AN EVALUATION OF THE EFFECTIVENESS OF ANTECEDENT-BASED MODIFICATIONS FOR TREATING SLEEP PROBLEMS AMONG CHILDREN ON THE AUTISM SPECTRUM

A thesis submitted in partial fulfilment of the requirements for the degree of Master of Science in Child and Family Psychology at the University of Canterbury

By Kate Ford

University of Canterbury

2022

Contents

List of Tables

List of Figures

List of Abbreviations

Achenbach System of Empirically Based Assessment	ASEBA
Adolescent Sleep Hygiene Scale	ASHS
Adolescent Sleep Wake Scale	ASWS
American Psychiatric Association	APA
Attention-deficit/hyperactivity disorder	ADHD
Autism spectrum disorder	ASD
Child Behavior Checklist	CBCL
Children's Sleep Habits Questionnaire	CSHQ
Depression, Anxiety and Stress Scale	DASS
Diagnostic and Statistical Manual of Mental Disorders	DSM
Evidence-Based Assessment	EBA
Faded bedtime with response cost	FBRC
Functional behavioural assessment	FBA
Gamma-aminobutyric acid	GABA
Gilliam Autism Rating Scale, Third Edition	GARS-3
Interobserver agreement	IOA
Long-term follow-up	LTFU
Motivating operations	MO
Multidimensional Anxiety Scale for Children, Second Edition	MASC 2
Night wakings	NWs
New Zealand	NZ
Phase 1, 2, 3	P1, 2, 3
Paediatric Quality of Life Inventory	PedsQL
Percentage below the mean	PBM

Percentage exceeding the mean	PEM
Pittsburgh Sleep Quality Index	PSQI
Post-extinction response burst	PERB
Questions About Behavioral Function	QABF
Relationship Quality Index	RQI
Repetitive Behaviour Scale-Revised	RBS-R
Short-term follow-up	STFU
Sleep Assessment Treatment Tool	SATT
Sleep Self-Report	SSR
Sleep onset delay	SOD
Sleep onset latency	SOL
Total sleep time	TST
Treatment Acceptability Rating Form-Revised	TARF-R
Vineland Adaptive Behaviour Scales, Third Edition	Vineland-3
Youth Self-Report	YSR

Acknowledgements

To my supervisors; Associate Professor Laurie McLay and Associate Professor Karyn France. Thank you for your time, feedback, and patience in the writing of this thesis, and for your support and guidance in my clinical work. I am very grateful to have learnt from distinguished experts in a field that I am passionate about and to have been able to develop my academic and clinical skills under your supervision. This research would not have been possible without your knowledge and expertise. I would also like to thank Emeritus Professor Neville Blampied for sharing his expertise in single-case research designs and data analysis, and Professor Philip Schluter for his time constructing the circadian graphs.

To Mum and Dad, thank you for always believing in me and never letting me doubt that my dreams were possible. Dad, thank you for being my biggest supporter, modelling hard work and perseverance, and offering lightness and humour. Mum, thank you for installing my passion for working with children and families and modelling endless kindness, compassion, and empathy as a teacher and a mother. Thank you also to Lucy, Tom and my extended whānau for your love and support across my time at university.

To my amazing friends, thank you for always being there for me and making sure I made time for self-care and to have fun. To my CFPY friends, I am so grateful to have met you and to be navigating this journey with you.

And finally, thank you to all the children and families that participated in this research. This thesis would not be possible without your time and dedication.

Abstract

Sleep problems are a commonly reported issue for children and adolescents on the autism spectrum. Given the impact of these problems on the child and their family, it is imperative to provide families with an effective intervention. More recently, research has demonstrated the effectiveness of behavioural interventions informed by functional behavioural assessment (FBA) for treating sleep problems among children and adolescents on the autism spectrum. There is, however, a paucity of research investigating the effectiveness of modification to sleep/wake schedules and other antecedent-based modifications alone, for treating sleep problems in this group. This thesis consists of two parts. Part one is a systematic review of the effectiveness of modification to antecedent variables (including modification to sleep/wake schedules) alone, for treating sleep problems in children and adolescents on the autism spectrum. Part two employed a single-case AB design to investigate the effectiveness of modification to sleep/wake schedules and other antecedent-based modifications alone or in combination, and consequence-based modifications if needed, for reducing sleep problems in children and adolescents on the autism spectrum. This study also examined parental ratings of treatment acceptability; any collateral benefits of behavioural sleep intervention for children's wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parents' sleep, mental health, and relationship quality; and the maintenance of treatment effects. Participants were six children and adolescents aged 3 to 19 years, with a diagnosis of autism spectrum disorder (ASD). Each participant's individualised intervention was informed by FBA and started with modification to sleep/wake schedules. The results showed that intervention was effective in reducing sleep problems for all participants, and most improvements were maintained at short- and long-term follow-up. Consequence-based modifications only needed to be implemented for one participant to fade sleep dependencies. In this case, modification to sleep/wake schedules were effective in improving the

participant's sleep problems, but a faded parental presence procedure was needed to eliminate parental presence and nighttime milk also needed to be eliminated. Overall, some positive change was evident in children's wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and in parent's mental health and sleep following intervention. In addition, all parents indicated that the intervention was acceptable; it was effective, reasonable, and they were willing to implement the strategies. Parents also reported clear understanding though a small number reported that it was costly, disruptive or time consuming, and there were side-effects. Overall, this study indicates the utility of antecedent-based modifications as a less restrictive, minimally sufficient intervention for treating sleep problems among children and adolescents on the autism spectrum.

Co-Authorship Form

Deputy Vice-Chancellor's Office
Postgraduate Research Office

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Co-Authorship Form - Masters

This form is to accompany the submission of any thesis that contains research reported in co-authored work that has been published, accepted for publication, or submitted for publication. A copy of this form should be included for each co-authored work that is included in the thesis. Completed forms should be included at the front (after the thesis abstract) of each copy of the thesis submitted for examination and library deposit.

Please indicate the chapter/section/pages of this thesis that are extracted from co-authored work and provide details of the publication or submission from the extract comes:

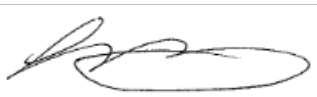
Chapter Two and Appendix A.

Chapter Two summarises the overall findings of the systematic review (Ford et al., 2021), and refers to the published version presented in Appendix A:
Ford, K. A., McLay, L. K., France, K. G., Blampied, N. M., & Gibbs, R. M. (2021). Systematic review of the effect of modification of antecedents in the treatment of sleep problems among children on the autism spectrum. *Advances in Neurodevelopmental Disorders*, 1-17. https://doi.org/10.1007/s41252-021-00224-8

Please detail the nature and extent (%) of contribution by the candidate:
The candidate (70%):
Conducted the search for the systematic review, screened and assessed the articles for eligibility, extracted the data for eligible articles, completed the study quality evaluation, wrote the original paper, and edited the paper based on feedback from the co-authors.

Certification by Co-authors:
If there is more than one co-author then a single co-author can sign on behalf of all
The undersigned certifies that:

- The above statement correctly reflects the nature and extent of the Masters candidate's contribution to this co-authored work
- In cases where the candidate was the lead author of the co-authored work he or she wrote the text

Name: *Laurie McLay* Signature:  Date: *10 Feb 2022*

Chapter One: Introduction and Literature Review

One of the most challenging issues reported by parents of children on the autism spectrum is sleep problems (Cohen et al., 2014). Estimates suggest that up to 80% of children and adolescents (henceforth referred to as children) on the autism spectrum experience some type of sleep problem (Hodge et al., 2014), including abnormalities in sleep/wake schedules, unwanted co-sleeping, reduced sleep duration, and dyssomnias such as early morning wakings, night wakings, and sleep onset difficulties (Goldman et al., 2012; Liu et al., 2006; Richdale & Prior, 1995). These problems have a significant impact on the health and wellbeing of children and their family (Carnett et al., 2021).

Sleep problems among children on the autism spectrum are thought to have a multi-factorial aetiology (Richdale & Schreck, 2009), comprised of biological, medical, psychological, social, and/or behavioural factors. A biopsychosocial framework was developed to conceptualise the complex interaction between these multiple factors (Richdale & Schreck, 2009). The salience of bio-behavioural factors is further described in the two-process model of sleep regulation (Borbély et al., 2016) and the behavioural model of sleep disturbance (Blampied & France, 1993).

There is an emerging body of research demonstrating the effectiveness of behavioural interventions for treating sleep problems in typically developing children (Meltzer & Mindell, 2014; Owens et al., 1999) and children on the autism spectrum (McLay et al., 2017; McLay et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021; van Deurs et al., 2019). Most recently, behavioural sleep interventions have been informed by Functional Behavioural Assessment (FBA) (for e.g., McLay, France, Blampied, van Deurs, et al., 2021). FBA is an evidence-based approach often used to identify the setting events, antecedents, and consequences of a behaviour, and develop a hypothesis about the function of that behaviour; this data is used to inform the design and implementation of an individualised behavioural

intervention plan (Blampied, 2013; Brown & Piazza, 1999; Jin et al., 2013). Within the context of this research, antecedent-based modifications include modification to sleep/wake schedules, sleep hygiene modifications, and modification to discriminative stimuli for sleep onset (McLay, France, Blampied, van Deurs, et al., 2021). Consequence-based modifications include standard extinction, modified extinction, and the use of rewards (Owens et al., 1999). Recent research suggests that FBA-informed interventions are effective in treating sleep problems in children on the autism spectrum (McLay et al., 2019; van Deurs et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021).

This thesis is comprised of two parts. Part one is a systematic review of the effectiveness of modification to sleep/wake schedules and other antecedent-based modifications for treating sleep problems in children on the autism spectrum. This systematic review, entitled "Systematic Review of the Effect of Modification of Antecedents in the Treatment of Sleep Problems Among Children on the Autism Spectrum", was accepted for publication in a Special Issue of *Advances in Neurodevelopmental Disorders* and has been published (see Chapter Two; Appendix A; Ford et al., 2021). Part two will use a single-case AB design to evaluate the effectiveness of modification to sleep/wake schedules, other antecedent-based modifications, and consequence-based modifications if needed, when implemented consecutively (i.e., modification to sleep/wake schedules; modification to sleep/wake schedules + antecedent-based modifications; modification to sleep/wake schedules + antecedent-based modifications + consequence-based modifications), to treat sleep problems in children on the autism spectrum. The maintenance of treatment effects, parent ratings of acceptability, and the collateral effects of reduced sleep problems for the child and their parents will also be reported.

In this chapter, an overview of the diagnostic features, characteristics, prevalence, and aetiology of autism is provided. Next, the prevalence, course, type, aetiology, and impact of

sleep problems is discussed. Then, the behavioural model of sleep disturbance is introduced. Common behavioural and pharmacological sleep interventions for children on the autism spectrum are then described, in the context of relevant literature. Finally, least restriction and minimal sufficiency are discussed as the guiding principles for this research.

Autism Spectrum Disorder

Definition

Autism spectrum disorder (ASD) is a neurodevelopmental disability characterised by abnormalities in social interaction and communication, and restricted, repetitive behaviours, activities, or interests (American Psychiatric Association [APA], 2013). Social differences may manifest as difficulties with social-emotional reciprocity, forming and maintaining relationships, understanding social contexts, and nonverbal communication. Behavioural differences include stereotyped speech (e.g., echolalia), movements (e.g., motor stereotypies) or play, strict adherence to routines, difficulty managing change, transitions or deviation from the typical routine, extremely restricted and fixated interests, and hypo- or hyper-reactivity to sensory stimuli (APA, 2013).

In accordance with the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) criteria, the onset of autism symptomology must be evident in early childhood (APA, 2013). This may be evidenced by developmental delays, a social or language skill loss or regression, and differences in social interactions. These symptoms cause impairment in social, occupational, or another domain of functioning, and are not more appropriately explained by global developmental delay or intellectual disability. The DSM-5 has conceptualised autism on a spectrum with level of severity ranging from requiring minimal support to requiring very significant support (APA, 2013).

Characteristics Associated with Autism

It is common for individuals on the autism spectrum to have an intellectual (La Malfa et al., 2004) and/or language impairment (APA, 2013). Moreover, individuals with average to high overall intellectual abilities may show variability in functioning across cognitive domains, and there are often discrepancies between overall intellectual abilities and adaptive functioning (APA, 2013). Also, individuals may demonstrate difficulties with motor skills as evidenced by clumsiness or abnormal gait (APA, 2013; Dziuk et al., 2007). Many children on the autism spectrum engage in challenging behaviours including stereotypy, aggression, tantrums, and self-injurious behaviour (Jang et al., 2011; Matson et al., 2008), and there are high rates of anxiety and/or depression in adolescents and adults on the autism spectrum (APA, 2013; Uljarević et al., 2019).

Prevalence

ASD is one of the most common developmental disabilities with an estimated prevalence of around 1 in 69 (14.5 per 1,000) children in the United States (Christensen et al., 2018). Internationally, the estimated prevalence of autism has significantly increased following advances in diagnostic assessment and conceptualisation of autism as a spectrum disorder (Bowden et al., 2020). However, there is a scarcity of data pertaining to the prevalence and incidence rates of ASD in the New Zealand population (Bowden et al., 2020). More recently, the 2018/19 New Zealand Health Survey reported that as many as 2% of children aged 2 to 14 years have a diagnosis of ASD (Ministry of Health, 2019). A local study using health administration data from 2015/2016 found that 1 in 102 New Zealand children (0.0098%) aged 8 years and 1 in 174 children and adolescents (0.0057%) aged 0 to 24 years, have been diagnosed with ASD (Bowden et al., 2020). In terms of demographics, the prevalence of ASD was higher for males than females and for those of New Zealand European descent compared to those of Māori or Pasifika descent (Bowden et al., 2020).

Aetiology

Over the past few decades, many theories have been proposed regarding the aetiology of autism. Recent research suggests that there are multiple risk factors and mechanisms, including biological factors, prenatal and environmental factors, and epigenetic factors (Yoon et al., 2020).

