reported the presence of siblings with an ASD diagnosis as a factor that increased the complexity of implementing a sleep intervention in their household. Several parents involved in the study reported a range of mental health difficulties such as anxiety and depression, along with sleep difficulties of their own. One family reported parental ASD traits. Two families also reported an inability for the mother to return to paid employment, as one or more children in their care were unable to attend school regularly due to ASD related challenges or reoccurring sickness. A summary of the complexity factors for study participants is shown in tables 2 and 3 below. To protect participant privacy, no names or pseudonyms have been displayed.

Table 2

*Summary of Child Complexity Factors by Participant*

Child	Child factors			
	Comorbid psychological diagnosis	Diagnosed physical health issues	Accident / injury prone	Gastrointestinal (eg reflux) / feeding / eating difficulties
A				
B	X	X		X
C	X	X	X	X
D		X		

Table 3

*Summary of Family/Household Complexity Factors by Participant*

Child	Family/Household factors							
	Siblings with ASD or ASD traits	Siblings with sleep issues	Parental psychological diagnosis	Parental relationship issues	Maternal inability to work due to child demands	Child shares room with a sibling	Lack of treatment consistency between parents	Parental perceived barriers to treatment
A	X	X	X		X	X	X	X
B	X	X	X		X	X	X	X
C	X	X	X	X	X			X
D			X	X				

## Assessment Measures

The FBA was conducted using a range of tools. The Sleep Assessment Treatment Tool (SATT; Hanley, 2005), was used to guide open-ended questions in the clinical interview, to assist in identifying factors contributing to each child's sleep disturbance. Parent recorded sleep diaries and analysis of video footage from the assessment phase, along with a brief functional assessment checklist known as the Questions About Behavioural Function (QABF; Matson & Vollmer, 1995) tool, was also used to establish the function of a target behaviour.

*Gilliam Autism Rating Scale –Third Edition (GARS-3; Gilliam, 2016).* The GARS-3 is a screening instrument for individuals aged 3-22 years that is used to assess the probability of ASD and level of symptom severity. Based on the diagnostic criteria of the DSM-V (APA, 2013), the GARS-3 has 58 items and has professional or parent report options. Normed on children and young adults diagnosed with ASD, the GARS-3 has good psychometric properties (Gilliam, 2016). The GARS-3 is consistent and discriminative, with good test-retest reliability, strong inter-rater reliability and fair to excellent internal consistency (for the 6 subscales alpha ranged from .79 to .94) (Gilliam, 2016). In the current study, the GARS-3 was used to corroborate ASD diagnoses and indicate level of symptom severity.

*The Vineland Adaptive Behaviour Scales-III, Parent/Caregiver Rating Form (Vineland-3; Sparrow, Cicchetti & Saulnier, 2016).* The VABS-II is a semi-structured interview which assesses an individual's ability to function socially and independently, within their everyday environment. The VABS-II is designed to measure adaptive functioning and is administered to parents or caregivers of individuals aged 0-90 years (Sparrow et al., 2005). As subsections of the Vineland-3 can be administered independently, only the communication subdomain was used for the current study (Sparrow et al., 2016).

This was used to ascertain each child's level of expressive and receptive language, to ensure interventions were tailored to meet their level of understanding.

### **Sleep Outcome Measures**

*Parent-reported sleep diaries.* The core data relating to the dependent variables was captured using parent reported daily sleep diaries.

*Videosomnography.* Video footage using night-time, infrared video cameras was used when available to supplement data relating to dependent variables.

#### *Child's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000).*

The CSHQ is a parent-report questionnaire consisting of 45 items relating to children's sleep patterns. Parents are asked to report on the frequency of particular sleep behaviours, observed in their child and in addition indicate whether these behaviours are a problem for the family. The CSHQ provides eight subscale scores, for commonly observed sleep disturbances in children, along with a total sleep disturbance score. The CSHQ has good psychometric properties and is frequently used to measure sleep problems both in typically developing and children on the autism spectrum (Goldman et al., 2011; Hodge, Parnell, Hoffman & Sweeney, 2012; Mazurek & Sohl, 2016; Owens et al., 2000). Scores > 41 are indicative of clinically significant sleep disturbance (Owens et al., 2000).

### **Child Daytime Functioning Measures**

The child daytime functioning measures were administered pre-treatment during the assessment phase and post-treatment during the maintenance phase.

*Child Behaviour Checklist for Ages 6-18 (CBCL; Achenbach & Rescorla, 2001).* A measure of daytime functioning, the CBCL is a 113-item caregiver report measure of behavioural and emotional problems in children, including externalising (eg., aggressive) and

internalizing (e.g., withdrawn) behaviour. There is evidence to support the validity of using the CBCL for youth with ASD and it has been used frequently in research investigating sleep in children with ASD and the impact on daytime functioning (Lambert et al., 2016; Pandolfi, Magyar & Dill, 2012). The CBCL was completed by parents during assessment and used to indicate the extent of additional behavioural difficulties experienced by participants.

*The Multidimensional Anxiety Scale for Children Second Edition (MASC-2; March, 2012).* assesses the presence of symptoms related to anxiety disorders in youth. Using parent and self-report formats, the measure aims to distinguish between important anxiety symptoms and dimensions.

### **Parent-wellbeing Measures**

The parent-wellbeing measures were administered pre-treatment during the assessment phase and post-treatment during the maintenance phase.

*The Depression Anxiety and Stress Scales (DASS-21; Lovibond & Lovibond, 1995).* A 21-item, self-report instrument, the DASS-21 is designed to provide an indication of the presence of psychological distress in adults, by quantifying features of depression, anxiety, and stress (Henry & Crawford, 2005). The DASS-21 has been used in research with parents of children on the autism spectrum (Giallo et al., 2011; Papadopoulos et al., 2019) and has good psychometric properties (Henry & Crawford, 2005).

*Relationship Quality Index (RQI; Norton, 1983).* A six-item, self-report questionnaire, the RQI is designed to assess perceptions of relationship quality and satisfaction for couples. Completed separately by each individual, scores are summed to indicate global relationship satisfaction. The RQI was used in the current study to monitor any changes in relationship quality and was completed separately by both parents during assessment and maintenance phases.

*Pittsburgh Sleep Quality Index (PSQI; Buysse, Reynold, Monk, Berman & Kupfer, 1989)*. The PSQI is a self-report, 18 item measure to evaluate the quality of sleep in adults. The PSQI has good psychometric properties and is commonly used to measure sleep quality of parents of children on the autism spectrum in research settings (Buysse et al., 1989; Giallo, Wood, Jellet & Porter, 2011; Hodge et al., 2013).

### **Treatment acceptability**

*Treatment Acceptability Rating Form-Revised (TARF-R; Reimers, Wacker, Cooper, & DeRaad, 1992)*. The TARF-R was administered to assess parents' perceptions of overall treatment acceptability, post-treatment. The majority of items examine ratings of treatment acceptability and three items assess problem severity and the participants' understanding of the intervention approach. A total treatment acceptability score is derived by summing ratings on six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time). In addition, post-treatment parent interviews were conducted to gain qualitative information regarding the social validity of treatment and treatment effects.

### **Data Analysis**

Data for each dependent variable was obtained through sleep diaries and video footage and graphed by phase for each child. A cornerstone of analysis in single case experimental design, is a visual analysis of graphed data to assess treatment outcomes (Cohen et al., 2014). For each participant data is graphed within and between conditions and includes an assessment of trend, level and stability of sleep behaviours.

Psychometric measures were administered pre- and post-treatment and analysed within participants, to identify other changes resulting from the study.

## Chapter 3. Results

The findings for each case study are presented below. First, the quality of the data is discussed, followed by sleep diary data outlining child sleep outcomes as a result of the sleep interventions. Child daytime functioning and parent well-being is then compared pre and post treatment, followed by data on parent perceptions of the assessment and treatment process.

### Quality of Data

After completion of phase 1 of intervention, Colin's family started intervention phase 2, but discontinued their involvement with the study before completion of this phase. Data for Colin is presented in the results up to the point of discontinuation.

Participants had varying levels of compliance with capturing sleep diary data and often reported being too busy or tried to do so. Parents recorded sleep diary data for all dependent variables across baseline and treatment on 23–98% of nights. For Anna and Jane some of these gaps could be supplemented with video footage, when available. On the nights where both data sources were available there was often a discrepancy (as frequently observed) between parental sleep diary and video recorded data for Anna and Jane. This appeared to be primarily due to a lack of parental supervision of Anna and Jane after they were put to bed at night. On day 30 of the intervention, Jane and Anna's parents requested the removal of the video camera as they believed it was interfering with sleep for Anna.

### Data Analysis

**Visual analysis.** A visual analysis across baseline and intervention phases was used by way of individual participant time series graphs, to display changes in sleep across nights and to assess the effectiveness of treatment. This included an evaluation of the level, variability, and trend in data across study phases (Kazdin, 2001).

**Effect size estimate.** To supplement visual analysis, the percentage below the median (PBM; conventionally called the PEM; Percent Exceeding the Median, assuming that increases in the median are desired) was calculated to estimate effect sizes. The PBM is derived by calculating the percentage of intervention data points below the baseline phase median (Parker, Vannest & Davis, 2011). PBM > 90% indicates high effectiveness of treatment; 70-90% moderate effectiveness; and < 70% represents ineffective treatment (Ma 2009).

## **Results – Anna**

**Sleep outcomes.** Anna's data for frequency of curtain calls (CC's) and duration of sleep onset latency (SOL) are shown in Figures 1 and 2.