Biological Factors. Multiple genes have been identified as being involved in the aetiology of autism (Yoon et al., 2020). Research has focused on the FOXP2, IMMP2L, RELN, and RAY1/ST7 genes on particular loci (7q22-7q33) of chromosome 7 (Muhle et al., 2004). The FOXP2, IMMP2L, RELN, and RAY1/ST7 genes have been associated with symptoms of autism including differences in the development of speech and language (Yoon et al., 2020). In terms of neurobiological factors, many children on the autism spectrum show abnormalities on an electroencephalogram (Hrdlicka et al., 2004; Yasuhara, 2010). For example, one study found that around 85% of children on the autism spectrum had abnormalities in epileptiform activity (Yasuhara, 2010). Further, estimates indicate that around 20% to 37% also have epilepsy (Hrdlicka et al., 2004; Yasuhara, 2010). As ASD and epilepsy frequently co-occur, this suggests that a common mechanism underlies the aetiology of these disorders, leading to this common co-occurrence (Lewis et al., 2018). The underlying neurobiology linking these two disorders is, however, unclear (Lewis et al., 2018). Another structural difference associated with ASD is abnormalities in grey matter volume and development (Greimel et al., 2013; McAlonan et al., 2008). For example, Greimel et al. (2013) examined grey matter volume in 47 children, adolescents, and adults on the autism spectrum, and 51 controls. Researchers used voxel-based morphometry to draw comparisons between the two groups. Results showed reductions in the grey matter volume in specific brain regions in individuals on the autism spectrum compared to the controls. Researchers also reported age-related differences in the grey matter volume of individuals on the autism

spectrum and different patterns of development of grey matter volume between the two groups (Greimel et al., 2013). Another interesting and widely replicated finding is that individuals on the autism spectrum often present with elevated levels of serotonin (Gabriele, et al., 2014). Gabriele et al. (2014) conducted a systematic review and meta-synthesis and found that individuals on the autism spectrum had higher levels of blood serotonin compared to controls (individuals without autism). Researchers concluded that serotonin levels are associated with ASD, and elevated levels of serotonin may be a reliable biomarker (Gabriele et al., 2014).

Environmental Factors. In the past 20 years, it has been hypothesised that the measles, mumps, and rubella vaccine causes ASD (DeStefano & Shimabukuro, 2019). However, this has been widely discredited as an environmental cause (DeStefano & Shimabukuro, 2019). In terms of prenatal factors, viral infections, zinc deficiency and parental age have been suggested to play a role (Yoon et al., 2020). For example, infection with chicken pox, rubella, measles, mumps, cytomegalovirus, and varicella zoster have been investigated though further research is warranted (Libbey et al., 2005). It is also postulated that autism may be caused by parental autoimmune disease (Keil et al., 2010), or bacterial infection in the mother's second trimester (Atladóttir et al., 2010) or third trimester following hospitalisation (Lee et al., 2015; Zerbo et al., 2015). Finally, zinc deficiency in mothers has been implicated as a risk factor for autism (Yoon et al., 2020). Zinc is involved in important processes including gene expression regulation, immune system functioning, and foetal and child growth and development (Pfaender et al., 2017).

Another widely reported risk factor for autism is parental age (Croen et al., 2007; Glasson et al., 2004). Several meta-analyses have found that the risk for autism increases as maternal and/or paternal age increases (Sandin et al., 2012; Wu et al., 2017). The reason for

this is unclear, however, it may be because the aging process causes an increase in de novo genetic mutations in developing germ cells (Croen et al., 2007).

Epigenetic Factors. Epigenetic factors are involved in the regulation of gene expression and chromatin structuring (Yoon et al., 2020). As epigenetic factors are implicated in genes and processes related to brain development, dysregulation in epigenetic mechanisms may play a role in the cause of autism and other neurodevelopmental disabilities (Yoon et al., 2020). Specific epigenetic factors that have been implicated in autism include DNA methylation, microRNA, and histone modification (Keil & Lein, 2015; Yoon et al., 2020). For example, environmental factors including chemical exposure, drug use, hormones, and stress may alter DNA methylation and thus, increase the risk of autism (Keil & Lein, 2015). Further, impaired microRNA synthesis or abnormalities in the regulation of proteins involved in histone modification (a process that affects gene expression) have been associated with the underlying aetiology of autism (Mbadiwe & Millis, 2013; Yoon et al., 2020).

Sleep Problems in Children on the Autism Spectrum

Prevalence of Sleep Problems

Sleep problems in children on the autism spectrum are widely reported with prevalence estimates suggesting that around 50% to 80% of children on the autism spectrum experience some type of sleep issue (Hodge et al., 2014; Kotagal & Broomall, 2012; Richdale & Schreck, 2009; Schreck & Mulick, 2000). This is compared to around 20% to 30% of typically developing children (Johnson & Malow, 2008; Owens et al., 2000) and 13% to 86% of children with other types of developmental disabilities (Cotton & Richdale, 2006; Didden & Sigafoos, 2001). The estimated prevalence is high, however, it should be viewed somewhat cautiously as variables such as age, cognitive functioning, sample size, sampling factors, and the psychometric instruments or criteria used to conceptualise sleep problems may affect these estimates (Richdale & Schreck, 2009). Further, there is a lack of consistency and

agreement between researchers in paediatric sleep in the definition of sleep problems (Richdale & Schreck, 2009). Additional measurement issues can arise when measuring sleep problems in children (Cortesi et al., 2010; Richdale & Schreck, 2009). For example, typically developing older children, adolescents, or adults can accurately self-report sleep problems, though sleep problems are reported to professionals by parents, caregivers, teachers, or other significant adults for children and adolescents with developmental disabilities (Richdale & Schreck, 2009). Informant reporting of sleep problems may therefore affect prevalence estimates (Richdale & Schreck, 2009). Despite these shortcomings, it is clear that children on the autism spectrum and children with other developmental disabilities are suspectable to experiencing sleep problems (Cortesi et al., 2010).

Course of Sleep Problems

The onset of sleep problems in many individuals on the autism spectrum occurs during childhood (Giannotti et al., 2006). One study found the peak onset of sleep problems in individuals on the autism spectrum occurs at age 2 (Giannotti et al., 2006). For children on the autism spectrum, it is common for sleep problems to persist overtime (Giannotti et al., 2006; Goldman et al., 2012; Hodge et al., 2014). To illustrate, in one study, parents of children on the autism spectrum were asked if their child currently has sleep difficulties or has had sleep difficulties in the past (Wiggs & Stores, 2004). In this study, 83% of these children had a history of sleep difficulties and these problems persisted for 67% of children (Wiggs & Stores, 2004). Children and adolescents on the autism spectrum also demonstrate changes in the types of sleep problems experienced overtime (Hodge et al., 2014). For example, a study by Hodge et al. (2014) examined age-related changes in sleep problems among children on the autism spectrum and typically developing children. Participants were mothers of children on the autism spectrum (n= 108) and typically developing children (n= 108), aged 3 to 17 years. Results showed that for children on the autism spectrum, overall

sleep, bedtime resistance, night wakings, and sleep anxiety increased with age between 3 to 5 years and 6 to 9 years, and then reduced between 6 to 9 years and 10 to 17 years. Further, daytime sleepiness increased consistently across these three age-groups. For typically developing children, however, overall sleep, bedtime resistance, night wakings, sleep anxiety, and daytime sleepiness reduced or remained constant across these age-groups. Studies have also demonstrated age-related changes in ASD symptoms, including those symptoms that are associated with sleep problems (Hodge et al., 2014). Thus, children on the autism spectrum may experience a change in sleep problems as these symptoms also change overtime (Hodge et al., 2014).

Types of Sleep Problems

Primary sleep disorders are typically categorised into two groups: parasomnias and dyssomnias (Schreck & Mulick, 2000). Parasomnias occur due to activation of the central nervous system, affecting the experience of falling asleep, sleep maintenance, transitioning between stages of sleep, and waking (Schreck & Mulick, 2000). Examples of parasomnias include disorders of partial arousals (e.g., sleep terrors, sleep walking), bruxism, and enuresis (Ming et al., 2009). Dyssomnias, defined as difficulty initiating or maintaining sleep, are the most frequently reported type of sleep problem experienced by children on the autism spectrum (Cortesi et al., 2010; Goldman et al., 2012; Schreck & Mulick, 2000). Dyssomnias affect sleep quality and duration, and include abnormalities in sleep/wake patterns, early morning wakings, night wakings, sleep onset delay, poor sleep quality, insufficient sleep duration, and unwanted co-sleeping (Cortesi et al., 2010; Richdale & Schreck, 2009). Parents of children on the autism spectrum also report challenging behaviour before bed and during night wakings including calling out, leaving their bedroom, and non-compliance with the bedtime routine (Richdale, 2013). Dyssomnia-related sleep problems and associated challenging behaviours are the focus of this research.

Aetiology of Sleep Problems

The aetiology of sleep problems among children on the autism spectrum is thought to be multi-factorial and include a combination of biological, medical, psychological, social, and/or behavioural factors (Blampied & France, 1993; Johnson & Malow, 2008). Recently, researchers have developed a biopsychosocial model of sleep problems to understand the interactions between factors that predispose and precipitate sleep problems in children on the autism spectrum (Richdale & Schreck, 2009).

Biological Factors. Genetic factors have been implicated in sleep problems in children on the autism spectrum as they can change brain architecture and cause abnormalities in hormones, clock genes, and circadian rhythms (Bourgeron, 2007; Richdale & Schreck, 2009). For example, Nicholas et al. (2007) investigated the hypothesis that autism is associated with clock gene abnormalities. Clock genes have a salient role in modulating circadian rhythms as they influence our internal biological clock (Bourgeron, 2007; Richdale & Schreck, 2009). Results demonstrated that there is an association between the single nucleotide polymorphisms (SNPs) of two clock genes PER1 and NPAS2, and autism which may be linked to sleep problems (Nicholas et al., 2007). Given that SNPs are a type of genetic variation, they may affect the functioning of PER1 and NPAS2 clock genes in people on the autism spectrum (Nicholas et al., 2007). Circadian rhythm abnormalities related to low levels of melatonin have consistently been found in individuals on the autism spectrum (Bourgeron, 2007; Rossignol & Frye, 2011). Circadian and melatonin physiology abnormalities will be further discussed in a later section (see *The Role of Circadian Rhythms in Sleep).*

Abnormalities in neurotransmitter expression of serotonin and gamma-aminobutyric acid (GABA) have also been reported in people on the autism spectrum (Rolf et al., 1993; Singh & Zimmerman, 2015). For instance, Rolf et al. (1993) studied the levels of serotonin

and GABA in 18 children on the autism spectrum compared to 14 typically developing children. Researchers found significant differences between the two groups, with increased serotonin and decreased GABA levels in children on the autism spectrum. Given that serotonin and GABA are important neurotransmitters for regulating the sleep/wake cycle, abnormal serotonin and GABA levels may lead to sleep disturbance (Richdale & Schreck, 2009; Singh & Zimmerman, 2015).

Medical Factors. Children on the autism spectrum may also be susceptible to sleep problems due to comorbid neurological and medical conditions. Examples include allergies, asthma, visual impairments, gastrointestinal problems (e.g., gastroesophageal reflux), seizure disorders, and obstructive sleep apnoea, most of which occur at higher rates in children on the autism spectrum when compared to typically developing children (Deliens et al., 2015; Johnson & Malow, 2008; Kotagal & Broomall, 2012; Singh & Zimmerman, 2015). For example, Mannion et al. (2013) examined coexisting disorders including gastrointestinal symptoms, epilepsy, and sleep problems in 89 children on the autism spectrum, aged 3 to 16 years. Results showed that around 80% of children had sleep problems, 79% had experienced gastrointestinal symptoms. and 10% reported having epilepsy. Abdominal pain and gastrointestinal symptoms significantly predicted sleep anxiety and parasomnias, respectively (Mannion et al., 2013).

Sleep problems in children on the autism spectrum may be further exacerbated by the side-effects of medications that are used to treat neurological, medical, and psychiatric disorders (for e.g., corticosteroids, antiepileptic drugs, stimulants, and selective serotonin reuptake inhibitors) (Johnson & Malow, 2008; Reynolds & Malow, 2011; Singh & Zimmerman, 2015). A study conducted by Liu et al. (2006) investigated correlates of sleep problems in 167 children on the autism spectrum, aged 2 to 18 years. They found that several

variables including medications and epilepsy were associated with sleep problems in children on the autism spectrum (Liu et al., 2006).

Psychological Factors. Internalising and externalising problems have been associated with sleep problems in children (Quach et al., 2018). As children on the autism spectrum have higher rates of anxiety and depression (DeFilippis, 2018; Leyfer et al., 2006), they may be more susceptible to experiencing sleep problems (Richdale & Schreck, 2009). Indeed, internalising disorders such as depressive disorders and anxiety disorders have been associated with problems initiating and maintaining sleep in children on the autism spectrum (Johnson & Malow, 2008; Kotagal & Broomall, 2012; Park et al., 2012). For example, Park et al. (2012) examined the relationship between coexisting psychological disorders and sleep problems in 166 children on the autism spectrum compared to 111 typically developing siblings aged 4 to 15 years. Results showed that sleep problems occurred in higher rates among the children on the autism spectrum (Park et al., 2012).

In addition to internalising disorders, increased rates of sleep problems have been associated with externalising disorders such as attention-deficit/hyperactivity disorder (ADHD) and oppositional defiant disorder (Stein, 1999; Sung et al., 2008). This is significant because many externalising disorders co-occur in high rates in children on the autism spectrum, for example, one study of comorbid disorders reported 31% children on the autism spectrum also met DSM-IV criteria for ADHD and 7% met criteria for oppositional defiant disorder (Leyfer, 2006). Further, parents report increased challenging behaviour in children with autism and sleep problems (Adams, Matson, & Jang, 2014). For example, Adams, Matson, and Jang (2014) studied the relationship between challenging behaviour and sleep problems in 311 children on the autism spectrum, aged 2 to 18 years, recruited from a database. Results showed an association between sleep problems and higher ratings of challenging behaviour (Adams, Matson, & Jang, 2014).

Sleep problems may also be linked to the social and behavioural differences associated with autism including abnormalities in social interaction and communication, stereotypy, strict adherence to routines, and developmental delays as well as overall symptom severity (APA, 2013; Richdale & Schreck, 2009). For example, individuals on the autism spectrum experience distress with deviations from their typical routine, therefore, children that are used to the routine of falling asleep in their parent's arms or with them present, may have difficulty falling asleep alone (Deliens et al., 2015). Similarly, if there are changes in their typical bedtime, this may cause distress and sleep onset difficulties (Kotagal & Broomall, 2012; Mazzone et al., 2018). Schreck et al. (2004) examined a pre-established database of parent responses on two psychometric instruments: the Behaviour Evaluation of Disorders of Sleep and the Gilliam Autism Rating Scale. Participants were parents of children on the autism spectrum aged 5 to 12 years (Schreck et al., 2004). Researchers reported that children with a shorter sleep duration had a greater overall severity of ASD symptoms, stereotypic behaviours, and social difficulties (Schreck et al., 2004).

Social Factors. Factors within the family or home environment may also contribute to sleep problems in children on the autism spectrum (Richdale & Schreck, 2009). Family factors related to sleep problems in children include family functioning (Bell & Belsky, 2008), parental stress (Hoffman et al., 2008), and parenting practices (Shetty et al., 2022). For example, parenting practices may not facilitate the onset of good quality sleep and therefore contribute to sleep problems in children on the autism spectrum (Richdale & Schreck, 2009). Staples et al. (2015) report that inconsistent parenting practices are associated with shorter sleep duration in typically developing children. There is, however, a paucity of research examining the relationship between family factors and sleep problems in children on the autism spectrum (Richdale & Schreck, 2009). Further, researchers have suggested that there is a bidirectional relationship between family functioning and sleep problems in children, but

this has not been studied extensively (El-Sheikh & Kelly, 2017). Environmental factors that are associated with sleep problems in children on the autism spectrum include an inconsistent bedtime routine and sleep/wake schedule, using technology before bed, and a bedroom environment that is not conducive for good quality sleep (i.e., too light, too hot or cold, and noisy) (Richdale & Schreck, 2019; van Deurs et al., 2019; see *Sleep Hygiene)*.

Impact of Sleep Problems

Sleep problems have a significant impact on the health and wellbeing of children, their parents and other family members (Carnett et al., 2021). For children, poor sleep has been associated with emotional problems (Malow et al., 2006), behavioural problems (Goldman et al., 2011), medical issues (Liu et al., 2006; Williams et al., 2004), cognitive functioning (Hollway et al., 2013), quality of life (Delahaye et al., 2014), and severity of autism symptomatology (Hoffman et al., 2008; Schreck et al., 2004). A large body of research has shown that sleep problems have an impact on the daytime behaviour of children on the autism spectrum (for e.g., Mazurek & Sohl, 2016; Park et al., 2012; Sikora et al., 2012). Sikora et al. (2012) examined the link between daytime behaviour and sleep problems in a sub-group of children on the autism spectrum, from a registry. Participants were children aged 4 to 10, with parent-reported sleep problems. They found that there is a negative association between sleep problems and daytime behaviour in this group; more internalising and externalising problems were reported for children on the autism spectrum with sleep problems (Sikora et al., 2012).

For parents, childhood sleep problems may negatively affect their sleep, mental health, stress levels, and quality of life (Davis & Carter, 2008; Hodge et al., 2013; Liu et al., 2021; Meltzer & Mindell, 2007) and for the wider family, their employment, sibling sleep, and family functioning (Carnett et al., 2021; Richdale & Wiggs, 2005). For example, a recent study investigated the impact of sleep problems in children on the autism spectrum on their

parent's quality of life (Liu et al., 2021). Participants were 440 children on the autism spectrum and 334 typically developing children of the same age. They found a negative association between sleep problems and parental quality of life for children on the autism spectrum (Liu et al., 2021).

The Role of Circadian Rhythms in Sleep

The Two-process Model of Sleep Regulation

A well-established framework in the sleep literature is the two-process model of sleep regulation (Borbély et al., 2016; Glickman, 2010). The model stipulates that sleep and wakefulness are regulated by homeostatic and circadian processes (Borbély et al., 2016; Glickman, 2010). Homeostatic sleep pressure can be defined as the body's biological drive for initiating or maintaining sleep (Glickman, 2010). Thus, an individual is able to fall asleep and remain asleep more easily with adequate sleep pressure. By contrast, insufficient sleep pressure may result in difficulty falling and staying asleep. Several variables influence sleep pressure including environmental factors (for e.g., darkness of the bedroom), and the duration of time since the individual's last sleep episode (Borbély et al., 2016). Sleep pressure progressively increases during wakefulness and decreases over the period of sleep (Glickman, 2010). This homeostatic process interacts with the circadian system to provide a consistent schedule of sleep and wakefulness. Circadian rhythms are behavioural and physiological processes with a cycle period of around 24-hours (Glickman, 2010). They are the product of our internal biological clock, the suprachiasmatic nucleus (SCN) in the hypothalamus of the brain (Borbély et al., 2016; Glickman, 2010). Circadian and homeostatic processes are both behavioural and physiological as they must be reset and synchronised with zeitgebers or environmental time cues daily (Borbély et al., 2016; Glickman, 2010). Natural sunlight is a salient zeitgeber as it entrains the individual to the 24-hour cycle (Borbély et al., 2016).

Circadian Rhythms and Sleep in Children on the Autism Spectrum

As previously described, commonly reported sleep problems in children on the autism spectrum include abnormalities in sleep/wake patterns, early morning waking, night wakings, and sleep onset delay. These types of disturbances indicate that circadian rhythm abnormalities may play a causal role in the sleep problems experienced by children on the autism spectrum (Geoffray et al., 2016; Glickman, 2010; Wiggs & Stores, 2004). In many cases, it is thought that disruptions to circadian rhythms may be the result of irregular timing of endocrine processes such as abnormal melatonin regulation, synthesis, and synaptic transmission, as well as low levels of melatonin (Bourgeron, 2007; Deliens et al., 2015; Geoffray et al., 2016; Rossignol & Frye, 2011). This is supported by a large body of research suggesting that there are abnormalities in melatonin physiology among individuals on the autism spectrum (Rossignol & Frye, 2011; Rossignol & Frye, 2014). Melatonin is a neurohormone produced in the pineal gland that is integral to the regulation of the sleep/wake cycle as it helps to entrain an individual's internal clock to the 24-hour rhythm (Hodge et al., 2014; Richdale & Schreck, 2009). Therefore, abnormal timing or levels of melatonin production may lead to circadian rhythm disruptions and sleep disorders such as irregular sleep/wake rhythm, delayed sleep phase disorder, or advanced sleep phase disorder (Glickman, 2010; Hodge et al., 2014; Souders et al., 2009). As mentioned above, our internal biological clock is synchronised and entrained by zeitgebers (e.g., sunlight). Social cues are also salient for this synchronisation, though individuals on the autism spectrum experience social differences (i.e., difficulties with interpreting social cues) (Deliens et al., 2015). Therefore, individuals on the autism spectrum may experience difficulty with this synchronisation process, resulting in problems in the timing of their circadian rhythm (Deliens et al., 2015).

Behavioural Model of Sleep Problems

A behavioural model of sleep problems draws upon the principles of Applied Behaviour Analysis (ABA) to explain the processes that directly contribute to the development and maintenance of sleep problems (Blampied, 2013; Blampied & France, 1993). This model outlines the relationship between sleep and stimulus cues (respondent behaviour) and Skinner's three-term contingency (operant behaviour) (Blampied, 2013). According to this model, sleep can be best described as a bio-behavioural state rather than an operant or respondent behaviour (Blampied & France, 1993), as the transition into sleep involves a combination of biological processes and respondent and operant behaviours.

Principles of classical conditioning stipulate that cues that are present in the environment when a person goes to bed are important for initiating and maintaining sleep (Blampied, 2013). This is because falling asleep is an operant behaviour and therefore, under stimulus control (Blampied & France, 1993) (i.e., good sleep is achieved through adequate stimulus control). By contrast, inappropriate or inconsistent stimulus control before bedtime (i.e., lack of discriminative stimulus for sleep) may be instrumental in the maintenance of sleep disturbances (Blampied & France, 1993; Blampied, 2013).

Principles of operant conditioning stipulate that contingencies of reinforcement are also instrumental in maintaining sleep disturbances (Blampied & France, 1993). Skinner's three-term contingency refers to the interrelationship between a behaviour and antecedents and consequences (Blampied & France, 1993). Antecedent variables are those that precede the behaviour (Cooper et al., 2020), and act as a discriminative stimulus that facilitates sleep onset (Blampied & France, 1993; McLay et al., 2019). Consequence variables are those that follow the behaviour and have some effect on the future likelihood of the behaviour (Cooper et al., 2020). Within the sleep context, if sleep-interfering (e.g., calling out, leaving bed, playing with toys) or sleep-conducive (e.g., lying quietly, closing eyes) behavior is

reinforced, it is more likely to occur in the future (Blampied & France, 1993). Therefore, it is imperative to identify and target the contingencies of reinforcement that maintain sleep-conducive and sleep-interfering behaviours (Jin et al., 2013).

An important concept in the behavioural model of sleep disturbance is operant behaviour chains (Blampied, 2013). There are two crucial ways in which antecedents affect these behaviour chains. Firstly, falling asleep represents the endpoint of a particular behaviour chain which begins with the bedtime routine and ends with behavioural quietude (i.e., lying down in bed with closed eyes) which is necessary for sleep onset (the reinforcer). As this operant chain is under stimulus control, each individual step in the chain (e.g., having a bath, brushing teeth, putting on pyjamas, reading stories) serves as a separate discriminative stimulus for the following component (Blampied, 2013). Thus, an absence of consistent discriminative stimuli for falling asleep will cause termination of the behaviour chain that leads to sleep onset (Blampied & France, 1993). Behaviours that are consistent with other behaviour chains (i.e., sleep-interfering behaviours such as leaving their bed, calling out, engaging in stereotypy) may also cause termination of the chain and result in sleep disturbance. Further, if inappropriate stimuli are paired with sleep, the child may rely on these cues for initiating and maintaining sleep, and absence of these cues may lead to sleep disturbance (Blampied, 2013). A quintessential example is observed when children become dependent on parental presence while falling asleep (for e.g., McLay et al., 2019).

Another way in which antecedent variables affect the behaviour chain is by means of sleep pressure. Sleep pressure can be defined as an individual's motivation to sleep (Borbély, 1982; Borbély et al., 2016), and therefore understood as a motivational state. Sleep pressure can operate as a motivational variable and increase the reinforcement value of sleep; if an individual is sleep deprived this may increase the reinforcement value of sleep and thus, the individual's motivation to sleep (i.e., sleep pressure) (Borbély & Achermann, 1992; Michael,

1982). By this means, sleep pressure has increased the effectiveness of sleep as the reinforcer, and the salience of the discriminative stimuli that cue the availability of sleep. The conditions that influence an individual's motivation to sleep (i.e., sleep pressure) and thereafter, the value of sleep are referred to as motivating operations (MO; Cooper et al., 2020; Michael, 1982). Factors that may increase the reinforcement value of sleep include the amount of time the individual has been awake, a delayed bedtime, and exhaustion for reasons unrelated to sleep (e.g., daytime behaviour) (Borbély & Achermann, 1992; Michael, 1982).

When conducting an assessment, based on the behavioural model, it is imperative to identify the setting events, antecedents and consequences that are instrumental in precipitating and maintaining the sleep problems, as well as the function of the behaviour (Blampied, 2013; Cooper et al., 2020). A tool that is widely used for this purpose in sleep research is Functional Behavioural Assessment (FBA) (Blampied, 2013) (see *FBA*).

Sleep Interventions

The most common approaches for treating sleep problems in children on the autism spectrum are pharmacological and behavioural interventions (Johnson & Malow, 2008). Interventions that fall within each of these classifications are outlined below.

Pharmacological Interventions

Pharmacological interventions for sleep problems in children on the autism spectrum include melatonin, atypical antipsychotics, anti-depressants, and benzodiazepines (Johnson & Malow, 2008). It is not uncommon to treat childhood sleep problems with medication. In Australia, a survey found that the most frequently used sleep medication was melatonin (Heussler et al., 2012). Melatonin was prescribed the most for children on the autism spectrum, followed by children with other developmental disabilities (Heussler et al., 2012). Similarly, in the United States, researchers found that 1 in 3 psychiatrists prescribed medication at least 50% of the time to children on the autism spectrum and children with

other developmental disabilities presenting with sleep problems (Owens et al., 2010). A local study investigated melatonin use in children on the autism spectrum aged 0 to 18 years using data from the Integrated Data Infrastructure (McLay, Schluter, et al., 2021). Results showed that close to 25% of New Zealand children on the autism spectrum use melatonin (McLay, Schluter, et al., 2021).

Melatonin. Exogenous forms of melatonin may be helpful for children with circadian rhythm disturbances, including children on the autism spectrum (Johnson & Malow, 2008). As previously noted, endogenous melatonin is the neurohormone involved in regulating the sleep/wake cycle and entraining an individual's internal clock. Exogenous melatonin may act through a similar mechanism and reduce sleep problems by entraining an individual's circadian rhythm to a socially conventional sleep/wake schedule by increasing levels of melatonin at the appropriate time (Bourgeron, 2007; Johnson & Malow, 2008; Richdale & Schreck, 2009). There is a large body of research supporting the use of melatonin for improving sleep problems in children on the autism spectrum (for e.g., Giannotti et al., 2006; Gringras et al., 2017; Malow et al., 2012; Maras et al., 2018; Wirojanan et al., 2009; Wright et al., 2011). A meta-analysis of randomised controlled trials found that melatonin can effectively increase sleep duration and reduce sleep onset latency and frequency of night wakings in children on the autism spectrum, and children with other developmental disabilities (Braam et al., 2009).

Other Medications. Other pharmacological interventions for treating sleep problems in children on the autism spectrum include atypical antipsychotics (e.g., risperidone), anti-depressants with sedative properties (e.g., trazodone, mirtazapine, fluvoxamine), and benzodiazepines (e.g., clonazepam) (Johnson & Malow, 2008). A review of pharmacological interventions for sleep problems in children with developmental disabilities, found that trazodone and mirtazapine demonstrated some improvements for sleep problems, though

further research is warranted (Hollway & Aman, 2011). Importantly, a study of sleep problems and the use of sleep medication in children on the autism spectrum aged 4 to 10 years, found that almost half of the children took medication for sleep problems including melatonin, atypical antipsychotics, antidepressants, and benzodiazepines (Malow et al., 2016). Of note, children that took medication for sleep problems had a lower paediatric quality of life and more challenging daytime behaviour compared to children that did not take sleep medication (Malow et al., 2016).