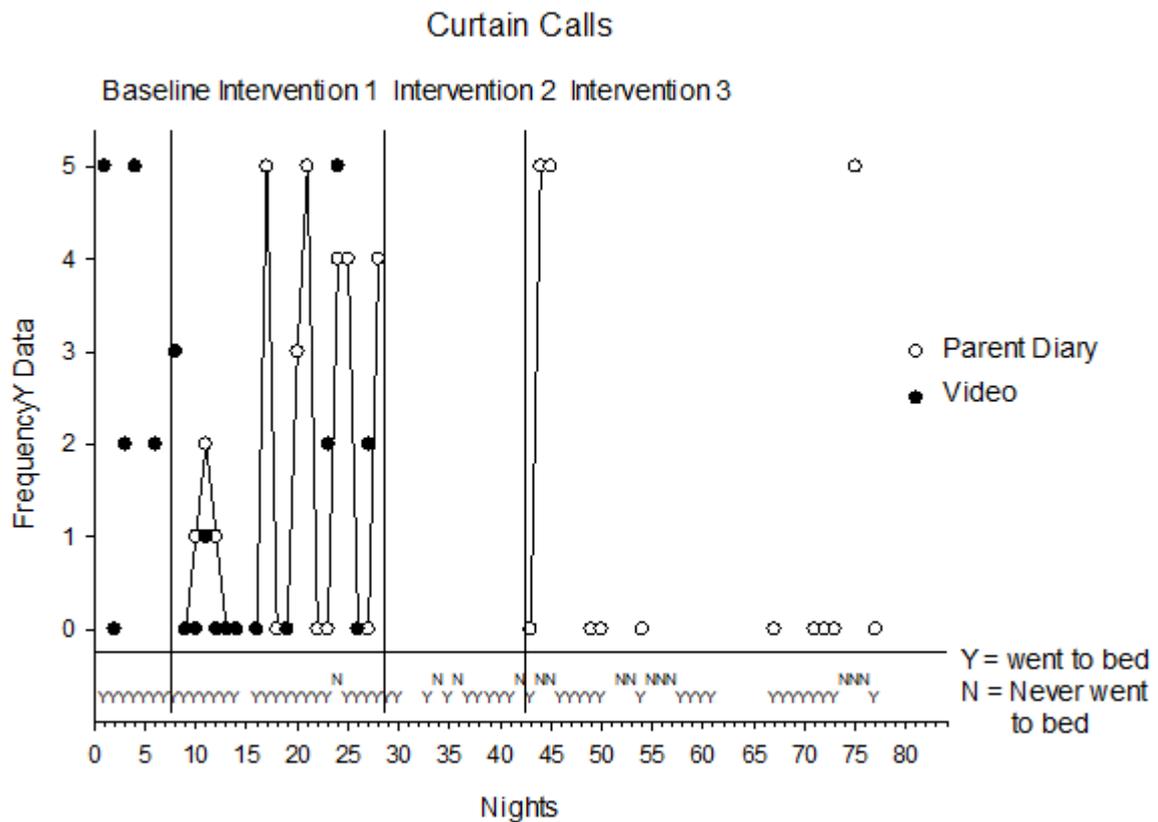


Figure 1. Frequency of curtain calls across baseline and intervention phases for Anna

**Effect on curtain calls.** In the baseline phase, the frequency of CC's for Anna, assessed by video, was variable, ranging from 0 to 5 (Fig. 1). During the first 10 nights of intervention phase 1, the frequency of Anna's CCs, recorded by both video and diary, decreased to between 0 and 2, showing some initial improvement. After this point the frequency of CCs became more variable ranging between 0 and 4 during the remainder of intervention phase 1 and intervention phase 3 and well above the clinical target of 0 (Fig. 1). There was no evidence of a treatment effect. Anna's parents reported not having the capacity to record CC data in intervention phase 2 and were inconsistent with recording CC data in intervention phase 3. They reported variable behaviour from Anna where she would either

have a good night, which is evidenced by several nights of zero CCs during intervention phase 3, or a difficult night where she would refuse to go to bed. Anna’s parents reported difficulty in recording CCs on these nights as Anna did not comply with requests for her to go to bed. Instead, she would sit or stand in the kitchen, often going to bed very late or after her parents went to bed and therefore the actual time she went to bed wasn’t always recorded by her parents. On Figure 1, N indicates the nights where Anna never went to bed before her parents did, and Y indicates the nights where Anna did go to bed before her parents.

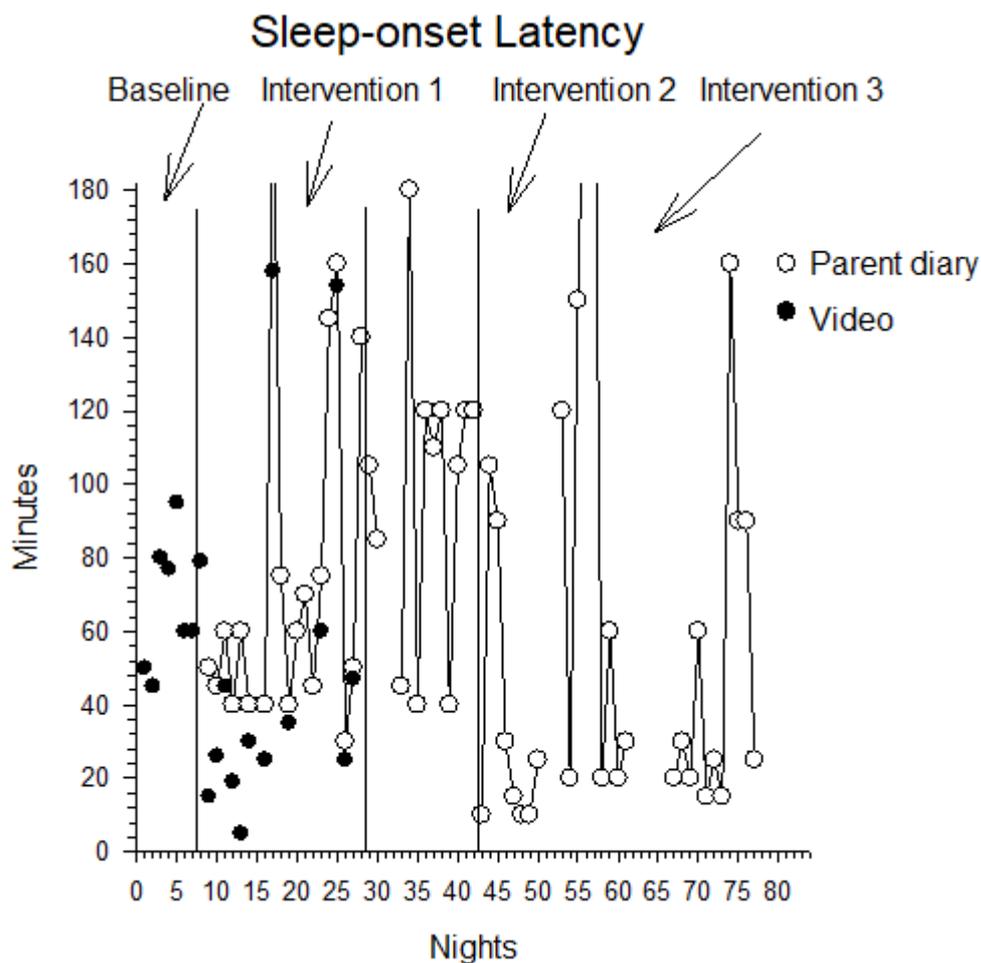


Figure 2. Sleep onset latency in minutes across baseline and intervention phases for Anna

**Effect on sleep onset latency.** Anna’s SOL was variable (between 45 and 90 minutes) and followed an increasing trend during baseline phase (Fig. 2). SOL for Anna during baseline, whether recorded by video or diary, fell outside the acceptable range of 15-

20 minutes. The first week of intervention phase 1 showed some initial improvement with SOL ranging between 5 and 60 minutes. During the remainder of intervention phase 1, Anna's SOL deteriorated and displayed high variability (between 40 and 180 minutes), which then settled into a pattern of approximately 120mins during intervention phase 2. The first week of intervention phase 3 showed some initial improvement before becoming highly variable. There was no evidence of a treatment effect. During intervention phases 2 and 3, her parents reported variable behaviour from Anna where she would either go to sleep within a short period of time or they reported it was very difficult for her to go to sleep. This is evidenced by the many nights (17) during intervention phases 2 and 3 where SOL was under 40 minutes, along with 10 nights where her SOL ranged from 60 to over 180 minutes (Fig. 2).

The rate of parental compliance with the intervention plan for Anna was high during intervention phase 1 (90%) but decreased markedly to only 7% during intervention phases 2 and 3. The sleep intervention for Anna was discontinued prematurely, as the research / therapy team were unable to gain agreement from her parents to adhere to intervention guidance.

## **Results – Jane**

**Sleep outcomes.** Jane's data on the frequency of curtain calls (CC's) and duration of sleep onset latency are shown in Figures 3 and 4.

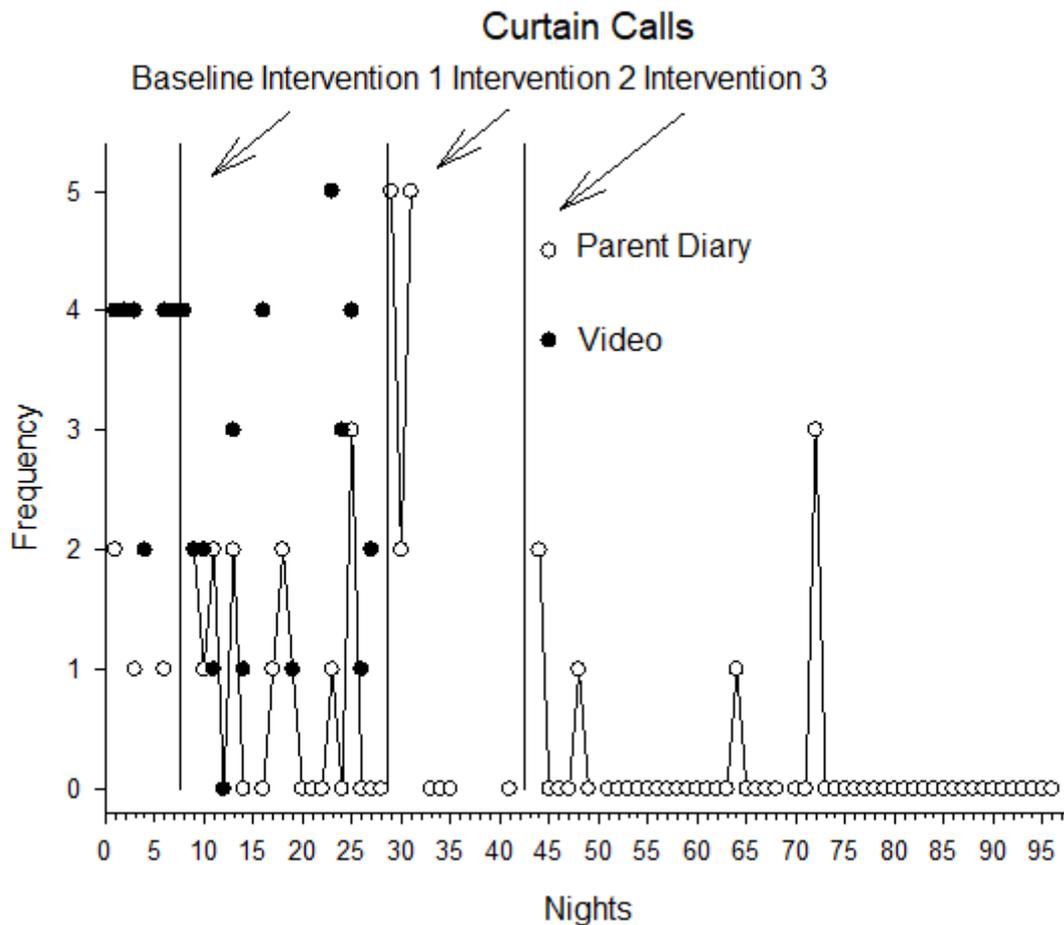


Figure 3. Frequency of curtain calls across baseline and intervention phases for Jane