Behavioural Interventions

Behavioural interventions are commonly used to treat sleep problems among children on the autism spectrum. Indeed, behavioural interventions are a widely recommended treatment in several, current literature reviews (e.g., Blackmer & Feinstein, 2016; Pattison et al., 2020). Behavioural interventions can be categorised into antecedent-based modifications and consequence-based modifications. A systematic review of the evidence supporting antecedent-based modifications for children on the autism spectrum is provided in Chapter 2.

Antecedent-based Modifications. Antecedent-based modifications for sleep problems include changes to sleep/wake schedules i.e., procedures that target circadian rhythms and physiological sleep pressure (e.g., implementing a consistent sleep/wake schedule, faded bedtime, chronotherapy, sleep restriction, scheduled awakenings), modification to sleep hygiene practices, and modification to discriminative stimuli for sleep onset (e.g., white noise, Gro-Clock™) (McLay, France, Blampied, van Deurs, et al., 2021). In addition to these procedures, there is emerging evidence that supports the use of visual supports (e.g., social stories, visual schedules, Gro-Clock™) to facilitate the implementation of these practices for children on the autism spectrum (McLay et al., 2019; van Deurs et al., 2019).

Sleep/wake Scheduling. Sleep/wake scheduling involves implementation of a consistent bedtime and wake time everyday including the weekend (Owens et al., 1999). The bedtime is set to the average time of sleep onset, and the wake time allows the child to achieve age-appropriate sleep and have sufficient sleep pressure (Owens et al., 1999). Age-appropriate sleep is calculated using appropriate guidelines, for example, the National Sleep Foundation's recommendations (Hirshkowitz et al., 2015).

Faded Bedtime. The faded bedtime procedure involves designating an initial bedtime that is within 15 minutes of the child's typical sleep onset time (Piazza et al., 1997; Vriend et al., 2011), in order to reduce sleep onset latency and increase homeostatic sleep pressure. The child is also woken at the same time each morning (Piazza et al., 1997; Vriend et al., 2011), with the wake time selected based upon age-appropriate sleep duration guidelines (e.g., Hirshkowitz et al., 2015). To maintain sleep pressure, daytime sleep is eliminated unless it is age-appropriate (Piazza et al., 1997; Vriend et al., 2011). Using this procedure, the initial bedtime is shifted progressively earlier until the goal bedtime is reached, with the designated wake time maintained (Piazza et al., 1997; Vriend et al., 2011). Faded bedtime with response cost (FBRC) is a variation of this procedure, with a consequence-based modification. The FBRC procedure includes all of the previous steps, with the addition of parents removing the child from their bed if sleep onset does not occur within an appropriate time period (Piazza et al., 1997; Vriend et al., 2011). The goal of this response cost (removal of the child) is to increase the child's incentive for falling asleep (Vriend et al., 2011). The child engages in a neutral activity when out of bed and is returned to bed typically after around 30 minutes (Vriend et al., 2011). Parents repeat this sequence until the child falls asleep. Previous research supports faded bedtime with and without response cost for typically developing children and children on the autism spectrum (Delemere & Dounavi, 2018; Kodak & Piazza, 2011; Moon et al., 2011). For example, Moon et al. (2011) investigated the effectiveness of

faded bedtime with response cost (and positive reinforcement) for sleep problems in three children on the autism spectrum, aged 8 to 9 years. Researchers found that sleep onset latency reduced for all participants, with treatment gains maintained at follow-up 12 weeks post-treatment (Moon et al., 2011).

Chronotherapy. Similar to faded bedtime, chronotherapy involves shifting the child's bedtime progressively (Piazza et al., 1998; Vriend et al., 2011). In contrast to faded bedtime procedures, chronotherapy shifts both the bedtime and wake time later rather than earlier (Piazza et al., 1998; Vriend et al., 2011). More specifically, chronotherapy involves shifting the child's bedtime and wake time on average progressively later every day by set intervals (for e.g., 2 hours in Piazza et al., 1998). The child's bedtime and wake time are shifted until the goal sleep/wake schedule is achieved, based upon an age-appropriate sleep duration and bedtime (Piazza et al., 1998). Importantly the child's typical daytime routine (e.g., eating breakfast, doing schoolwork, lunchtime) is maintained notwithstanding the unconventionality of the sleep/wake schedule (for e.g., the child's bedtime is 2.00pm and wake time is 11.00pm). Chronotherapy is effective for reducing sleep problems in children on the autism spectrum (Piazza et al., 2018) and typically developing adolescents (Okawa et al., 2002). For example, Okawa et al. (2002) investigated the effectiveness of chronobiological treatments (e.g., chronotherapy, melatonin, bright light therapy) in 20 adolescents with delayed sleep phase syndrome, aged 10 to 19 years. Researchers treated one participant with chronotherapy (in addition to melatonin use) and found it was effective for delayed sleep phase syndrome (Okawa et al., 2002).

Sleep Restriction. Sleep restriction is based upon the assumption that spending too much time in bed awake exacerbates sleep problems (Christodulu & Durand, 2004). The procedure involves placing a restriction on the amount of time the child is allowed to spend in bed (Christodulu & Durand, 2004; Durand & Christodulu, 2004), in order to reduce sleep

onset latency, consolidate sleep, and increase homeostatic sleep pressure. Using this procedure, the child's bedtime and wake time are determined based upon 90% of the child's average sleep duration (i.e., if the child sleeps for 8 hours on average, the child is allowed to sleep for 7.2 hours each night) (Christodulu & Durand, 2004; Durand & Christodulu, 2004). If the child does not fall asleep quickly, the child is removed from bed, engages in neutral activities, and is returned to bed when they seem tired. The child's sleep is progressively increased (e.g., around 15 minutes per week) until the goal sleep/wake schedule is reached (Christodulu & Durand, 2004; Durand & Christodulu, 2004). Sleep restriction is an effective treatment for sleep problems in adults (Spielman et al., 1987), however, there is a paucity of research in typically developing children and children on the autism spectrum. Spielman et al. (1987) investigated the effectiveness of sleep restriction in 35 adults (mean age = 46 years), with a history of sleep problems. Researchers found that the sleep restriction procedure improved sleep onset latency, total sleep duration, and sleep efficiency (Spielman et al., 1987).

Scheduled Awakenings. The scheduled awakenings procedure involves parents waking the child before the time that the child typically wakes during the night in order to reduce night wakings and consolidate sleep (Owens et al., 1999). Parents progressively increase the amount of time in-between these scheduled awakenings, with the goal of the child sleeping through the night uninterrupted by night wakings (Owens et al., 1999). In other words, scheduled awakenings are reduced or eliminated following a reduction or elimination in the child's night wakings (Durand, 2002). This procedure is not appropriate for children with settling difficulties (Owens et al., 1999). There is, however, some evidence that it is effective for reducing night wakings in infants and young children (Rickert & Johnson, 1988). To illustrate, one study investigated the effectiveness of scheduled awakenings, in comparison to systematic ignoring and a control condition (Rickert & Johnson, 1988).

Participants were 33 infants and young children aged 6 months to 4.5 years, with at least one night waking each night. Researchers found that the scheduled awakenings procedure reduced night wakings across participants, with further improvements at 6 weeks follow-up (Rickert & Johnson, 1988).

Sleep Hygiene. Sleep hygiene practices that facilitate good quality sleep include a consistent sleep/wake schedule, structured bedtime routine (e.g., have a bath, put on pyjamas, brush teeth, read a story in bed), a sleep-conducive environment (e.g., a comfortable, quiet, dark bedroom), and minimising access to objects that compete with sleep (e.g., toys) or promote hyperarousal (e.g., electronic devices, social media engagement) (Bathory & Tomopoulos, 2017; Blampied, 2013; Mindell et al., 2009; van Deurs et al., 2019). Further, daytime behaviours such as exercise (Galland & Mitchell, 2010), use of stimulants (e.g., caffeine), and levels of exposure to natural light also contribute to sleep quality (McLay, France, Blampied, van Deurs, et al., 2021; van Deurs et al., 2019). Sleep hygiene is an effective and essential component of function-based sleep interventions for children on the autism spectrum (McLay, France, Blampied, van Deurs, et al., 2021). Interventions addressing sleep hygiene practices (e.g., implementing a consistent bedtime routine) have been effective for treating sleep problems in typically developing children (Mindell et al., 2009). More recently, Richdale and Schreck (2019) investigated the sleep and sleep hygiene practices of both children with and without a diagnosis of ASD. Participants were 101 children aged 2 to 5 years: 71 typically developing children, 28 children on the autism spectrum, and 2 children with global developmental delay (Richdale & Schreck, 2019). They found that children with autism and sleep problems slept less than the other children, and electronic device use and bedroom temperature (i.e., too hot or too cool) were associated with poorer sleep (Richdale & Schreck, 2019).

Stimulus Substitutions and Matched Sensory Stimulation. Stimulus substitutions and matched sensory stimulation have also been used as part of function-based sleep interventions for children on the autism spectrum (McLay et al., 2020). Stimulus substitution can be used as part of a faded parental presence procedure. For example, a sleep item can be introduced to replace other stimuli that interfere with the child falling asleep independently (i.e., a soft toy can replace parental presence) (McLay et al., 2020). Matched stimulation can be helpful for sensory modulation in children on the autism spectrum. For example, white noise can be introduced to modulate vocal stereotypy (McLay et al., 2017). White noise can reduce the reinforcement children receive when they engage in vocal stereotypy, and provide matched sensory stimulation i.e., auditory stimulation (McLay et al., 2017).

Social Stories. Social stories are a tool used to provide social information to individuals on the autism spectrum, to help with social and behavioural difficulties (Gray & Garand, 1993). They can be used, for example, to teach skills (e.g., academic skills, functional skills) and socially appropriate behaviour (Gray & Garand, 1993; Kokina & Kern, 2010). Within the context of sleep interventions, social stories use text and pictures to help the child understand the procedure, and model expectations for sleep behaviour based on their current treatment plan (McLay et al., 2017; McLay et al., 2019). There is evidence supporting social stories as an effective visual support that can be used as part of a behavioural sleep intervention for children on the autism spectrum (for e.g., McLay et al., 2017; McLay et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021; van Deurs et al., 2019). To illustrate, McLay et al. (2019) used social stories with children on the autism spectrum to demonstrate the steps comprising the child's bedtime routine, sleep and nighttime expectations, and associated rewards. Social stories were read to the children on a nightly basis before bedtime and as requested by the child (McLay et al., 2019). These were

written based on Carol Gray's guidelines for creating social stories and individualised for each child (Gray, 2010; McLay et al., 2019).

Visual Schedules. Visual schedules have also been used to communicate social behaviour and expectations to children on the autism spectrum in a number of settings (e.g., at home, in the classroom) (Schneider & Goldstein, 2010). Visual schedules typically contain pictures, photographs, and/or words that represent each step in an activity or routine (Delemere & Dounavi, 2018). Parents or adults supporting the child may prompt them to attend to the picture on the visual schedule, immediately before completing the activity or routine. Visual schedules can also be used with adolescents to enhance independence, including to support completion of work-related tasks (Lora et al., 2020). More recently, visual schedules have been used to support the implementation of behavioural interventions for sleep problems in children on the autism spectrum (Delemere & Dounavi, 2018). For example, Malow et al. (2014) incorporated visual schedules in a sleep education intervention delivered to parents. They found that the sleep education intervention reduced sleep onset latency in children on the autism spectrum (Malow et al., 2014).

Gro-Clock™. A Gro-Clock™ is another visual support that has been used within behavioural sleep interventions (McLay et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021; van Deurs et al., 2019). A Gro-Clock™ is a clock-like device that provides a visual aide for sleep/wake times (McLay et al., 2019). The clock face lights up blue and shows a star for bedtime, and then switches to yellow with a picture of a sun to represent wake time (McLay et al., 2019). Gro clocks have been successfully implemented for children on the autism spectrum to provide a discriminative stimulus for sleep time or waking time (McLay et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021; van Deurs et al., 2019).

Consequence-based Modifications. Consequence-based modifications are those that involve modifying contingencies of reinforcement (McLay, France, Blampied, van Deurs, et

al., 2021). This includes elimination or withdrawal of reinforcement (e.g., social attention) for sleep-interfering behaviours as well as the use of reinforcement (e.g., rewards) for sleep-conducive behaviours (Blampied, 2013; Owens et al., 1999). Consequence-based modifications include standard extinction, modified extinction procedures, and the use of rewards (McLay et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021; Weiskop et al., 2001).

Standard Extinction. Extinction targets problem behaviour by removing the reinforcement (e.g., social attention) that is maintaining the behaviour (Owens et al., 1999). This in turn has the effect of reducing the likelihood of the behaviour occurring again in the future (Owens et al., 1999; Weiskop et al., 2005). For example, if the child's sleep problem is maintained by social attention, the parent is instructed to minimise attention in response to sleep-interfering behaviour (France & Blampied, 1999; Owens et al., 1999; Weiskop et al., 2005). Likewise, if the behaviour is maintained by access to a bottle of milk upon waking, then access to this bottle will be eliminated (McLay et al., 2019; Weiskop et al., 2005). There is empirical support for the efficacy of extinction procedures for children on the autism spectrum and typically developing infants (France & Hudson, 1990; Weiskop et al., 2001). For example, Weiskop et al. (2001) published a case study examining the effectiveness of a behavioural intervention for sleep problems in a five-year-old boy on the autism spectrum. Within this study, parents were provided with training in behaviourally based strategies over three sessions (Weiskop et al., 2001). Parent coaching sessions focused on multiple strategies including extinction, reinforcement, implementing a bedtime routine, instruction delivery, and strategies for the parents supporting each other. Researchers found that the intervention improved night wakings, difficulties settling, and unwanted co-sleeping, however, improvements were not demonstrated until the implementation of extinction (Weiskop et al.,

2001). In many studies, extinction has been used in combination with antecedent-based modifications (Vriend et al., 2011).

Modified Extinction. Modified extinction procedures provide a more gradual approach to the withdrawal of reinforcement (Owens et al., 1999). Examples of such procedures include minimal check, graduated extinction, and faded parental presence (Owens et al., 1999). Minimal check involves the parents checking the child quickly and regularly (e.g., every 10 minutes for around 1 minute) during night wakings (Owens et al., 1999; France & Blampied, 2005). Checking includes reassuring the child (e.g., verbally, with touch) and restoring their sleeping position (France & Blampied, 2005). Research shows that the minimal check procedure is effective for treating sleep problems in infants (France & Blampied, 2005). For example, France and Blampied (2005) investigated the effectiveness of minimal check as well as extinction (i.e., ignoring) and faded parental presence for sleep problems in infants. Participants were 15 infants, ranging in age from 6 to 15 months old. Researchers found reduced night wakings with all procedures including minimal check (France & Blampied, 2005). There is, however, a paucity of literature on the effectiveness of minimal check for treating sleep problems in children on the autism spectrum.