**Effect on curtain calls.** During the baseline phase, the frequency of CC's for Jane was varied (Fig 3). There was a discrepancy between data sources, with CCs ranging from 0 to 4 according to video data and 0 to 2 according to parent diary data. During intervention phase 1 parent and video diary was again quite different, with parents reporting fewer curtain calls (between 0 and 3) compared with data captured via video (between 0 and 5). This may be due to lack of parent supervision once Jane had been put to bed. There was no evidence of a treatment effect during intervention phase 1. During intervention phase 2 there is limited data on CCs recorded for Jane as her parents' report that her sister Anna, who Jane shared a room with, was very disruptive and they were unable to record data. There was an overall reduction in CCs for Jane during intervention phase 3 (Fig.3), other than nights of parental

non-compliance with treatment plans. A large treatment effect was observed during intervention phase 3 with a PBM = 100%.

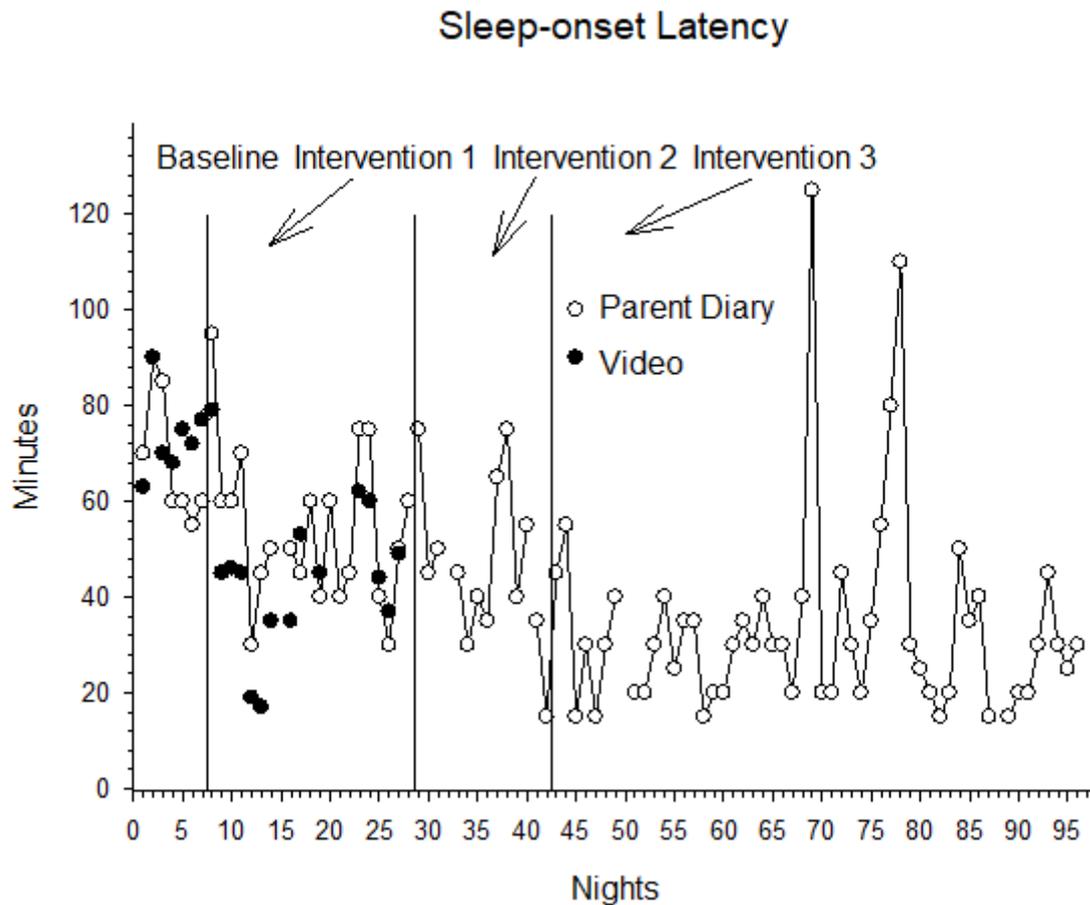


Figure 4. Sleep onset latency in minutes across baseline and intervention phases for Jane

**Effect on sleep onset latency.** During the baseline phase, there was a discrepancy between video and parent recorded data. Based on the video data, SOL for Jane was high (60 minutes and above) and following an increasing trend (Fig 4). A shift downwards in the level of SOL was observed during intervention phase 1, which became reasonably stable. A treatment effect was observed during intervention phase 1, with a PBM of 81%. During intervention phase 2 the new level of SOL for Jane was maintained. A further slight reduction in the level of SOL and an overall treatment effect was observed in intervention phase 3 (PBM=94%). However, the level of SOL for Jane was still considered to be above clinical threshold of 15-20 minutes, that has been used for many years in the Canterbury Autism and

Sleep project. There was also still a significant level of variability present, which was often associated with nights of parental non-compliance with treatment plans (Fig 4).

## Results – Colin

**Sleep outcomes.** As Colin was able to watch his PED in bed until he fell asleep, sleep onset latency for Colin has been referred to as duration of device use (DDU) and is shown in Figure 5. The data for frequency of night waking (represented by the line with data point) and duration of night wakings (represented by the columns) for Colin is shown in Figure 6.

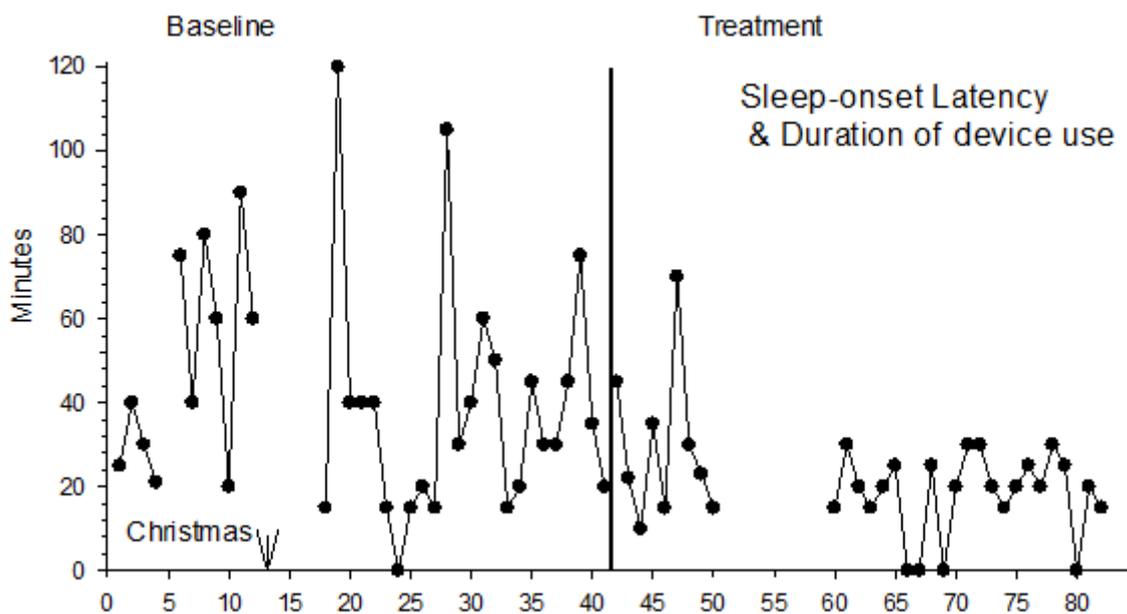


Figure 5. Duration of device use in minutes across baseline and treatment phases for Colin