Graduated extinction involves gradually decreasing the rate or quality of reinforcement for the problem behaviour (Owens et al., 1999). For example, if the sleep problem is reinforced by parental attention, graduated extinction may involve increasing the duration of time before the parents respond to the child crying (incremental) or reducing the duration of time that the parents interact with the child during sleep onset and night wakings (decremental) (Owens et al., 1999). Research suggests that graduated extinction is effective for reducing bedtime disturbances in children on the autism spectrum (Durand et al., 1996) and night wakings in infants (Lawton et al., 1991). For example, Durand et al. (1996) investigated the effectiveness of a behavioural intervention for treating bedtime disturbances

and night wakings in children with developmental disabilities. Participants were two children on the autism spectrum, one child with an intellectual disability, and one child with Down syndrome. Intervention consisted of graduated extinction in addition to a consistent bedtime routine. Researchers found that the intervention improved bedtime disturbances in two children on the autism spectrum and reduced night wakings in the other two children (Durand et al., 1996).

Another modified extinction procedure involves systematic fading of parental presence (McLay et al., 2019). This entails a number of steps wherein the parent may begin by sitting at the child's bedside and leave the room once the child is asleep (McLay et al., 2019). Parents are instructed to avoid interacting verbally or socially with the child unless there are safety concerns. If the child leaves their bed, parents are asked to return them with minimal interaction. As the procedure progresses, the parent is faded from the room over the course of many nights. This may involve sitting about one meter from the child's bed, sitting two to three meters from their bed, sitting next to the bedroom door, sitting behind the door with their legs in sight, and then sitting outside of the child's room (McLay et al., 2019). The progression between these steps will be dependent on the child's response on previous nights. The faded parental presence procedure draws upon multiple behavioural concepts; there is extinction of the parent's presence and responsiveness (McLay et al., 2019), and there is transfer of stimulus control i.e., discriminative stimuli for sleep onset shift from the parent to the child's bed (Blampied, 2013). Thus, faded parental presence can be classified as an antecedent-based modification as well as a consequence-based modification (McLay et al., 2019). Research supports the effectiveness of the faded parental presence technique for typically developing infants (e.g., Kahn et al., 2020), however, there is scarce evidence for children on the autism spectrum. In one study, McLay et al. (2019) implemented multiple component behavioural interventions for sleep problems in seven children on the autism

spectrum, aged 2 to 4 years. Researchers found improvements across all sleep outcomes for two children, and this was associated with starting the faded parental presence procedure (McLay et al., 2019). However, clear treatment effects were not demonstrated for the other three children that completed the faded parental presence procedure (McLay et al., 2019).

Rewards. Reinforcement is used to increase the likelihood a behaviour will reoccur (Cooper et al., 2020). Positive reinforcement adds something rewarding to the environment and negative reinforcement removes something aversive from the environment (Blampied, 2013). In the context of sleep interventions, reinforcement includes reinforcement of appropriate sleep behaviours, differential reinforcement, and planned access to reinforcement (Owens et al., 1999). For example, positive reinforcement of appropriate sleep behaviours may involve providing a reward (e.g., tangible, attention) immediately after the child wakes if they remain in their bed following night wakings (McLay et al., 2017). Differential reinforcement may involve providing reinforcement for appropriate sleep behaviours and withholding reinforcement for inappropriate sleep behaviours (Owens et al., 1999). Planned access to reinforcement may involve providing opportunities for parental attention or toys prior to bedtime (Owens et al., 1999). There is a paucity of research on reinforcement-based interventions for sleep problems, though rewards have been effective in the treatment of sleep problems in children on the autism spectrum alongside other behavioural interventions (e.g., McLay, France, Blampied, van Deurs, et al., 2021; Moon et al., 2011; Weiskop et al., 2005).

FBA-informed Interventions. FBA is an evidence-based approach to assessment that is used to identify the setting events, antecedents, and consequences, and to develop a hypothesis about the potential function of a behaviour (Blampied, 2013; Brown & Piazza, 1999). FBA is used to inform the development of individualised treatment plans that target these identified variables (Brown & Piazza, 1999; Jin et al., 2013). Common functions for sleep problems in children on the autism spectrum include attention, escape, and tangible

(McLay, France, Blampied, van Deurs, et al., 2021). In some cases, sleep problems are multi-functional and an individualised, FBA-informed intervention will target multiple functions (McLay, France, Blampied, van Deurs, et al., 2021). For example, if a child has frequent night wakings and the functions are to obtain attention (e.g., social attention from parents) and access a tangible item (e.g., bottle), intervention will target these functions by eliminating parental presence and the bottle (McLay et al., 2019).

FBA is often used in assessment and treatment formulation to address a number of challenging behaviours in children on the autism spectrum, for example, aggression, inappropriate vocalisations, noncompliance, tantrums, and self-injurious behaviour (Hanley et al., 2014; O'Reilly et al., 2010; Lodge, 2001). More recently, a growing body of evidence has demonstrated the utility of FBA-informed interventions in the treatment of sleep problems for children on the autism spectrum (Didden et al., 2002; McLay et al., 2017; McLay et al., 2019; van Deurs et al., 2019). For example, McLay et al. (2021) collated and analysed 41 cases in which FBA-informed sleep interventions were implemented (McLay, France, Blampied, van Deurs, et al., 2021). All participants were children and adolescents on the autism spectrum. Researchers found that FBA-informed interventions improved sleep problems for 38 out of the 41 participants (McLay, France, Blampied, van Deurs, et al., 2021).

Post-extinction Response Bursts (PERBs)

Despite the overwhelming empirical support, many parents report reservations about the use of extinction procedures (Gradisar et al., 2016). This is typically due to the occurrence of a post-extinction response burst (PERB) and the possibility of child distress (Gradisar et al., 2016; Owens et al., 1999). A PERB is a transient increase in the frequency, duration or intensity of a problem behaviour following the removal of reinforcement (Lerman & Iwata, 1995; Owens et al., 1999). For instance, when a child is accustomed to receiving

parental attention for a specific behaviour, and the parent's attention is suddenly removed, there may be a temporary increase in the child's intensity and duration of challenging behaviour in order to reinstate parental attention (France & Blampied, 1999). This can manifest in the child crying out louder (intensity) or for a longer period (duration) (Turner & Johnston, 2012). For this reason, parents may experience difficulty adhering to the procedure, resulting in the abandonment of a treatment plan (Gradisar et al., 2016). Research has been conducted investigating the effect of extinction-based procedures (e.g., graduated extinction) on parent-child attachment and the development of emotional and behavioural problems (Gradisar et al., 2016; Price et at., 2012). Researchers reported no evidence of negative long-term consequences on these outcomes, nonetheless, in many cases, these beliefs persist (Gradisar et al., 2016; Price et at., 2012).

Principles of Least Restriction and Minimal Sufficiency

In this thesis, the principles of least restriction and minimal sufficiency guide the research design and methodology. This is accomplished using FBA to inform each participant's individualised intervention. These principles are discussed below in relation to sleep interventions.

Least Restriction

Least restrictive is the term used to refer to the process in which procedural, relational, and physical restrictions are minimised and implemented only if needed (Sustere & Tarpey, 2019). From a behavioural perspective, restrictiveness can be defined in a number of ways, including perceived acceptability of the intervention, the loss of liberty as a consequence of implementing the intervention, the level of irreversibility of treatment effects, and the amount of time, risk, stress, and discomfort associated with the intervention (Johnston & Sherman, 1993). In the context of this research, it is likely that the least restrictive interventions are antecedent-based modifications including modification to

sleep/wake schedules (van Deurs et al., 2021). Modification to antecedent variables are likely less restrictive as they are relatively simple for parents to implement and are not associated with high levels of distress. By contrast, the most restrictive interventions are likely to be those which involve modification to contingencies of reinforcement (i.e., extinction-based procedures) because of the possibility of a PERB and the associated distress for both the parent and child (Gradisar et al., 2016; Owens et al., 1999). In this thesis, the term least restrictive is used to refer to the least restrictive of the available alternative treatment components.

In Part two of this thesis, restrictiveness is measured post-treatment through parent-report ratings of acceptability. Each child's individualised intervention will start with modification to sleep/wake schedules (i.e., the less restrictive treatment component). It is imperative to explore parental perceptions of treatment acceptability to ensure this intervention provides a less restrictive intervention approach. This is important because parental adherence to a treatment plan will be higher with a less restrictive alternative; thus, there is a higher chance of successfully treating the child's sleep problems which would otherwise persist overtime (van Deurs et al., 2021).

Minimal Sufficiency

Minimal sufficiency is defined as "the selection of interventions aimed at achieving a meaningful clinical outcome in the most cost-effective and time-efficient manner" (Sanders et al., 2014, p. 339). In the context of this research, minimal sufficiency refers to implementing treatment components that are effective for treating the child's sleep problems, without implementing more components than necessary (i.e., a treatment component will only be introduced if sleep problems did not improve with the current procedure). This is important because it means that if sleep problems improve with modification to sleep/wake schedules or other antecedent-based modifications, families have received an effective

intervention that avoided the use of consequence-based modifications. As previously mentioned, most behavioural interventions for sleep problems are multi-component (i.e., comprised of modification to both antecedents and consequences), meaning that it is difficult to determine which modifications alone are minimally sufficient. In Part two of this thesis, treatment components are introduced from less restrictive to most restrictive and only implemented, if necessary, to provide the family with a minimally sufficient approach. This treatment hierarchy starts with modification to sleep/wake schedules, followed by antecedent-based modifications, and then consequence-based modifications.

Rationale for this Thesis

The summarised literature demonstrates the efficacy of behavioural interventions for treating sleep problems in children on the autism spectrum. Many studies included both modification to antecedents and consequences thus, making it difficult to isolate the most effective, minimally sufficient, and acceptable treatment component(s) for the child and family. This is important because providing a minimally sufficient, less restrictive alternative has potential to increase parental adherence and therefore the likelihood of effectively treating sleep problems (van Deurs et al., 2021). The significance of a minimally sufficient, less restrictive sleep intervention may extend beyond the child's sleep problems, as there can be collateral benefits for the child, their parents, and other family members (McLay, France, Blampied, Hunter, et al., 2021).

The aim of this research is to identify whether modification to sleep/wake schedules and other antecedent-based modifications (i.e., modifications which are less restrictive and minimally sufficient) alone, are effective in reducing sleep problems in children on the autism spectrum. To achieve this, this thesis has been divided into two parts. Part one involves a systematic review of the effectiveness of procedures that involve modification to sleep/wake schedules and antecedent-based modifications for treating sleep problems in

children on the autism spectrum. The following research questions were used to guide the design of Part one:

1. Are modifications to sleep/wake schedules alone, effective in reducing sleep problems in children on the autism spectrum?
2. Are antecedent-based modifications alone, effective in reducing sleep problems in children on the autism spectrum?

Part two will use a single-case AB design in order to separately evaluate: (a) the effectiveness of antecedent-based modifications (including modifications to sleep/wake schedules) and consequence-based modifications for treating sleep problems in children on the autism spectrum. This will involve a staggered approach to intervention in which each treatment component will be introduced in phases, beginning with modification to sleep/wake schedules, followed by antecedent-based modifications and then consequence-based modifications; (b) the maintenance of treatment effects at short-term and long-term follow-up; (c) parental perceptions of treatment acceptability; and (d) the impact of reduced sleep problems on the children's wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parental mental health, sleep, and relationship quality.

Part two was designed to answer the following four research questions:

1. Are modifications to sleep/wake schedules and antecedent-based modifications alone or in combination with one another, effective in reducing sleep problems in children on the autism spectrum?
2. How will parents of children on the autism spectrum rate the acceptability of modifications to sleep/wake schedules, antecedent-based modifications, and consequence-based modifications?

3. Is there an impact of reduced sleep problems on the children's wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parental mental health, sleep, and relationship quality?
4. Will any reduction in sleep problems be maintained at short-term follow-up (STFU) and long-term follow-up (LTFU)?

Chapter Two: Systematic Review (Part One)

In this chapter, the findings of the systematic review entitled "Systematic Review of the Effect of Modification of Antecedents in the Treatment of Sleep Problems Among Children on the Autism Spectrum" are briefly summarised (Ford et al., 2021). This systematic review was first published in (*Advances in Neurodevelopmental Disorders,* 1-17*)* by Springer Nature (see Appendix A). As author of this thesis, I (KF) led this systematic review and wrote the original manuscript.

The purpose of this systematic review was to appraise the evidence for modification to antecedent variables for treating sleep problems in children on the autism spectrum. This would also highlight the degree to which modification of antecedents have been implemented without the use of consequence-based modifications (e.g., extinction, modified extinction procedures, use of rewards) in the treatment of sleep problems for children on the autism spectrum. As the overarching aim of this thesis is to identify whether modification to antecedent variables including modification to sleep/wake schedules are effective in treating sleep problems, it was necessary to appraise the current corpus of research while implementing selected procedures with a group of participants.

The systematic review (Part one) aimed to identify whether modification to sleep/wake schedules and other antecedent-based modifications alone, are effective in reducing sleep problems in children on the autism spectrum. The findings showed that modification to antecedent variables can be effective for treating a number of sleep problems in this group. Improvements were reported for modification to sleep/wake schedules (e.g., faded bedtime, sleep restriction, chronotherapy, bedtime scheduling, scheduled awakenings) and other antecedent-based modifications (e.g., sleep hygiene modifications, stimulus control). Of note, faded bedtime was implemented in 5/12 studies and improved sleep

problems for all participants. Overall, modification to antecedent variables have been effective alone, for treating sleep problems for children on the autism spectrum.

The findings of Part one provide rationale for Part two of this thesis. Part two will also aim identify whether modification to sleep/wake schedules and other antecedent-based modifications alone, are effective in reducing sleep problems in children on the autism spectrum. In Part two, a single-case AB research design will be used in order to separately investigate the effectiveness of these modifications, as well as consequence-based modifications if necessary. A single-case AB research design will allow for treatment components to be introduced in phases. This staggered approach to intervention will start with the less restrictive treatment component i.e., modification to sleep/wake schedules, followed by antecedent-based modifications and then consequence-based modifications if needed. Part two will also aim to: (1) explore how parents of children on the autism spectrum will rate the acceptability of modification to sleep/wake schedules and other antecedent-based modifications, (2) examine the effect of an improvement in children's sleep on child wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parental mental health, sleep, and relationship quality, and (3) examine whether reductions in sleep problems are maintained at STFU and LTFU.