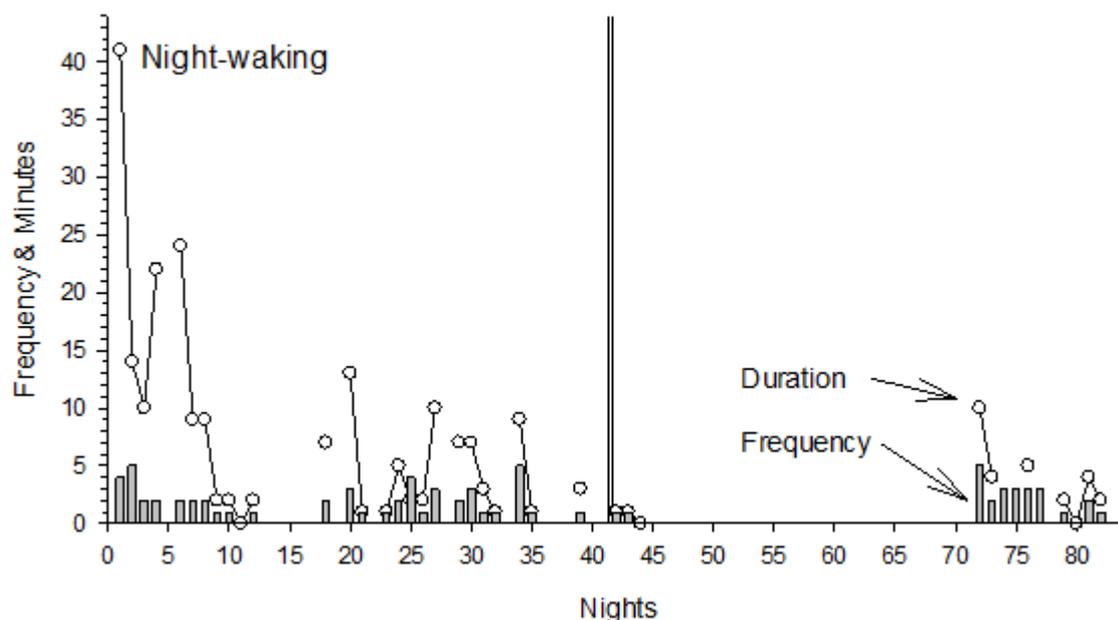


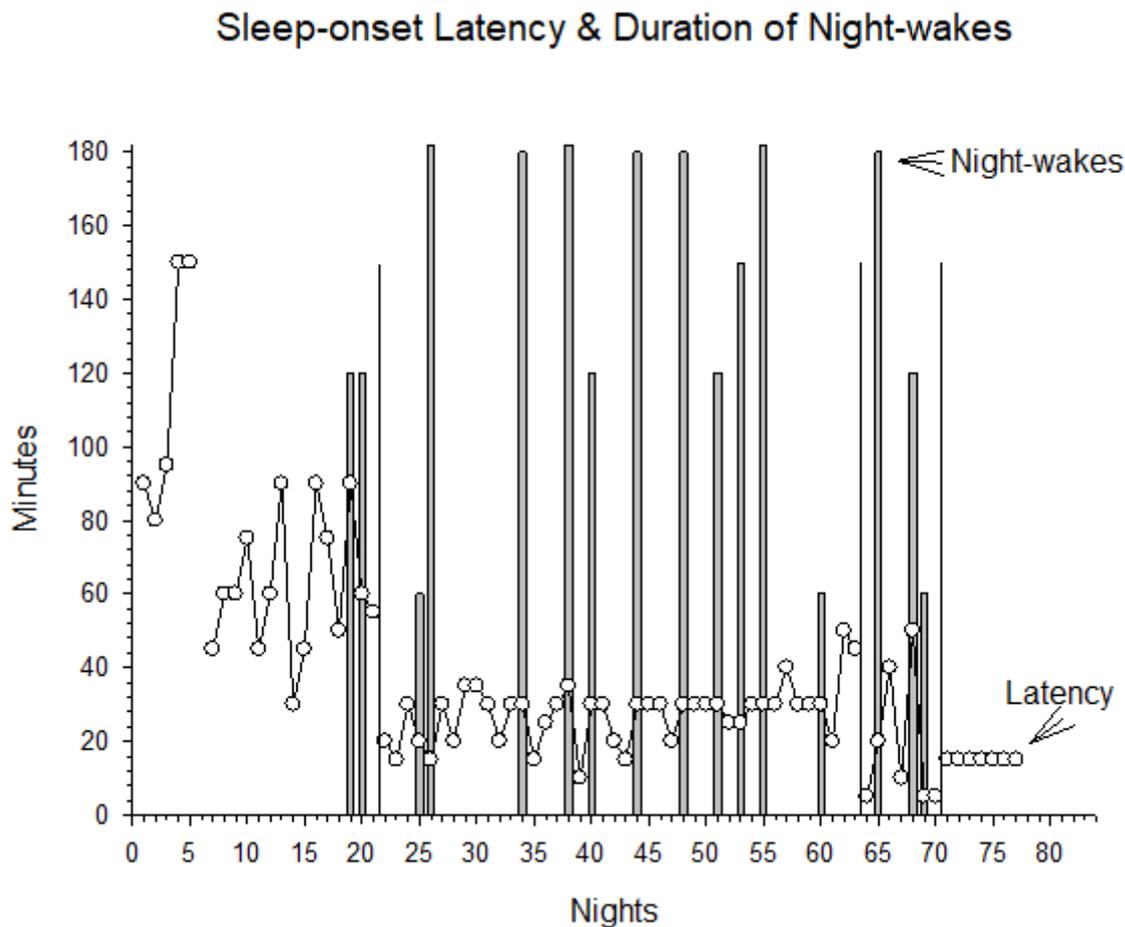
Figure 6. Frequency and duration of night wakings in minutes across baseline and treatment phases for Colin

**Effect on sleep onset latency/Duration of device use (DDU).** During the baseline phase, DDU for Colin was highly variable with few exceptions (between 0 and 120 minutes; see Fig 5). During the treatment phase, where Colin was now falling asleep in his bedroom rather than the lounge room, DDU was 30 minutes and under for most nights, showing an overall reduction from baseline (Fig 5). The new level of SOL during the second half of the treatment phase is on the threshold of being clinically acceptable (i.e., 20 mins or under; close to the clinical threshold adopted by the Canterbury Sleep and Autism project) and there is evidence of a large treatment effect (PBM = 93%; Ma, 2009).

**Effect on night waking.** During the baseline phase Colin was waking between 1 and 5 times per night (Fig 6). The duration of time awake for Colin associated with these night wakings post-Christmas is consistently under 15 minutes and quite variable. Colin’s parents reported little change in the duration or frequency of night wakings during the first week of treatment, after which, the family decided to stop treatment and leave the study due to challenging family circumstances.

## Results – Max

**Sleep outcomes.** Max's data for sleep onset latency (SOL) (represented by the line with data points) and duration of night wakings (represented by the vertical bars) is shown in Figure 7.



*Figure 7.* Sleep onset latency and duration of night wakings in minutes across baseline, intervention and follow up phases for Max. Duration of night wakings has only been included for nights where there was at least one night waking episode. There were zero night waking episodes on the nights where duration data has not been provided.

**Effect on sleep onset latency.** Max's SOL during baseline was highly variable (between 30 and 150 minutes; see Fig 7) and well above clinically acceptable levels used by the Canterbury Autism and sleep project (15-20 minutes). After the initiation of treatment an

immediate treatment effect was observed (PBM=100%), with Max's SOL showing an overall reduction and some nights achieving clinically acceptable levels. This improvement was improved upon at long term follow up with SOL recorded as 15 minutes for each night.

**Effect on night waking duration.** During the baseline phase, Max was waking on only approximately 10% of nights, and the duration of each waking was approximately 120 minutes (Fig 7). The frequency of night waking increased during the treatment phase to approximately 29% of nights, with the duration of these night waking episodes highly variable (60 -270 minutes). There was no evidence of a treatment effect for night waking. Night waking frequency and duration remained at a similar level at short term follow up, but had improved greatly at long term follow up, with no night waking episodes reported.

### **Comparison of Pre and Post Treatment Sleep Questionnaire Data**

**Child Sleep Habits Questionnaire (CSHQ).** Results of the CSHQ, a parent-report questionnaire relating to children's sleep patterns (Owens et al.,2000), are presented in Table 4. A higher score indicates more difficulties in that area, with total difficulty scores > 41 indicative of 'clinically significant' sleep disturbance (Owens et al. 2000). These results show that before intervention, Anna, Jane, Colin and Max were all rated by their parents as having 'clinically significant' sleep difficulties. Following intervention, parents total sleep difficulty ratings had increased for Anna (45 to 63) and Jane (41 to 43) and were over the cut-off for 'clinically significant' sleep difficulties. Max's total sleep difficulty score had reduced following intervention but was still indicative of 'clinically significant' sleep disturbance. Post treatment ratings were not available for Colin who did not complete the intervention.

## **Comparison of Pre and Post Treatment Questionnaire Data for Child Behaviour and Parent Wellbeing**

**Child Behaviour Checklist (CBCL).** The results of the CBCL at pre- and post-intervention are presented in Table 5. The CBCL identifies possible behavioural and emotional problems in children as rated by their parents. A higher score indicates more difficulties in that area.

Prior to intervention, Anna's parents scored her within the 'clinical' range for the internalising and total behaviour problem domains and within the 'normal' range for externalising behaviours. Following intervention, Anna's scores for internalising and total behaviour problems were still within the 'clinical' range and her score for externalising behaviours was now in the 'borderline' range. These results suggest no improvement in Anna's behaviour and that her externalising behaviour became slightly worse from pre- to post-treatment.

Prior to intervention, Jane's parents scored her within the 'normal' range for the internalising, externalising and total behaviour problems domains. Following intervention, all domains remained within the 'normal' range and reductions were seen in internalising behaviour and total problem behaviour domains, while externalising behaviour remained constant.

Prior to intervention, Max's parents scored him in the 'clinical' range for the internalising, externalising and problem behaviour domains. Following intervention reductions were seen across all domains with internalising behaviour scoring within the 'normal' range and externalising and total problem behaviour domains remaining in the 'clinical' range.

Colin's parents rated him to be in the 'clinical' range for each of the internalising behaviour, externalising behaviour and total behaviour problem domains. No post-treatment data was collected for Colin as his family withdrew from the study.

Table 4

*Comparison of Pre and Post Intervention Scores on the CSHQ*

Variable scores	Anna		Jane		Colin		Max	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Bedtime Resistance	7	<b>15</b>	6	6	<b>16</b>	N/A	<b>12</b>	<b>8</b>
Sleep Onset Latency	<b>3</b>	<b>2</b>	<b>2</b>	<b>2</b>	<b>2</b>	N/A	<b>3</b>	<b>3</b>
Sleep Duration	<b>5</b>	<b>6</b>	<b>5</b>	<b>4</b>	<b>6</b>	N/A	<b>6</b>	<b>7</b>
Sleep Anxxiety	<b>6</b>	<b>9</b>	4	4	<b>11</b>	N/A	<b>8</b>	<b>5</b>
Night Wakings	<b>4</b>	<b>5</b>	3	3	<b>8</b>	N/A	<b>4</b>	<b>6</b>
Parasomnias	<b>7</b>	8	7	8	<b>11</b>	N/A	<b>9</b>	<b>11</b>
Sleep Disordered Breathing	3	3	<b>4</b>	<b>4</b>	3	N/A	<b>4</b>	3
Daytime Sleepiness	<b>10</b>	<b>15</b>	<b>10</b>	<b>12</b>	<b>14</b>	N/A	<b>18</b>	<b>12</b>
Total Difficulties	<b>45</b>	<b>63</b>	<b>41</b>	<b>43</b>	<b>71</b>	N/A	<b>64</b>	<b>55</b>

*Note.* bold = above average scores / total difficulties score > 41 indicates clinically significant sleep disturbance; Owens et al. (2000)

Table 5

*Comparison of Pre and Post Intervention T-Scores on the CBCL*

Variable	Anna				Jane				Colin				Max			
	Pre		Post		Pre		Post		Pre		Post		Pre		Post	
	T-Score	R	T-Score	R	T-Score	R										
Internalising Behaviours	66	C	64	C	58	N	43	N	73	C	N/A	N/A	67	C	58	N
Externalising Behaviours	57	N	62	B	57	N	57	N	76	C	N/A	N/A	73	C	64	C
Total Score	67	C	65	C	58	N	52	N	83	C	N/A	N/A	75	C	70	C

*Note.* R=Range; N=Normal; B=Borderline; C=Clinical

## Parent Wellbeing Measures

**DASS-21.** The results of the DASS-21 for parent participants, are presented in Table 6. A higher score indicates a greater likelihood of psychological distress in the area of depression, anxiety or stress. At pre-treatment all scores were above the ‘normal’ range for both of Colin’s parents. Post-treatment scores were not collected for Colin’s parents as they withdrew from the study. For parents of children who completed intervention, the impacts on depression, anxiety and stress were variable. Jane and Anna’s father scored in the ‘normal’ range for all dimensions pre- and post-intervention. Their mother’s stress score had increased from 3 to 10, taking it over the ‘moderate’ symptom cut-off, her depression score had increased slightly from 1 to 2, however this was still within the ‘normal’ range and her anxiety score remained constant. For Max’s father, his scores had increased from pre- to post-treatment for all dimensions; depression from 5 to 13 (over the ‘severe’ symptom cut-off), stress from 8 to 12 (over the ‘moderate’ symptom cut-off) and anxiety from 0 to 3 (within the ‘normal’ range). Max’s mother’s scores had also increased from pre- to post-treatment and were over the ‘extremely severe’ symptom cut-off for anxiety (11 to 13) and stress (15 to 19). Her depression score decreased slightly from 16 to 15, however, was still over the ‘extremely severe’ symptom cut-off.

**PSQI.** The results of the PSQI for all parents are presented in Table 7. This gives an indication of each parent’s sleep quality pre-and post-intervention. Higher scores indicate worse sleep quality and lower scores indicate better sleep quality. Jane and Anna’s parents both indicated an improvement in their sleep post-intervention, with the Global PSQI score decreasing from 7 to 5 for their mother and 3 to 0 for their father. Max’s parents however, indicated a worsening of sleep quality post-treatment with the Global PSQI score increasing from 12 to 16 for his mother and 8 to 14 for his father. Post treatment scores were not collected for Colin’s parents as intervention was not completed.

**RQI.** Table 8 presents the results of the RQI for all parents. The global RQI score indicates the overall level of partner satisfaction for each individual, with a higher score indicating greater satisfaction. Both Jane and Anna's parents indicated an extremely high level of relationship satisfaction, scoring 45/45 at both pre- and post-intervention time points. Max's father indicated a notably higher level of partner satisfaction than his wife pre-intervention (32/45 and 18/45 respectively). At post-treatment both parents indicated an increase in their level of partner satisfaction, with Max's mother indicating a notable increase (from 18/45 to 26/45) compared to her husband (from 32/45 to 35/45). Colin's father indicated a higher level of partner satisfaction compared to his wife (31/45 and 29/45 respectively). Post-intervention comparisons were not made because of family circumstances at that time.

Table 6

*Comparison of Pre and Post Intervention Scores on the DASS-21*

Variable	Anna				Jane				Colin				Max			
	Mother		Father		Mother		Father		Mother		Father		Mother		Father	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Depression	1	2	0	0	1	2	0	0	<b>9</b>	N/A	<b>9</b>	N/A	<b>16</b>	<b>15</b>	<b>5</b>	<b>13</b>
Anxiety	1	1	0	0	1	1	0	0	<b>5</b>	N/A	<b>3</b>	N/A	<b>11</b>	<b>13</b>	0	<b>3</b>
Stress	3	<b>10</b>	0	0	3	<b>10</b>	0	0	<b>13</b>	N/A	<b>9</b>	N/A	<b>15</b>	<b>19</b>	<b>8</b>	<b>12</b>

Note. Bold = above the normal range

Table 7

*Comparison of Pre and Post Intervention Scores on the PSQI*

Variable	Anna				Jane				Colin				Max			
	Mother		Father		Mother		Father		Mother		Father		Mother		Father	
	Pre	Post														
Global PSQI Score	7	5	3	0	7	5	3	0	10	N/A	6	N/A	12	16	8	14

Note. higher scores indicate worse sleep quality – scores of 4 and below indicate good sleep quality

Table 8

*Comparison of Pre and Post Intervention Scores on the RQI*

Variable	Anna		Jane		Colin		Max							
	Mother		Father		Mother		Father							
	Pre	Post	Pre	Post	Pre	Post	Pre	Post						
Global RQI	45	45	45	45	45	45	29	N/A	31	N/A	18	26	32	35

*Note.* higher scores indicate better relationship satisfaction

Table 9

*Post-Intervention Treatment Acceptability Scores from TARF-R*

Variable scores	Anna		Jane		Colin		Max - incomplete	
	Mother	Father	Mother	Father	Mother	Father	Mother	Father
<b>Total Acceptability</b>	<b>74</b>	<b>106</b>	<b>74</b>	<b>106</b>	<b>98</b>	<b>N/A</b>	<b>N/A</b>	<b>N/A</b>
Reasonableness	12	20	12	20	21	N/A	N/A	N/A
Willingness	14	18	14	18	15	N/A	N/A	N/A
Effectiveness	16	12	16	12	20	N/A	N/A	N/A
Cost	14	14	14	14	14	N/A	N/A	N/A
Negative Side-Effects	6	21	6	21	15	N/A	N/A	N/A
Disruption/Time	12	21	12	21	8	N/A	N/A	N/A
Problem Severity	10	8	10	8	10	N/A	N/A	N/A
Understanding of Treatment	5	7	5	7	7	N/A	N/A	N/A

## **Social Validity and Treatment Acceptability**

**TARF-R.** The TARF-R scores have a possible range of 17 to 119, with higher scores indicating higher acceptability. Parent ratings ranged from 74 to 106 (see Table 9). Anna and Jane's parents noted that it was difficult for them to rate the overall acceptability of the treatment, as in their view, the intervention had proven effective for one of their children (Jane) but not the other (Anna).

## **Post-Treatment Discussions**

During post-treatment interviews parental report, suggested that the components implemented as part of an FBA-informed sleep intervention were generally acceptable and well received by parents. Parental report regarding effectiveness varied by participant. The perception of Anna, Jane and Colin's parents was that just changing the content on the PED and allowing the child to take this to bed, had been initially helpful for minimising distress and taking the first intervention steps, without removing the device completely.

Anna and Jane's mother reported that overall, she had learnt a lot and knows what strategies will work to improve her children's sleep going into the future. This is consistent with previous research which suggests that parental acquisition of new knowledge and understanding is a crucial step to progress being made in improving their child's sleep (Beresford et al., 2016). She reported that allowing Anna and Jane to take their PED with revised content to bed had represented a 'path of least resistance' and had helped the family to take the first step in changing requirements around bedtime and sleep. She believed this had worked particularly well for Jane who was younger and better able to accept the change, but not for Anna who she believed was older and therefore more resistant to change.

She reported feeling supported during the intervention, which increased her confidence to follow things through. She believed that a consistent bedtime routine and

ensuring the children had enough time to ‘wind down’ before bed was important to intervention success. She reported that the intervention was effective for Jane and that she was sleeping more hours overall each night. She believed that the additional sleep had a positive impact on her daytime behaviour, including being more relaxed and helpful. Jane’s mother also reported noticing an improvement in Jane’s communication ability and energy levels. In terms of positive impacts for the family, she reported that herself and her husband were now able to have less disrupted time in the evenings.

Anna and Jane’s mother reported the intervention initially looked like it was going to work for Anna too, but believed that Anna did not like the changes and gave up. She reported several perceived barriers to treatment including that Anna:

- was older and more set in her ways, which made change more difficult
- had stomach issues which impacted on the outcome
- was more stimulated from reading a book compared to using a PED, where her mind was maybe less active, thus facilitating relaxation
- was resistant to entering her bedroom as she did not like the presence of the video camera in her room

Anna and Jane’s mother also highlighted the challenge of having multiple children in her house with ASD and that parents in such households ‘don’t have the energy to fight’. She also noted that interventions such as these require more effort when you have ‘challenging children’.

Despite leaving the study prematurely, Colin’s mother was able to share her impression of the treatment approach. Colin’s mother reported that the intervention worked really well for their family, as taking the PED to bed (with revised content) had allowed a

smooth and distress free transition, from Colin falling asleep in the lounge room to him falling asleep in his own bed without parental presence. She indicated that managing her personal stressors with no support was a barrier to maintaining changes implemented during intervention phase 1, as well as continued participation in the sleep study, and that Colin was also impacted by the stress of these events and has since co-slept with his mother.

## Chapter 4. Discussion

### Research Questions

The current study was driven by the following five research aims: 1) to examine the effect of using an age and developmentally appropriate bedtime story presented on a PED, to improve sleep in children on the autism spectrum; 2) to examine the effect of using a Video Self-Model Social Sleep Story presented on a PED, to improve sleep in children on the autism spectrum; 3) to evaluate parent's perceptions of the treatment approach using PEDs; 4) to examine the effect of a FBA informed individualised intervention to improve sleep in children on the autism spectrum; 5) to examine the effect of improving child sleep on parent and child wellbeing.

### Effectiveness of the PED Interventions

In the current study, three participants (Anna, Jane and Colin) received an intervention using a PED. They all received an intervention using an age and developmentally appropriate bedtime story and after this Anna and Jane also received a video self-modelled social story. The effectiveness of these interventions will be discussed for each child below.

**Jane.** For Jane, the bedtime story PED intervention resulted in an improvement in sleep onset delay, which became relatively stable. This improvement was maintained during the video self-model PED intervention; however, no additional improvement was evident. The bedtime story PED intervention resulted in no improvement in curtain calls for Jane and there was very limited data available to assess the video self-model PED intervention, as the parents reported that Jane's sister Anna (who shared the same room) was very disruptive during this phase and they were unable to record data. There is some evidence to suggest that

restricting the content on the PED to be less stimulating and reinforcing had the desired effect, contributing to one of the goals for this family, that is, to reduce device use at bedtime. During the video self-modelled social story phase, Jane took the PED to bed and watched this for the first four nights, after which she elected not to take the PED to bed as she described the content as 'boring'. Factors which added to the complexity of this sleep intervention for Jane, included the need to share a room with her sibling and her parent's report that their attention was directed elsewhere during part of this intervention to address disruptive behaviour from her sibling.

**Anna.** For Anna, the bedtime story and video self-model PED interventions resulted in no sustained improvement for curtain calls or sleep onset delay, which remained highly variable. Anna's parents reported an oscillating pattern of behaviour, where Anna would either have a good night and go to sleep quite easily, or have a bad night and refuse to go to bed at all, often staying up past when her parents went to bed. Early in the intervention, Anna had also long elected not to take the PED to bed as she was not interested in the content. While this had the desired effect of reducing reinforcement for using the PED, it also meant that Anna had limited exposure to the video self-model social story, presented on the PED.