Chapter Three: Method (Part Two)

In this chapter, the general method for all participants is outlined. Next, the specific details of clinical cases and individualised interventions are presented.

Sleep Research Team

This research was conducted as part of a wider research project, the Good Nights Programme, investigating the effectiveness of FBA-informed interventions for sleep problems in children and adolescents on the autism spectrum, and the collateral benefits of such interventions on children's daytime behaviour and parent and child well-being. The focus of this thesis is on modification to antecedent variables including modification to sleep/wake schedules and was prospectively established as a discrete project within the Good Nights Programme. This programme is led and supervised by two Associate Professors and a Professor at the University of Canterbury, and comprised of registered psychologists, intern psychologists, and postgraduate students. As author of this thesis, I (KF) was part of this team. I was the main clinician for two of the cases discussed in this study. I completed this academic and clinical work under the supervision of senior team leaders. Other clinicians in the study team completed the clinical work for the remaining four participants, and I collated the data, coded video recordings, calculated interobserver agreement and treatment fidelity, scored psychometric instruments, and analysed the data for these participants as they met inclusion criteria for this study.

Ethics and Participant Consent

This research has received ethical approval from the University of Canterbury Human Ethics Committee (HEC 2018/47). Parents signed a written consent form to participate in the study. Consent or assent was obtained from child and adolescent participants depending upon chronological age, developmental stage, and/or cognitive ability. Jack, Mason, and Luna signed a children/young person's consent form, and Hannah signed an assent form. Further

consent was given by two families for nighttime video recording. Three families did not consent to video recording and consented to use of an actigraph as an alternative measure. One family did not consent to either video recording or actigraph use. A copy of the consent and information forms given to all participants are attached (see Appendix B-J).

Research Design

A single-case AB design was employed to investigate treatment effects. Within the single-case design, participants acted as their own controls and were randomly assigned a baseline [A] length of 1, 2, or 3 weeks. Intervention [B] was introduced using a staggered approach, starting with modification to sleep/wake schedules, modification to antecedent variables, and then consequence-based modifications. Intervention phases introduced following sub-phase one [B, P1] were labelled P2, P3, or P4 and evident on the graph with a phase change line. Treatment effects for each intervention phase and follow-up phases were assessed by comparing the change from baseline and with the previous intervention phase using visual analysis.

Participants

Recruitment

Participants were recruited throughout New Zealand via organisations and service providers that support children on the autism spectrum and their families (e.g., Explore, Autism New Zealand, Child Development Service) or via self-referral. An advertisement was sent to organisations and service providers that included information about the study and how to become involved. Families who contacted the research team either directly or by referral, were provided with further information and screened for eligibility.

Screening and Confidentiality

Families interested in participating completed a screening assessment over the phone with a member of the study team. The purpose of the screening phone call was to ascertain

whether they were eligible for inclusion in the study. The researchers started the phone call with a discussion about confidentiality and limits of confidentiality. Families deemed eligible were then provided with relevant information sheets and consent forms to return to the study team if they were interested in proceeding.

Inclusion/Exclusion Criteria

Participants were eligible for inclusion if they were: (a) between the ages of 3 and 21 years; (b) meet diagnostic criteria for ASD as verified by a registered psychologist, psychiatrist and/or paediatrician (APA, 2013); (c) had parent-reported difficulties with sleep onset and/or maintenance (e.g., sleep onset delay, frequent or prolonged night wakings, early morning wakings); and (d) parent(s) and children/adolescents (henceforth referred to as children) were willing and able to adhere to the treatment plan and research procedures. Participants were excluded from the study if they had a co-occurring medical condition that may have comprised their ability to participate in the programme safely (e.g., epilepsy, obstructive sleep apnoea, or suicidal ideation).

Participant Characteristics

Participants were four males and two females on the autism spectrum with a mean age of 10 years (range = 3 to 19 years) at the start of intervention. Four of the six participants were NZ European, one participant was NZ European/Samoan, and one participant was Chinese/Vietnamese. All of the participants had been diagnosed with ASD by a registered psychologist, psychiatrist and/or paediatrician, and experienced parent-reported sleep disturbances, corroborated by sleep diary and/or video recordings. Four participants took melatonin regularly before sleep onset during the intervention. The other two participants had been prescribed melatonin but took it occasionally or rarely. Participant characteristics are summarised in Table 1. Participant's names have been replaced with pseudonyms in order to maintain confidentiality.

Table 1

Summary of Participant Characteristics

Name	Age (Y-M)	Gender	Ethnicity	Diagnoses (DSM-5)	Vineland-3 Receptive/Expressive/ Written Communication Age Equivalent (Y-M)	Sleep problems	Medication
Jack	19-4	Male	NZ European	ASD, intellectual disability	3:4 6:9 8:9	SOD, TST, irregular sleep/wake schedule	Melatonin
Hannah	5-8	Female	NZ European	ASD	2:8 4:0 <3:0	Freq. and duration of NWs, sleep dependencies (milk, parental presence), irregular sleep/wake schedule	Melatonin
Luna	12-7	Female	NZ European/ Samoan	ASD	-	SOD, irregular sleep/wake schedule	
Mason	13-5	Male	NZ European	ASD, Twice Exceptional	11:0 11:6 9:4	SOD, freq. and duration of NWs, irregular sleep/wake schedule	
Liam	8-1	Male	NZ European	ASD, ADHD	1:10 1:9 3:10	SOD, irregular sleep/wake schedule	Melatonin
Ethan	3-9	Male	Chinese/Vietnamese	ASD	0:3 0:3 <3:0	Irregular sleep/wake schedule	Melatonin

Note. ASD= autism spectrum disorder, ADHD= attention-deficit/hyperactivity disorder, DSM-5= Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, SOD= sleep onset delay, M= months, NWs= night wakings, TST= total sleep time, Y=years.

Setting

Interventions were implemented in the family home by each child's parent(s), with support from the clinician and study team. For Jack, the intervention was implemented with occasional parental support i.e., Jack was in daily contact with the researcher, adhered to the treatment plan, and asked his parents for help if needed. In-person meetings (i.e., to complete the clinical interview, see below) with the five local families occurred in either the family home or in the Pukemanu Centre clinic located at the University of Canterbury. For the

remaining family, the clinical and child interviews were completed via Zoom. Following the clinical interview, contact with families was primarily maintained via email, phone (call or text), or video call (Skype, Zoom) unless in-person support was needed (e.g., to assist with completing psychometric forms, to teach and practice strategies such as diaphragmatic breathing).

General Materials

Parent- and/or self-report sleep diaries, video equipment, and/or actigraphs were used to collect data and measure change in dependent variables across study phases. Visual supports (e.g., Gro-Clocks™, social stories) were also implemented for some children as part of their individualised treatment plan and are described below (see *Individual Cases*).

Sleep Diaries

Parent- and/or self-report sleep diaries were completed each night during baseline, intervention, and follow-up. Parent-report sleep diaries alone were used for three participants (Hannah, Liam, and Ethan); Jack and Mason completed self-report sleep diaries; and both parent- and child-report diaries were completed by Luna and her parents. Parent-report sleep diaries included information about the sleep setting, the time their child was put to bed, the frequency of curtain calls, the child's behaviour during curtain calls and parent responses to these behaviours, time of sleep onset, the time and duration of each night waking, a description of their child's behaviour and their own response during each awakening, morning wake time, and other variables that may have affected their child's sleep (e.g., illness). The self-report sleep diaries were formatted differently, to enable reporting in a developmentally appropriate manner. Jack, Mason, and Luna recorded what time they went to bed, duration of sleep onset, how easily they fell asleep (i.e., "did you fall asleep": 1= easily, 2= after some time, 3=with difficulty), the frequency, timing and duration of night wakings, total sleep duration, reasons for sleep disturbance, how they felt in the morning (i.e.

"how do you feel this morning": 1= refreshed, 2= okay, 3= tired), and morning wake time. In addition, Jack was asked to record the activities that he engaged in, in the lead up to bedtime and if he took melatonin that night. A copy of the standard parent- and self-report sleep diaries are attached (see Appendix K-M). These standard diaries may have been altered to suit the individual's needs.

Videosomnography

Infrared video recording equipment (Swann Advanced-Series DVR4-1200, D-Link HD Cloud, or TP-Link Tapo camera) was used for Hannah and Liam. These cameras were set up in a discrete location facing the bed. Video footage was recorded from the time the children were put to bed until the time they woke up the next morning, to provide an additional measure of dependent variables and sleep-related behaviour. Video recording was downloaded and coded by the researcher for 25% of the nights across study phases, to triangulate the data, measure the reliability of parent-report sleep dairies, and calculate inter-observer agreement (IOA).

Actigraphs

An actigraph is a small watch-like device, worn on a person's wrist. It is able to differentiate between sleep and wake states and provides information on SOL, wake after sleep onset, TST, and sleep efficiency (SE). GT9X-BTActiGraph devices were worn by three participants (Jack, Luna, and Mason) for at least 30% of nights, across study phases. Actigraph data were downloaded by the researchers using ActiLife 7 desktop analysis software, for the purpose of triangulation, measuring the reliability of parent- and/or self-report, and calculating IOA. Jack and Mason wore the actigraph most nights of the study, however, there were technical issues with the device and only 10 and 3 nights provided usable data, respectively.

Dependent Variables

Dependent variables were measured for each participant based upon their unique sleep problems and the treatment goals. Sleep diaries, video equipment, and/or actigraphs were used to collect this data. Dependent variables in this study include asleep, awake, sleep onset latency (SOL), frequency of night wakings (NWs), duration of NWs, and total sleep time (TST). The following definitions were conceptualised by the study team and have been used widely in research published by the study team (for e.g., McLay, France, Blampied, van Deurs, et al., 2021).

Asleep

Asleep was defined as the participant lying down still in bed with closed eyes and no voluntary movements or vocalisations associated with wakefulness.

Awake

Awake was defined as the participant engaging in voluntary movements or vocalisations indicative of wakefulness with their eyes open.

SOL

SOL was defined as the duration of time measured in minutes between the child going to bed (i.e., being bid goodnight) and falling asleep.

NWs

NWs were defined as an awakening that occurred between the child's set bedtime and wake time and lasted for at least five minutes and/or was signalled. The frequency and duration of NWs were recorded in sleep diaries.

TST

TST was defined as the duration of time in minutes between the child falling asleep and waking up, minus the duration of night wakings.

Assessment Measures

Clinical Interview

A Child and Family intern or registered psychologist conducted a clinical interview with the parents of each participant. Parents were asked open questions following the clinical interview format used in the Child and Family Psychology Programme and the Pukemanu Centre clinic. The standard format covers introductions, consent and confidentiality, referral information, diagnostic/medical information, the child's strengths and interests, presenting problems, history of presenting problem (including previous support), parental goals for intervention, parental mental health and wellbeing, impact on family functioning, and developmental history. The Sleep Assessment Treatment Tool (SATT) was also administered during the clinical interview. The clinician demonstrated the correct way to complete sleep diaries and supported parents to complete the psychometric measures if needed.

Child Interview

The clinician also conducted a child interview with Jack and Mason. Jack was asked questions about his current sleep difficulties, bedtime routine, previous strategies that he has tried, what has been helpful or unhelpful for him, and goals for the study. Mason was asked about his perception of his sleep problems, bedtime routine, thoughts and feelings related to his sleep problems, and goals for the study.

SATT (Hanley, 2005)

The SATT is an interview tool that is used to inform FBA. It consists of open-ended questions about the type of presenting sleep problems, history of sleep problems, parental goals for the child's sleep, bedtime routine, sleep environment, sleep-interfering behaviours, and the antecedents and consequences underlying sleep problems (Hanley, 2005). The SATT has been used widely in research with children on the autism spectrum (Jin et al., 2013; McLay et al., 2017, McLay et al., 2019; van Deurs et al., 2019).

FBA Measures

FBA was completed using data collected from the clinical interview, the SATT, sleep diaries, and/or video recording. FBA was used to identify the antecedents and consequences of the sleep problem and to develop a hypothesis regarding the function(s) of the participant's sleep problem(s). This data was used by clinicians and the study team to formulate an individualised sleep intervention.

Questions About Behavioral Function (QABF; Matson & Vollmer, 1995). The QABF is a 25-item instrument, designed to assess the function of problem behaviour (Paclawskyj et al., 2000). It is used to obtain parent-report responses on five subscales: Attention, Escape, Non-social, Physical, and Tangible. Parents are instructed to consider how often the child engages in the behaviour, and rate items on a four-point scale: 0 ("never"), 1 ("rarely"), 2 ("some"), and 3 ("often") (Paclawskyj et al., 2000). The sum of item scores provides the five subscale scores, and higher subscale scores indicate the function of the behaviour. The QABF has good psychometric properties, for example, good validity and reliability in a group of children and adolescents on the autism spectrum (Healy et al., 2013).

Sleep Outcome Measures

The following sleep measures were administered at baseline and post-treatment to assess sleep problems and associated behaviours, sleep hygiene, and sleep quality. Measures were selected for each participant based on their chronological age and developmental stage.

Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000)

The CSHQ is a 33-item, parent-report instrument, designed to measure the frequency of behaviours associated with sleep problems in children aged 4 to 10 years (Owens, Spirito, & McGuinn, 2000). It includes eight subscales: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness. Parents are instructed to consider the past week or a recent typical

week for their child and rate each item on a three-point scale: 0 to 1 (“rarely”), 2 to 4 (“sometimes”), and 5 to 7 (“usually”) times per week (Owens, Spirito, & McGuinn, 2000). Parents are also asked whether the behaviours are a problem, and instructed to circle “yes”, “no”, or “not applicable (N/A)”. The sum of item scores provides a total CSHQ score in addition to the eight subscale scores. Higher CSHQ total and subscales scores indicate worse sleep problems, and the total score clinical cut-off is ≥41. The CSHQ has demonstrated good psychometric properties, for example, clinical and control subjects were identified using the cut-off score (≥41) with a sensitivity of 0.80 and specificity of 0.72 (Owens, Spirito, & McGuinn, 2000). In addition, test-retest reliability in community subjects for the subscales was 0.62 to 0.79, and the internal consistency of the CSHQ total score was 0.78 for the clinical group and 0.68 for the community group (Owens, Spirito, & McGuinn, 2000). The CSHQ has been used in research to assess sleep problems in both typically developing children and children on the autism spectrum (Lambert et al., 2016; Hoffman et al., 2006), and achieved a “well-established” rating according to the American Psychological Association Division 54 Evidence-Based Assessment (EBA) Task Force criteria (Lewandowski et al., 2011).