**Colin.** For Colin, the PED intervention was supported by a behavioural intervention which included a new bedtime routine, a paper-based social story to help him understand routine changes and expectations around sleep and withdrawal of parental presence. Prior to intervention, Colin was able to watch his preferred content on his PED in the lounge room until he fell asleep. The priority for Colin's family was for him to initiate sleep alone in his own bedroom. Therefore, instead of sleep onset delay, the time Colin viewed restricted content on the PED until he fell asleep was referred to as duration of device use. For Colin, the bedtime story PED intervention resulted in an overall improvement in duration of device use, reaching clinically acceptable levels when assessed using sleep onset delay data. A

secondary behavioural intervention was designed to address Colin's night waking, which is discussed in a later section.

### **Parents' Perceptions of the PED Interventions**

Prior to treatment, parents in the study reported common themes regarding why they used PEDs as part of their children's bedtime routine. These themes included, facilitating the transition from evening activities to bedtime, reducing conflict with siblings and parents, creating a calm environment before sleep, and allowing parents more time for relaxation in the evening. All parents, however, expressed a desire to reduce PED use at bedtime.

Overall, parent perceptions of the interventions using a PED was positive. Anna and Jane's parents reported that they represented an acceptable intervention, in that it met the children halfway in terms of their desire to take a PED to bed with them. They found the intervention easy to implement, without causing distress to the children which was important to them. Colin's parents were also positive regarding how taking the PED to bed had facilitated transitioning Colin's sleep onset location from the loungeroom to his bedroom, without causing him distress. As a result of implementing the intervention, all parents also reported no perceived risk of harm or side effects. These findings support previous research that has found that parents are more likely to use intervention techniques if they are not too burdensome and fit with the family's values (Koegal et al, 2003).

### **Effectiveness of Secondary FBA-Informed Behavioural Intervention**

All participants who had completed the PED interventions were identified as requiring further behavioural intervention. Only Jane went on to receive a full secondary behavioural intervention. Anna's intervention ended early due to parental noncompliance and Colin's mother decided to withdraw from the study only a few days into this phase. Max's

device use at bedtime was addressed as part of a multi-component behavioural intervention. Responses to the behavioural interventions are discussed below for each child.

**Max.** Along with the change of Max's device use at bedtime from TV to audio (music) only, he received a multi-component behavioural intervention comprising parental presence (Sadeh, 1994), with planned ignoring and positive reinforcement. This combined intervention resulted in the resolution of sleep onset difficulties for Max, with improvements maintained at short term and long term follow up. Night waking problems were not resolved for Max during intervention or short-term follow-up, however, they were resolved at long term follow up. It should be noted that the duration of long-term follow-up was only seven days and may have sampled an atypical week. Furthermore, due to the gap between the end of intervention and long term follow up, conclusions are unable to be made regarding the factors contributing to the resolution of night waking problems for Max.

**Jane and Anna.** Within the same household, Anna and Jane received a further behavioural intervention including a staggered bedtime routine (as they shared the same room), that included the introduction of a bedtime story with a parent for Jane (the youngest) and reading time in bed for Anna. The intervention also included a set time each night for when device time ended, which was at least 30 minutes prior to commencing the bedtime routine. For Jane, this intervention resulted in the near resolution of CC issues with zero curtain calls recorded for 49 out of 53 nights. Of the four nights where curtain calls were recorded for Jane, her parents had not complied with the treatment plan, either putting her sister Anna to bed in the same room early or missing out the bedtime story component. The behavioural intervention also resulted in an overall improvement in Jane's SOD, however, the level of SOD was still considered to be above clinically desirable levels. There was also a significant level of variability present for Jane's SOD, which was often associated with parental non-compliance with the treatment plan.

For Anna, the secondary behavioural intervention resulted in no overall improvement in CCs or SOD. She continued to display oscillation between 'good' and 'bad nights, as described above. It is possible that the nights where she was able to fall asleep quickly was due to an increased level of sleep pressure from staying up very late the night before.

It is impossible, however, to draw conclusions about the effectiveness of the behavioural intervention for Anna as the rate of parental compliance with the treatment plan was very low, approximately 7%. There was also inconsistency between her parents in terms of following the treatment plan. Anna's mother would ensure that her sister Jane was asleep before putting Anna to bed in the same room, whereas, Anna's father would regularly attempt to put Anna to bed before her agreed bedtime and when Jane was still awake. Anna's parents would also regularly give in to her requests to sleep on a mattress in their room or for one of her parents to sleep on a mattress in her room.

The consistent deviations from the treatment plan, resulted in the maintenance of sleep interfering behaviours for Anna through a lack of appropriate stimulus control and differential reinforcement for sleep facilitative behaviour. The research / therapy team had offered Anna's parents assistance and support to encourage adherence to the treatment plan, but the decision was eventually made by the research / therapy team to cease intervention for Anna. Anna's parents reported that they were unable to follow through on the limits required to extinguish the sleep interfering behaviour for Anna and agreed with the decision to cease intervention. Reviewing the case, the research / therapy team report that they should have halted Anna's intervention earlier, given the reluctance of her parents to receive assistance.

**Colin.** To address Colin's night waking and eliminate bed-sharing, a secondary behavioural intervention phase was implemented. This included settling him in his own bed and gradually removing parental presence when he woke during the night. After the first

week of treatment there was little to no improvement for Colin, however, his mother reported only be able to follow through with the treatment plan on four out of ten nights. Colin's family then decided to stop treatment and leave the study, due to family circumstances which were likely to have been stressful for Colin.

**CSHQ.** Despite the results regarding improvement in SOL and CCs for Jane, her parent-rated post-treatment sleep disturbance score on the CSHQ was still within the 'clinically significant' range. It is possible that the changes to Jane's sleep had not been in place long enough to be reflected in parent overall ratings.

Despite the resolution of sleep onset difficulties for Max, his parents CSHQ rating of his sleep disturbance was still within the 'clinically significant' range, although it had dropped a little. This might be explained by the persistence of Max's night waking problems post intervention and at short term follow-up.

### **Impact on Children and Parents**

The intervention had a varied impact on child behaviour and parent wellbeing across participants. There is no data available for Colin and his family who withdrew from the study. The resolution of sleep onset difficulties for Max may have contributed to a significant reduction in internalising behaviour, which shifted from the 'clinical' to 'normal' range of the parent reported CBCL post-treatment. Reductions in externalising behaviour was also reported, although his CBCL externalising behaviour and total score remained in the 'clinical' range. Some reductions in internalising behaviour was also seen for Jane, although all her behaviour ratings post-treatment remained in the 'normal' range. Post-treatment data for Anna, who gained no improvement through the intervention, showed an increase in externalising behaviours shifting from the 'normal' to 'borderline' range on the CBCL. It is possible that the stress of the intervention, which was characterised by a high level of

resistance from Anna, combined with prolonged sleep difficulties contributed to this increase in externalising behaviours.

The impact of the intervention on parent wellbeing was also varied across participants. Max's parents indicated worsening sleep quality post-treatment, which might be partly explained by Max's prolonged night waking difficulties, that remained unresolved post-treatment. Jane and Anna's parents endorsed an improvement in sleep quality, that was already quite high pre-treatment. This might be explained by the scarcity of night waking difficulties for Jane and Anna.

In term of relationship quality, Max's parents both reported an increase in satisfaction post-treatment. This may have been influenced in part by the resolution of Max's sleep onset difficulties, which possibly gave them more time together in the evening and the active role that Max's father also played during the intervention. Jane and Anna's parents reported no change in their relationship satisfaction, although it should be noted that they both rated this a perfect score (45/45) at pre-treatment, meaning there was no scope for improvement at post-treatment (a ceiling effect).

In reference to parental mental health as part of well being, there was also variation across participants. At post-intervention for Jane and Anna's parents, the only change was an increase in stress levels for their mother, although her score only took her within the 'moderate' symptom range. All other ratings on depression, anxiety and stress remained the same for Jane and Anna's parents and within the normal range, suggesting a relatively low level of mental health distress for these parents. In comparison, depression, anxiety, and stress ratings had all increased for both Max's parents, with many of the ratings in the 'severe' symptom range. The lack of resolution of Max's night waking difficulties may have in part contributed to this increase.

No significant reduction in mental health distress for parents was evident in the present study. It is likely that the commitment required in implementing in-home behavioural interventions can be challenging and stressful for some parents (Eikeseth, Klintwall, Hayward & Gale, 2015). Longer term data may therefore be required to assess the impact to parent wellbeing after sustained sleep improvements.

### **Overall Intervention Effectiveness**

Overall, the findings from these case studies adds to research that has identified PED use as a factor, which frequently contributes to sleep disturbance for children on the autism spectrum (Mazurek et al., 2016; Mazurek & Engelhardt, 2013; Mazurek & Wenstrup, 2013; Van Deurs et al., 2019). Jane, Colin and Max's case study findings also contribute to previous research that has found that passive content may be less detrimental to sleep than interactive content (Cain & Gradisar, 2010). The findings for Jane and Colin suggest that using a PED at bedtime with less stimulating content, while not being able to completely resolve sleep problems for children with ASD, may represent a useful stepping stone for families in facilitating change, while reducing child resistance and distress and being manageable for families to implement.

The findings for Jane and Max, support the use of a FBA-informed behavioural intervention to improve sleep in children on the autism spectrum. This is consistent with previous research that demonstrates the effectiveness of such interventions delivered in a naturalistic setting, such as the home (Jin et al., 2013; McLay et al., 2017; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019; Moore, 2004; van Deurs et al., 2019). A comprehensive FBA-informed approach is considered to be more effective, as the variables precipitating and maintaining behaviour for each child drive the choice of intervention (McLay et al., 2017; McLay et al., 2020).

## **Factors Impacting on Intervention Delivery and Results**

There were a range of child and family factors that were observed to increase the level of complexity present during the intervention. Each case study provided some interesting clinical observations relating to child and family complexity factors and how such factors can impact the delivery and results of parent facilitated interventions for children on the autism spectrum. To protect the privacy of the families, these observations are discussed without reference to a particular family or participant.

**Family factors.** Previous research has highlighted that the added complexity faced by parents of children with neurodevelopmental disorders including ASD, may impact on their ability to prioritise a sleep intervention and consistently focus their energy and attention on improving their child's sleep (Beresford et al., 2016). One participant family were also responsible for other siblings with neurodevelopmental and sleep difficulties. Consistent with previous research, parents reported that the responsibility for caring for multiple children with additional needs had impacted their level of energy available to consistently record sleep diary data and comply with treatment plan instructions. They noted that parents of families with only one child with additional needs might be able to follow through on the changes, but for a family with multiple children with neurodevelopmental and sleep problems, they reported not having the energy to fight, when any child resistance was encountered.

Factors including an increased frequency of child illness and an inability for full time school attendance had increased the childcare demands for two families in the study. This had prevented a desired return to paid employment for both mothers. This may have impacted their overall capacity to adhere to treatment requirements, which is in line with previous research findings that an inability to return to work can diminish parent's financial, social and emotional support resources (Karst & van Hecke, 2012).

Some environmental complexity factors were also observed during the study. Two study participants shared a bedroom with a sibling for the duration of the study and this appeared to add further complexity to the sleep interventions. One factor identified as maintaining sleep difficulties for both children included social attention from their sibling. Parents were often unaware of the social interaction between the children in the bedroom, which was captured via video. Another participant's progress during intervention was impacted by the noise level in their household, close to their bedroom. These observations are consistent with previous research that has identified a lack of sleeping environment options, such as sibling room sharing, as a significant barrier to parent led sleep interventions (Beresford et al., 2016).

Inconsistency between parents in terms of implementing the treatment plan was also observed as adding complexity to the sleep intervention. This is consistent with previous research which has identified that inconsistency between parents, or other caregivers undermining the intervention, as factors that increase the difficulty of implementing new sleep management strategies (Tse & Hall, 2007). A lack of support from one parent during the intervention, also reportedly reduced the capacity of the other parent to adhere to the intervention requirements, due to the increased stress created by a lack of support from their partner.

After seeing initial improvement as a result of the sleep intervention, the level of complexity greatly increased for one family, involving circumstances that were likely to have been stressful for the child. This resulted in the family withdrawing from the study and the gains of the first phase of intervention being lost, with the child reverting to pre-intervention sleep patterns. This outcome is consistent with previous research, that has identified parental relationship quality as a factor that may impact sleep treatment outcomes (Chow, 2019; Mindell et al., 2006). This outcome also suggests that, even if treatments have shown

effectiveness for children, that interventions can put so much strain on parents that if the parent believes they cannot withstand the impact on their mental health, that child gains are not maintained or parents withdraw from treatment (Karst & van Hecke, 2012).

Parental perceived barriers associated with treatment participation, is another factor found to be significantly associated with less therapeutic change compared with families perceiving fewer barriers (Kazdin & Whitely, 2006). In the current study, parents reported several perceived barriers to participation in treatment. Child resistance to change during intervention was often perceived by parents as a barrier to treatment participation. A range of factors were perceived by parents to be driving the resistance from the child, including regular stomach upsets, the older age of the child, bedroom fears and concerns regarding the presence of the video camera. Parents would regularly deviate from the treatment plan to accommodate these factors. A strong belief from parents for one child that they were more stimulated from reading a book compared to watching something on a PED, represented a perceived barrier to full treatment participation for this family. Child distress was another form of child resistance that was identified as another perceived barrier to treatment participation. Some parents were unwilling to tolerate any level of child distress during intervention, such as that triggered by setting boundaries and limits around bedtime as a part of the treatment plan. There was less therapeutic change for children of parents with more perceived barriers and less treatment adherence in this study, which is consistent with some research in this area (Kazdin & Whitely, 2006). The implication for clinicians is that parents may need a great deal of support to implement a FBA-informed treatment plan.

In this study some parents who reported mental health difficulties were able to follow through with treatment, which resulted in improved sleep for their child. On the other hand, other families with reported parental mental health difficulties struggled to adhere to treatment requirements and this appeared to impact sleep improvement and treatment

completion. Autism traits in one or both parents, which has been found in one recent study to predict sleep disturbance in children with ASD, was also present for one family within the study, which may have impacted on the level of treatment adherence and sleep outcomes for this child (Waddington et al., 2020).

Another interesting observation is that the participant who appeared to have relatively less complexity present, appeared to gain the most improvement in sleep. This family had no additional siblings with mental health or sleep difficulties and both parents remained committed to the intervention, with the father also playing a particularly active role compared to other families.

**Child factors.** Also present for a participant in this study were two of the most commonly occurring psychological conditions for children on the autism spectrum, namely ADHD and anxiety, which have also been associated with sleep difficulties (Hollway et al., 2013; Mayes & Calhoun, 2009; Mazurek & Petroski, 2014). Apart from a possible direct amplification of this child's sleep difficulties, the presence of comorbidities may have also had a secondary impact in terms of their parent's capacity to follow through on intervention requirements. This would be consistent with previous research which has found that for children with ASD, the severity of comorbid ADHD also predicted parenting stress and coping which, in turn, impacted on parent's ability to engage in interventions (Manohar et al., 2018).

**Complexity conclusions.** Many of the complexity factors appeared to impact the level of treatment adherence and treatment outcomes in this study. Indeed, for the child with the most family and child complexity factors, this intervention, which was characterised by a lack of parental treatment adherence, was not successful in improving sleep for this child. In contrast, improved sleep was achieved for the child with the least complexity present.

Interestingly, the sleep intervention was relatively successful for one child who was part of an environment with many family complexities. This success may in part be due to relatively fewer child-related complexity factors present in this case. This outcome suggests that a sleep intervention can be successfully delivered and achieve results, despite the presence of multiple complexity factors. This is consistent with previous research involving children with oppositional defiant disorder and conduct disorder, which found that case complexity and comorbidity did not necessarily influence treatment outcomes (Kazdin & Whitely, 2006). It is also consistent with some recent Master's thesis research on the efficacy of FBA-informed sleep interventions for children on the autism spectrum, which found that child complexity was not a deterrent to treatment response (Ng, 2019).

The varied outcomes across case participants in this study, highlight the importance for clinicians to be aware of the limitations involved in working with families with a high of case complexity. It also highlights, however, that positive outcomes can be achieved despite the presence of complexity.

**Medication.** The variation in use of medication was one factor that potentially impacted on the results for the participants. Three out of the four participants had been prescribed and were taking melatonin or Phenergan prior to the commencement of the study. Despite the research / therapy team requesting that the use of melatonin remain consistent throughout the study, parents reported a wide variation in melatonin use, providing it to their child on some nights but not on others. As an example, one child had not been prescribed melatonin prior to or during the study, however, her mother reported giving her a dose of her sibling's melatonin if they asked for it. Another parent also reported that they perceived melatonin to be detrimental to their child's daytime behaviours, so would avoid providing it, unless the child asked for it. Varied use of melatonin for participants throughout the study, may have impacted on the ability to draw valid and accurate conclusions from results.

## **Limitations and Recommendations for Future Research**

Limiting the generalisability and external validity of the study is the small number of participants with heterogeneous presentations of ASD and sleep problems, along with the presence of varied complexity factors. It should be noted, however, that it is expected that case study research will have limited generality.

Replication with robust methodological design and larger samples/more replications, is essential to determine the contribution of a PED component as a stand-alone intervention and as part of a larger behavioural intervention. Only a qualitative discussion of complexity, in the form of clinical observations, was included in the study, with no attempt made to quantify the level of complexity factors present for each child. Although one attempt has been made previously to operationalise complexity, there is currently no standard measure of case complexity (Kazdin & Whitely, 2006). Further research is required to establish measures of complexity and determine the impact of family and child related complexity factors on sleep outcomes, for interventions involving children on the autism spectrum.

Another factor limiting the reliability and validity of results in this study was the inability of some parents to regularly and comprehensively complete sleep diaries, which resulted in missing or incomplete data for participants. Consistent with previous research, some parents, particularly those who were already overwhelmed by the demands of bedtime, found the completion of sleep diaries laborious and were therefore unable to capture required data (Moore et al., 2017). In other cases, video-recorded data suggested that parents were unaware of child sleep and wake times and possibly misinterpreted silence as sleep. Even when video data was consistently recorded, it was limited by any time a child moved their sleep location during the night (e.g., to their parent's room) and parents forgetting to initiate video recording (Moore et al., 2017). Overall, there was limited availability of video data in

the current research, which meant that reliability of parent sleep diary data could not always be verified.

While it is well recognised that there are differences in sleep characteristics for TD children and children on the autism spectrum, data based on TD has been cited in the current study due to the lack of normative sleep data available for children on the autism spectrum. This was also a limitation of the sleep and behaviour-related questionnaires used in the study, which have not been developed with norms for children on the autism spectrum (Moore et al., 2017).

### **Clinical Implications**

In terms of the clinical application of sleep interventions, especially those informed by FBA, it is important to consider the presence of complexities and the unique needs of children on the autism spectrum and their families. To assist in shifting barriers to treatment and preventing parent burnout, interventions should be tailored not only the varied characteristics of the child but also the competencies and preferences of the parents and family (Vriend et al., 2011). The families in the current study, reported using PEDs to manage behaviour and encourage a calm bedtime transition for their child, which aligns with similar themes identified in previous research (Mazurek et al., 2016; Nally et al., 2000). In cases where PED use is identified as a primary sleep-interfering behaviour, clinicians might consider incorporating a component into their treatment planning which utilises a PED with less stimulating content. Future research could explore the balance between ‘boring’ PED content, which had the desirable reinforcement from use, against sufficient reward to maintain use, if use is required as part of the intervention, such as the video self-model social story in this study. While a PED option may not completely resolve sleep issues, it may offer

a manageable path of least resistance to implementing changes around bedtime and sleep for parents and children on the autism spectrum.

In order to increase treatment effectiveness and minimise cases of non-responders it is important to have an awareness and understanding of what specific child and familial complexities need to be considered and addressed in treatment planning (Vriend et al., 2011; Webster-Stratton, 1985). In cases where there are family complexity factors present and noted at initial presentation, especially relationship discord or a discrepancy in the level of parent commitment to improving their child's sleep, the therapist may need to provide additional education regarding the importance of creating a favourable environment, to facilitate the success of the intervention. To improve the rate of adherence to treatment plans, additional components may be needed to address child behavioural problems, including parent education and training regarding the importance of enforcing limits and boundaries during intervention. It is also important for clinicians to seek clarity on best practice methods to support the delivery of the treatment program.