Sleep Self-Report (SSR; Owens, Spirito, McGuinn, & Nobile, 2000)

The SSR is a 26-item instrument, designed to measure the frequency of behaviours associated with sleep problems in children aged 7 to 12 years (Owens, Spirito, McGuinn, & Nobile, 2000). It was developed to measure similar sleep problems to the CSHQ thus, many SRR items correspond to CSHQ items. It is used to obtain self-report responses on six subscales: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, and Daytime Sleepiness. Children are instructed to think about their sleep, answer “yes” or “no” questions, and then rate items on a three-point scale: 0 to 1 (“rarely”), 2 to 4 (“sometimes”), and 5 to 7 (“usually”) times per week. The sum of item scores provides a total

SSR score in addition to the six subscale scores. Higher SSR total and subscales scores indicate worse sleep problems. The SSR has demonstrated good psychometric properties, for example, in a non-clinical sample of school children the internal consistency of the total SSR score was 0.88 (Owens, Spirito, McGuinn, & Nobile, 2000). The SSR has been used to assess sleep problems in children on the autism spectrum as well as typically developing children in research settings (for e.g., Paavonen et al., 2008; Richdale & Baglin, 2015), and achieved an "approaching well-established" rating according to the American Psychological Association Division 54 EBA Task Force criteria (Lewandowski et al., 2011).

Adolescent Sleep Hygiene Scale (ASHS; LeBourgeois et al., 2005)

The ASHS is a 33-item self-report instrument, designed to measure sleep hygiene practices in adolescents aged 12 to 19 years (LeBourgeois et al., 2005; Storfer-Isser et al., 2013). It is used to obtain the adolescent's responses on eight subscales: Physiological Factor, Behavioural Arousal Factor, Cognitive/Emotional Factor, Sleep Environment Factor, Sleep Stability Factor, Daytime Sleep Factor, Substances Factor, and Bedtime Routine Factor. Adolescents are instructed to consider their sleep habits over the past month and rate the frequency of each item on a six-point scale: "never" (0%), "once in a while" (20% of the time), "sometimes" (40%), "quite often" (60%), "frequently if not always" (80%), and "always" (100%). In scoring the ASHS, responses for each item are given the following points (opposite for reverse-coded items): never (6), once in a while (5), sometimes (4) quite often (3) frequently if not always (2), and always (1). The sum of items divided by the total number of items (the mean) is calculated for each subscale, and the mean of the eight subscale scores provides a total ASHS score. Higher ASHS and subscale scores indicate the adolescent is engaging in better sleep hygiene practices (LeBourgeois et al., 2005). The ASHS has demonstrated satisfactory psychometric properties, for example, the internal consistency of the ASHS total score was 0.84 in a community sample of adolescents (Storfer-

Isser et al., 2013). The ASHS has been used in research to assess sleep problems in typically developing children and children on the autism spectrum (for e.g., Goldman et al., 2017), and achieved an "approaching well-established" rating (Lewandowski et al., 2011).

Adolescent Sleep Wake Scale – short version (ASWS; Essner et al., 2015)

The ASWS is a 28-item self-report instrument, designed to measure subjective sleep quality in adolescents aged 12 to 18 years (Essner et al., 2015). It is used to obtain the adolescent's responses on five subscales: Going to Bed, Falling Asleep, Maintaining Sleep, Reinitiating Sleep, and Returning to Wakefulness. Adolescents are instructed to consider their sleep over the past month, and rate the frequency of each item on a six-point scale: "never", "once in a while", "sometimes", "quite often", "frequently if not always", and "always". To score the ASWS, responses for each item are given the following points (opposite for reverse-coded items): never (6), once in a while (5), sometimes (4) quite often (3) frequently if not always (2), and always (1). The sum of item scores provides a total ASWS score in addition to the five subscale scores. Higher ASWS total and subscales scores indicate better sleep quality (Essner et al., 2015). The ASWS has demonstrated good psychometric properties, for example, in a community sample the internal consistency of the ASWS total score ranged from 0.80 to 0.86 (Essner et al., 2015). The ASWS has been used to assess sleep problems in research settings (for e.g., Palermo et al., 2008), and achieved an "approaching well-established" rating (Lewandowski et al., 2011).

Measure of Communication

Vineland Adaptive Behaviour Scales- Third Edition (Vineland-3; Sparrow et al., 2016)

The Vineland-3 is a comprehensive instrument, designed to measure adaptive functioning in individuals aged 0 to 90 years (Sparrow et al., 2016). It measures parent-report responses across four domains: Communication, Daily Living Skills, Socialisation, and Motor Skills. Parents are instructed to consider what the person is able to do on their own

without support or reminders and rate the items on a three-point scale: 0 (“never”), 1 (“sometimes”), 2 (“usually or often”). Parents are also instructed to check the box beside the three options if they were uncertain about the frequency and provided their best guess. The Vineland-3 has an overall Adaptive Behaviour Composite score as well as domain standard scores. All domain standard scores and the Adaptive Behaviour Composite score have a mean of 100 and standard deviation of 15 (Sparrow et al., 2016). In this study, only the Communication sub-domains of the Vineland-3 were administered at baseline. These provided information about each child’s receptive, expressive, and written communication, to ensure that interventions were developmentally appropriate.

Measures of Child Wellbeing, Behaviour, and Functioning

The following measures were administered at baseline and post-treatment, to assess change in children’s wellbeing, daytime behaviour, ASD symptomatology and quality of life.

Gilliam Autism Rating Scale, Third Edition (GARS-3; Gilliam 2013)

The GARS-3 is a 58-item instrument, designed to assess autism symptomatology in children and adolescents aged 3 to 22 years (Gilliam, 2013). The items are based on DSM-5 criteria (APA, 2013) for ASD. It is used to obtain parent-report responses on six subscales: Restrictive/Repetitive Behaviours, Social Interaction, Social Communication, Emotional Responses, Cognitive Style, and Maladaptive Speech. Parents are instructed to consider how well the item describes their child’s typical behaviour, and rate items on a four-point scale: 0 (“not at all like their child”), 1 (“not much like their child”), 2 (“somewhat like their child”), and 3 (“very much like their child”). The sum of the six subscale scores provides the Autism Index Score which indicates the probability (Unlikely, Probable, and Very Likely) and severity level (Level 1, 2, or 3) of ASD. The GARS-3 has good psychometric properties, for example, in individuals diagnosed with ASD the internal consistency of the Autism Index was 0.93 (Gilliam, 2013).

The Achenbach System of Empirically Based Assessment (ASEBA; Achenbach & Rescorla, 2000, 2001)

The ASEBA is a family of instruments designed to measure emotional and behavioural problems in children and adolescents (Achenbach & Rescorla, 2000, 2001). In this research, two versions of the Child Behavior Checklist (CBCL 1.5-5, 6-18) and the Youth Self-Report (YSR) were used to obtain parent-and/or self-report responses on syndrome scales and DSM-orientated Scales. The CBCL 6-18 and YSR syndrome scales measure eight areas: Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, Rule Breaking Behaviour, and Aggressive Behaviour. The CBCL 1.5-5 syndrome scales measure seven areas: Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Attention Problems, Aggressive behaviour, and Sleep Problems. Parents and/or adolescents are instructed to rate items on a three-point scale: 0 ("not true"), 1 ("somewhat or sometimes true"), and 2 ("very true or often true"). The sum of item scores provides an overall Total Score as well as the Syndrome and DSM-orientated Scale Scores. The Syndrome Scales are also divided into two broadband scales of Internalising and Externalising Scores. All scores are converted to T scores to be interpreted in the Normal Range, Borderline Range, or Clinical Range (Achenbach & Rescorla, 2000, 2001). The ASEBA is well-established and has demonstrated good to excellent psychometric properties in children on the autism spectrum (Pandolfi et al., 2012). In a sample of children with ASD aged 6 to 18 years, reliability of syndrome scales ranged from 0.76 to 0.94 (mean = 0.85) for example (Pandolfi et al., 2012).

Multidimensional Anxiety Scale for Children, Second Edition (MASC 2; March, 2012)

The MASC 2 is a 50-item instrument, designed to measure symptoms associated with anxiety disorders in children and adolescents aged 8 to 19 years (March, 2012). It is used to obtain parent- and/or self-report responses on six scales: Separation Anxiety/Phobias,

Generalized Anxiety, Social Anxiety, Obsessions and Compulsions, Physical Symptoms, and Harm Avoidance. Raters are instructed to consider how often each item has been true for the child/adolescent recently and rate the items on a four-point scale: 0 (“never”), 1 (“rarely”), 2 (“sometimes”), and 3 (“often”). The sum of item scores provides a Total Score in addition to the six subscale scores. Higher Total Scores indicate higher levels of anxiety, and a higher number of elevated *T* Scores on three subscales (Separation Anxiety/Phobias, Generalized Anxiety and Social Anxiety) increases the Anxiety Probability Score (March, 2012). The MASC 2 has demonstrated acceptable to excellent psychometric properties in children on the autism spectrum, for example, the internal consistency of the MASC 2 Total Score was 0.92 for parents and 0.90 for youth, with subscale scores ranging from 0.70 to 0.90 for parents and 0.69 to 0.80 for youth (Kaat & Lecavalier, 2015). The original MASC has been used in research with children on the autism spectrum (Storch et al., 2013).

Paediatric Quality of Life Inventory (PedsQL; Varni et al., 1999)

The PedsQL is a 23-item instrument designed to measure health-related quality of life in children and adolescents aged 2 to 18 years (Varni et al., 1999). It is used to obtain parent- and child self-report responses across four domains of functioning: physical, emotional, social, and school. Raters are instructed to consider how much of a problem each item has been for the child/adolescent over the past month, and rate items on a five-point scale: 0 (“never”), 1 (“almost never”), 2 (“sometimes”), 3 (“often”), and 4 (“almost always”). Items are reverse scored and transformed on a scale of 0 to 100. The sum of transformed item scores divided by the number of items provides a Total Score in addition to the four subscale scores. Higher scores indicate the child has a better health-related quality of life. The PedsQL has demonstrated good to excellent psychometric properties, for example, the internal consistency of the Total Score was 0.88 for child-report and 0.90 for parent-report in a paediatric sample of healthy children and patients (Varni et al., 1999). The PedsQL has been

used in research with children and adolescents on the autism spectrum (e.g., Shipman et al., 2011).

Repetitive Behaviour Scale- Revised (RBS-R; Bodfish et al., 1998, 2000)

The RBS-R is a 43-item instrument, designed to measure repetitive behaviour in children, adolescents, and adults on the autism spectrum (Bodfish et al., 2000). It is used to obtain parent-report responses on six subscales: Stereotyped Behaviour, Self-Injurious Behaviour, Compulsive Behaviour, Routine Behaviour, Sameness Behaviour, and Restricted Behaviour. Parents are instructed to consider how much of a problem each behaviour has been for the person over the past month, and rate items on a four-point scale: 0 ("does not occur"), 1 ("mild problem"), 2 ("moderate problem"), and 3 ("severe problem") (Bodfish et al., 2000). The RBS-R has acceptable to good psychometric properties, for example, the internal consistency was at least 0.72 for all subscales in a sample of preschool children on the autism spectrum (Mirenda et al., 2010).

Measures of Parental Wellbeing

The following measures were completed by parents at baseline and post-treatment, to assess changes in parental mental health, sleep, and relationship quality.

Pittsburgh Sleep Quality Index (PSQI; Buysse et al., 1989)

The PSQI is a 10-item instrument, designed to measure subjective sleep quality (Buysse et al., 1989). It is used to obtain parent-report responses on seven subscales: Subjective Sleep Quality, Sleep Latency, Sleep Duration, Sleep Efficiency, Sleep Disturbance, Use of Sleep Medication, and Daytime Dysfunction. Raters are instructed to consider their sleep habits over the past month, answer open-ended questions, and rate items on a four-point scale: 0 ("not during the past month"), 1 ("less than once a week"), 2 ("once or twice a week"), and 3 ("three or more times a week"). The PSQI is scored by calculating the subscale scores on a scale of 0 to 3. The sum of the seven subscale scores provides the

Global PSQI Score. Higher Global PSQI Scores indicate worse sleep quality. The PSQI has demonstrated good psychometric properties, for example, “good” sleepers and “poor” sleepers were identified using a cut-off score >5 with a sensitivity of 89.6% and specificity of 86.5%. In addition, test-retest reliability was 0.83, and the internal consistency was 0.84 for the subgroup that completed the PSQI twice over time (Buysse et al., 1989).

Depression, Anxiety and Stress Scale (DASS-21; Lovibond, & Lovibond, 1995)

The DASS-21 is a 21-item instrument, designed to measure symptoms of depression, anxiety, and stress (Lovibond & Lovibond, 1995). It is used to obtain parent-report responses on three axes: Depression, Anxiety, and Stress. Raters are instructed to consider how much the items have applied to them over the past week, and rate items on a four-point scale: 0 (“never”), 1 (“sometimes”), 2 (“often”), and 3 (“almost always”). The sum of seven items provides each axes score, and these scores have different cut-off points to define scores as Normal, Mild, Moderate, Severe, and Extremely Severe (Lovibond & Lovibond, 1995). The DASS-21 has demonstrated good psychometric properties in adults across a number of cultures (Norton, 2007).

Relationship Quality Index (RQI; Norton, 1983)

The RQI is a 6-item instrument, designed to measure relationship quality among married and cohabitating couples (Norton, 1983). Raters are instructed to consider the statements about the level of satisfaction they feel in different aspects of their relationship, and rate items five items on a seven-point scale: 1 (“very strong disagree”), 2 (“strongly disagree”), 3 (“disagree”), 4 (“neither agree nor disagree”), 5 (“agree”), 6 (“strongly agree”), and 7 (“very strongly agree”). In addition, raters also provide an overall rating of their level of happiness in the relationship on a 10 point scale: 1 (“unhappy”) to 10 (“perfectly happy”). The sum of items provides a total RQI Score; higher scores indicate better subjective relationship quality, and scores ≤ 29 indicate relationship distress (Norton, 1983).