## **Conclusion**

Despite the limitations, the current research makes a number of important contributions. Firstly, it adds to the emerging research base recognising the possible benefits of using a PED to facilitate behaviour change with children on the autism spectrum (Flores et al., 2014; Goldsmith & LeBlanc, 2004; Kuoch & Miranda, 2013), and to our knowledge it is the first study to use a PED as part of a sleep intervention. The findings suggest that using a PED with less stimulating content was perceived well by parents and may show some promise as a path of least resistance for initiating change around bedtime and sleep for children on the autism spectrum. An intervention component using a PED might be able to compliment a wider behavioural intervention, especially in cases where children and parents

are reliant on PED use around bedtime. The use of PEDs to facilitate video self-modelling and present social stories worked well together in this study.

Secondly, the study contributes to a growing literature base that has effectively used a FBA process to inform sleep intervention plans for children on the autism spectrum (Jin et al., 2013; McLay et al., 2017; McLay et al., 2019; Van Deurs et al., 2019). Thirdly, it adds to the discussion around child and family complexity factors and how they can impact treatment adherence and outcomes in sleep interventions for children on the autism spectrum. It also highlights, however, that positive outcomes can be achieved despite the presence of complexity. Finally, the findings indicate that improvements in child sleep does not necessarily lead to measurable improvement in child behaviour and parental wellbeing, which highlights the complexity of these associations. Future research incorporating robust methodological designs and larger samples/replications could focus on determining the contribution of a PED component as a stand-alone intervention and as part of a larger behavioural sleep intervention. Additional research is also required to establish standardised measures of complexity and determine the impact of family and child related complexity factors on sleep outcomes, for interventions involving children on the autism spectrum.

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## Appendices

### Appendix A. Ethics Approval



HUMAN ETHICS COMMITTEE

Secretary, Rebecca Robinson  
Telephone: +64 03 369 4588, Extn 94588  
Email: [human-ethics@canterbury.ac.nz](mailto:human-ethics@canterbury.ac.nz)

Ref: HEC 2018/47

23 July 2018

Dr Laurie McLay  
Health Sciences  
UNIVERSITY OF CANTERBURY

Dear Laurie

The Human Ethics Committee advises that your research proposal "An Investigation into the Effectiveness of Treatments for Sleep Disturbance in Children With Autism" has been considered and approved.

Please note that this approval is subject to the incorporation of the amendments you have provided in your email of 16<sup>th</sup> July 2018.

Best wishes for your project.

Yours sincerely

*pp. R. Robinson*

Professor Jane Maidment  
**Chair**  
*University of Canterbury Human Ethics Committee*

## Appendix B. Children's Information Sheet



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

#### Children's Information Sheet



Hello. My name is XXX and I am a teacher/student/psychologist at the University of Canterbury. I am doing a project about how to help children to 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 to sleep better. This means that I might be Skyping you, coming to your house, or your parent/s will be coming to see me at the University.

There will 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. XX, a research assistant/Masters/PhD 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.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email [human-ethics@canterbury.ac.nz](mailto:human-ethics@canterbury.ac.nz)

If you do not want to be a part of this project, you can tell me or your parents 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.

Now we need to decide if you would like to do this. If you do want to be a part of my project then you can say "yes". If you do not want to be a part of this project then you can say "no" and no one will mind.

If you say yes, you or one of your parents can sign the form for you.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email [human-ethics@canterbury.ac.nz](mailto:human-ethics@canterbury.ac.nz)

## Appendix C. Children's Consent Form



### **“An investigation into the efficacy of treatments for sleep disturbance in children with autism”**

#### **Children's Consent Form**

My name is \_\_\_\_\_.

- XXX has told me about the work that she is going to be doing with me and my parent/s.
- XXX told me that she is going to be working with me and my parent/s to help me to learn to sleep better.
- While XXX does this she will be asking my parents about my sleep each night and there will be a video camera in my room on some nights that is recording my sleep.
- I know that if at any time I want to stop being a part of this project then XXX will stop recording data and this will be destroyed.
- If I want XX to stop video recording my sleep then the camera will be taken out of my room and that will be fine. If I want any video footage to be deleted, I can tell XXX or my parents.
- I was told that my parents/caregiver may sign this form for me and I think that is OK.
- I would like a summary of the results of this project.

Child's name: \_\_\_\_\_

Date: \_\_\_\_\_

Signature: \_\_\_\_\_

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

Parent/caregiver: \_\_\_\_\_

Date: \_\_\_\_\_

Signature: \_\_\_\_\_

***Please return this form to XXX.***

## Appendix D. Child Assent Form

### Children's Assent Form



The project that Laurie 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 Laurie, Jenna, or my parents.

- I am happy to be a part of the project and for Laurie 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 Laurie now.

## Appendix E. Parent Information Sheet



### **An investigation into the effectiveness of treatments for sleep disturbance in children with autism or features of autism**

#### **Information for Parents/Caregivers**

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

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 Child and Family Intern psychologists or registered psychologist also work on this project.

We would like you and your child with autism 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 autism. 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 parents 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 well-being 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 (insert name), a psychologist/intern psychologist/Masters/PhD student will be working closely with you to conduct the necessary assessments and formulate interventions. XX, a research assistant/Masters/PhD 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: [aurie.mclay@canterbury.ac.nz](mailto:aurie.mclay@canterbury.ac.nz)

## Appendix F. Parent Consent Form



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

#### **CONSENT FORM FOR PARENTS/ CAREGIVERS**

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

- I wish to participate in the project, “An investigation into the efficacy of treatments for sleep disturbance in children with autism”
- I have read and been given a full explanation of this project and have had the opportunity to ask questions.
- I understand what will be required of myself and my child/the child in my care during this project.
- I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study. However, if the intervention results in an increase in family stress, the staff working with us will provide support.
- I understand that all information about my family will be treated as confidential unless there is concern about anyone’s safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published
- I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of my child and I will be maintained
- I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data
- I understand that all research data that is collected will be securely stored at the University of Canterbury for a minimum of ten years
- 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 G. Parent Audio-Visual Recording Consent



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

#### **AUDIOVISUAL RECORDING CONSENT FORM**

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

Please read the statements below, which explain the purpose of audiovisual recording and how your 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
- The audiovisual file will only be seen by the researchers
- The audiovisual recording will be securely stored at the University of Canterbury for a minimum of ten years

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

Signed: \_\_\_\_\_

Date: \_\_\_\_\_

## Appendix H. Screening Phone Call Example

The purpose of our call today is to tell you a little bit more about what we do here and if you are still interested then I will ask you some more about xxx and your concerns for their sleep.

I am part of a team of researchers and clinicians (team of psychologists, interns, Masters and PhD students) at UC – received funding from Marsden Royal Society and Health fund to research/treat children with sleep problems and developmental disabilities generally. We currently have funding for working with children with a diagnosis of ASD and rare genetic disorders.

We are looking at the effectiveness of behaviourally based interventions for sleep difficulties.

### Process:

- Gain consent from you to proceed if you are interested.
- Start by doing a clinical interview where we find out a bit more information around the family context, xxx's developmental history and more specific information about the sleep difficulties.
- We use this information to formulate a treatment plan.
- Because you are based in xxx it is likely we will do that over the phone or skype.
- OR as you are in Christchurch you can either come into the University or we can come to you.
- Once we have done that we ask the families to record sleep diaries every night (in the morning - take about 5 minutes every night usually) which are done through all the stages of the study. This records number of times waking, what is happening when he is awake etc.
- Usually a week of diaries to begin with and a few times throughout treatment – helps us to monitor progress and see what is/isn't effective and anything we may need to change.
- We also ask families to record video because we find that really helpful in terms of determining what is going on when the child at times that you may not be aware of – it also gives us a chance to observe exactly what the behaviour looks like.
- We provide video equipment and instructions on how to set this up – if you are in Christchurch we can come and set this up for you.
- We download all the footage and code it etc. so you don't have to worry about that – just the initial set up.
- We get consent and tell the child that the camera will be in their room.
- We usually record every 2<sup>nd</sup> or 3<sup>rd</sup> night.
- We usually work with families until they are happy with the progress they have made or they to pull out from the study.
- It varies a bit in length as the rate of progress varies rate of progress varies. It can be anywhere from a few weeks to a few months.
- We usually expect to see positive change within a few nights – if we don't then that is why we collect the diaries so we can see what is going on.
- We know there is a relationship between sleep and day time challenging behaviours so one of the things we are interested in is not only in the effectiveness of sleep interventions but also investigating any secondary benefits of sleep improvement – parent and child wellbeing, day time behaviour etc.
- We ask families to fill out some questionnaires at the beginning and some at the end so that we can see if there has been any improvement in terms of sleep and daytime behaviours, parent and child wellbeing etc.

### Sleep Question Examples:

- Tell me a bit more about xxx's sleep. Start at the beginning of the night with the bedtime routine etc.
- Check any day time sleeping.
- What time does xxx go to bed?
- How often do you use medication (e.g. melatonin)?
- Check where the child is sleeping (e.g. own bedroom/bed)
- When xxx wakes what does he do?
- Do you often do xxx (e.g. lie with child) and does that help?
- Sounds like you've tried quite a few different things to help xxx settle back to sleep

### Ending:

- Would you like me to send you through some more information sheets and some consent forms?
- It's possible that we will need to get in touch with xxx's paediatrician just to make sure we weren't doing anything that could potentially be unsafe.
- Check if they have a scanner that they can send the consent forms back or if post is better.
- Will also send forms for xxx as it sounds like they want to be involved in the process.
- When we get the forms back then we will get in touch regarding how to proceed from here.

Consent for separated parents – does xxx stay with their father/mother? Is he/she aware that xxx may be a part of the programme? We can call zzz and tell them more about it. Does zzz notice a difference in xxx sleep? Ask zzz to get in touch with us or ask zzz if it is ok to share their number.

# Appendix I. Parent Sleep Diary

## Sleep Diary

Child's Name:

	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 <sup>st</sup> Night time awakening	Time & Duration of awakening	_____ mins						
	Behaviour while awake (Describe)							
	Your responses (Describe)							
		~~~~~		~~~~~		~~~~~		~~~~~
2 <sup>nd</sup> Night time awakening	Time & Duration of awakening	_____ mins						
	Behaviour while awake (Describe)							
	Your responses (Describe)							
		~~~~~		~~~~~		~~~~~		~~~~~
Time awake in the morning								

\*Curtain calls: Any behaviour such as leaving the bed (or bedroom) or calling parents into the room, between the time of being put to bed and falling asleep

Notes:

Appendix J. Example Bedtime Story for PED

Cars Read Along Story book, Read Aloud Story Books, Cars 3 Storybook



Adapted by  
**Victoria Saxon**

Illustrated by  
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Designed by  
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