Measures of Treatment Acceptability

Post-treatment Interview

An interview was conducted immediately following treatment to explore parents' or adolescents' experience with the assessment and treatment process. The post-treatment interview was conducted with a member of the study team who had no prior involvement with the family and followed a standard format (see Appendix N).

Treatment Acceptability Rating Form-Revised (TARF-R; Reimers et al., 1992)

The TARF-R is a 20-item instrument, designed to measure ratings of treatment acceptability (Reimers et al., 1992). It is administered post-treatment to obtain parent-report responses on eight subscales: Reasonableness, Effectiveness, Side Effects, Disruptive/Time Consuming, Cost, Willingness, Problem Severity, and Understanding. Parents are instructed to rate the items on a seven-point scale. The sum of six subscales (all excluding Problem Severity and Understanding) provides the Total Acceptability Score. Higher Total Acceptability Scores indicate higher levels of treatment acceptability. The TARF-R has demonstrated good to excellent psychometric properties, for example, the internal consistency ranged from 0.89 to 0.96 in a sample of parents who were seeking support for their child's behavioural difficulties (Reimers et al., 1992).

Measures of Reliability

Inter-observer Agreement (IOA)

Parent- or self-report sleep diaries and either video recording or actigraph data were compared to calculate IOA, for 25% of nights across study phases. It should be noted that the researcher calculated IOA for all participants and coded Hannah and Liam's video recording herself. Frequency of NWs were recorded as in agreement if both the parent- and researcher-report sleep diaries noted the presence of the awakening, and disagreement if the awakening was not reported on both diaries. Duration of NWs and SOL as well as bedtime and wake

time were recorded as in agreement if the parent- and researcher-report sleep diaries recorded a similar time (± 15 minutes). For Jack, Luna, and Mason, actigraphs provided information pertaining to bedtime, wake time, SOL, and TST. This information was used to calculate IOA. For all participants, IOA was calculated for each variable (frequency of NWs, duration of NWs, SOL, TST, bedtime, wake time) using the following equation: [agreements/(agreements + disagreements)] x 100.

Treatment Fidelity

Treatment fidelity was calculated for 25% of nights across intervention and follow-up phases. This provided a measure of adherence to the treatment plan by parents and by Jack himself. This was completed by examining parent- or self-report sleep diaries, contact notes, and video recording if applicable, and comparing this information with a checklist that outlined the steps in the treatment plan, for each child. All participants started intervention with modification to sleep/wake schedules; the researcher marked each bedtime and wake time as adhered to if reported in the sleep diary at a similar time (± 15 minutes). For Jack, P2 (sleep hygiene modifications) was marked as adhered to or not adhered to based upon the activities that he reported engaging in before bed (e.g., it was adhered to if he reported not using electronic devices). This was the most objective measure for this treatment component. For Hannah, P2 (faded parental presence) and P3 (fading nighttime milk) was marked as adhered to for each step Hannah's parents followed for a particular night i.e., there were multiple steps to follow, and these were marked for adherence individually. Other treatment components for Jack, Hannah, and Liam were subjective or there was insufficient data to precisely measure treatment fidelity (i.e., relaxation exercises, positive coping self-statements). The overall treatment fidelity score was calculated using the formula: (number of treatment components implemented/total number of treatment components) x 100.

Study Phases

There were five distinct study phases; assessment, baseline, intervention, maintenance, and follow up.

Assessment

Assessment involved completing the clinical interview, SATT, FBA, and all pre-treatment measures for the child and parents (for e.g., the Vineland-3, the CSHQ, the CBCL, the PSQI).

Baseline

Following these initial assessment procedures, participants were randomly assigned a baseline length of one, two, or three weeks. During baseline, parent- and/or child self-report sleep diaries, video recording, and/or actigraphs were used to collect data. Parents were asked to maintain the child's typical sleep routine and their own responses to the child's sleep problems during this phase. Intervention occurred immediately after baseline, provided a stable baseline was evident. This ensured that the researchers were able to attribute change to treatment components rather than other extraneous variables (Blampied, 2013).

Intervention

Intervention was started directly following the conclusion of the baseline period. Interventions were informed by assessment data including FBA and were individualised according to the participant's needs and goals. Prior to commencing intervention, the clinician discussed the steps in the treatment plan with the family. Treatment components were progressively introduced in sub-phases, starting with less restrictive procedures. Treatment sub-phase 1 (P1) consisted of modification to sleep/wake schedules, for example, sleep/wake scheduling (i.e., setting a consistent bedtime and wake time) and faded bedtime. Treatment sub-phase 2 (P2), other antecedent-based modifications, were introduced if sleep problems persisted following sub-phase 1. Antecedent-based modifications included sleep

hygiene modifications, stimulus control, visual supports, and relaxation exercises. Consequence-based modifications were implemented following P1 or P2 if needed.

Throughout the intervention, there was daily contact between the clinician and family via phone (call or text), email, and/or video call (Zoom, Skype). This contact was necessary for the clinician to support the family to adhere to the plan, resolve any problems, amend the plan if needed, and discuss the next steps. Further, it allowed the clinician to track the participant's progress and discuss this with the study team. Intervention proceeded until the sleep problem was reduced or eliminated, treatment goals were met, and parents were satisfied with the progress made to date.

Maintenance

Following the intervention phase, parents immediately began a maintenance phase of four to six weeks. During this period, post-treatment assessments were administered and the post-treatment interview was conducted. Aside from this, there was no contact between the study team and the family during this phase. This was to allow for the strategies to be incorporated into the family's routine without support from the study team.

Follow-up

Parent- and/or self-report sleep diaries were completed and video recording or actigraph data was collected for seven days. Typically, short-term follow-up (STFU) was completed four to six weeks post-treatment, and long-term follow-up (LTFU) was completed twelve to fourteen weeks post-treatment. In several cases, however, STFU or LTFU was completed earlier or later to suit the family's availability (see *Individual Cases* below).

Description of Data Analysis

Visual analysis was used to evaluate the effectiveness of the intervention. Data collected from participants during baseline, intervention, and follow-up phases is evidenced on multiple types of graphs. The first type of graph depicts all participants' sleep

wake/schedules and the longest periods of consolidated sleep across the intervention. Thus, depicting shifts in the participants' circadian rhythm over time. The second type of graph presents the studied dependent variables (SOL, frequency of NWs, duration of NWs, TST), arranged by cases. For Ethan, only the first type of graph is presented as an irregular sleep/wake schedule was the dependent variable. The primary method of data analysis for both graph types was visual analysis. Visual analysis is commonplace in single-case research, as it provides a visual of the data, allows researchers to identify and analyse changes in behaviour in real-time, and assess whether intervention caused these changes (Blampied, 2013; Hanley et al., 2003). Visual analysis can be a powerful method for analysing data with appropriate use (Blampied, 2013). In this research, it involved visual inspection of the data presented on graphs, to examine changes in behaviour between the study phases within each case (Hanley et al., 2003). Visual inspection was completed for six variables: the mean, trend, level, latency, variability, and consistency (Cohen et al., 2014).

Further, the percentage of data points exceeding the median (PEM; Ma, 2006) in baseline was used to analyse the effectiveness of intervention. The PEM approach assumes that if intervention has no effect on behaviour, there is a 50% chance the data points will be above the baseline median and a 50% chance the data points will be below the baseline median (Ma, 2006). The PEM score ranges from 0-1 and can be thought of as the effect size (Ma, 2006). In this study, intervention was considered effective if there was a reduction in SOL, frequency of NWs and duration of NWs, and an increase in TST. Thus, to calculate the PEM: the percentage of data points below the baseline median was used for SOL and NWs, and the percentage above was used for TST. PEM scores can be interpreted qualitatively: 0.9 or more is "highly effective", 0.7-0.89 is "moderate", 0.50-0.69 is "mild", and 0.50 or less is "ineffective" (Ma, 2006, p. 600).

In addition, it was noted for each dependent variable (i.e., SOL, frequency and duration of NWs, TST) if there was clinically substantive change between pre- and post-treatment. The following clinical cut-offs for poor sleep quality are based upon guidelines by the National Sleep Foundation (Ohayon et al., 2017): >45 mins SOL, ≥ 4 NW frequency, and ≥ 5 mins NW duration. Further, TST was measured for Jack; 7 to 9 hours of sleep is recommended for Jack's age by the National Sleep Foundation (Hirshkowitz et al., 2015) thus, the clinical cut-off of <7 hours (i.e., 420 mins) was selected.

Pre- and post-treatment psychometric outcome measures of sleep, child wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parental mental health, sleep, and relationship quality are presented in tables and analysed within and between participants, to measure changes attributed to intervention.

Individual Cases

The following section provides a description of each participant's characteristics, presenting problems, goals for treatment, results from the FBA, and individualised intervention. Table 2 provides an overview summarising the participants' sleep problems, factors precipitating and/or maintaining sleep problems, hypothesised function(s), and sub-phase treatment components.

Jack

Jack was a 19 year, 4-month-old adolescent male. Jack's age-equivalent score on the receptive and expressive domains of the Vineland-3 were 3:4 and 6:9, respectively. Jack lived at home with his parents and completed the intervention with parental support. Jack was first prescribed melatonin (4mg slow releasing) at around 3 years old and has taken it inconsistently in the past. Jack took melatonin for the duration of the study.

Presenting Problems. Jack's parents initially contacted the researchers and expressed interest in the study. Subsequently, Jack worked directly with a member of the research team

to complete an initial interview. Jack reported that he had difficulty falling asleep and engaged in sleep-competing activities before bed in response to feeling anxious and worrying (i.e., sleep-interfering thoughts). Information about the nature and history of Jack's sleep problems was obtained from Jack and his parents. The assessment information indicated that Jack went to bed between 12.00am and 4.00am, took 5 to 15 minutes to fall asleep, woke up around 7.00am on weekdays and 10.50am to 12.50pm on the weekend. **Goals.**
Jack's goals were to: (1) improve the consistency of his sleep schedule (i.e., fall asleep and wake up at a conventional time every night) in order to improve his mood and feel more refreshed, and (2) have a better bedtime routine (i.e., eliminate habits negatively impacting sleep).

FBA. Jack's sleep problems included SOD, short TST, and an irregular sleep/wake schedule. There were several factors precipitating and maintaining these sleep problems, as seen in Table 2. Jack had an inconsistent sleep/wake schedule on weekdays compared to the weekend which disrupted his circadian rhythm and decreased sleep pressure. When Jack experienced a delayed bedtime, this increased the reinforcement value of sleep and in turn, Jack's motivation to sleep (i.e., sleep pressure). When Jack tried to go to bed earlier however, he had insufficient motivation to sleep i.e., inadequate sleep pressure. Jack would sleep in until midday on the weekend and then go to bed late as his sleep pressure was insufficient for him to fall asleep at a conventional bedtime. In addition to insufficient MO for sleep, Jack experienced sleep-interfering thoughts and would avoid settling to sleep until late and increase his wakefulness by engaging in activities or using an electronic device. It was hypothesised that the function of Jack's sleep problems was access to tangible items or preferred activities.

Baseline. Jack was randomly assigned to a baseline length of two weeks.

Intervention. Jack's individualised intervention was comprised of three distinct sub-phases: (P1) modification to sleep/wake times, (P2) sleep hygiene modifications, and (P3) relaxation exercises. Table 3 provides the days that Jack's sleep/wake schedule was modified, as well as the days that sleep hygiene modifications and relaxation exercises were introduced.

Phase One (P1). In order to reset Jack's circadian rhythm and increase sleep pressure, a set bedtime and wake time was implemented. Initially, Jack was asked to go to bed at 1.00am and wake up at 7.00am every day. From the assessment information, 1.00am was the time Jack appeared to be falling asleep on average and he woke up at 7.00am on weekdays. Jack's bedtime was faded earlier to 12.30am once sleep onset was stable and then to 12.00am, 11.45pm, 11.30pm, 11.15pm, and 11.00pm. To consolidate Jack's sleep, his bedtime was delayed from 11.00pm to 11.15pm at the end of P1.

Phase Two (P2). To facilitate the onset of good quality sleep, Jack was asked to avoid engaging in activities or using electronic devices before going to sleep. Jack also found it difficult to sleep when the weather was hot or muggy. Jack was asked to check the weather prior to completing his bedtime routine and open his window to modify the bedroom temperature if necessary.

Phase Three (3). In order to alleviate anxiety and sleep-interfering thoughts, Jack met with the researcher to learn relaxation exercises (e.g., diaphragmatic breathing, progressive muscle relaxation). Jack chose to use the breathing exercise in bed before going to sleep.

Follow-up. STFU and LTFU were completed five- and 13-weeks post-treatment, respectively.

Hannah

Hannah was a 5 year, 8-month-old girl. Hannah received age-equivalent scores of 2:8 and 4:0 on the receptive and expressive domains of the Vineland-3, respectively. Hannah was prescribed melatonin (2mg) and took it an hour before bed throughout the study.

Presenting Problems. Hannah's parents contacted the researchers and expressed concern about sleep dependencies (parental presence, milk) and night wakings. Hannah had also spoken about nightmares waking her up, and often shared these with her parents during awakenings. Information about the nature and history of Hannah's problems was obtained from Hannah's parents.

Goals. Hannah's parents' goals were for Hannah to: (1) to sleep independently all night in her own bed, and (2) remain asleep until after 5am.

FBA. The assessment information showed that Hannah had an inconsistent sleep/wake schedule. As a result, Hannah had a disrupted circadian rhythm and inadequate sleep pressure. Results also showed that Hannah's dependence on nighttime milk and parental presence during sleep onset and night wakings was interfering with her ability to settle independently. When Hannah woke up and called out, she received positive social reinforcement through attention from her mother, coupled with tangible reinforcement (i.e., access to milk). It was hypothesised that the functions of Hannah's sleep problems were access to social attention and tangible items (e.g., milk) (see Table 2).

Baseline. Hannah was randomly assigned a baseline length of two weeks.

Intervention. Intervention was comprised of four distinct sub-phases: (P1) modification to sleep/wake times, (P2) faded parental presence, (P3) fading nighttime milk and rewards, and (P4) relaxation, positive coping self-statements and introducing a night light.

Phase One (P1). In order to increase sleep pressure, the researchers asked Hannah's parents to implement a set bedtime and wake time. Hannah's parents were asked to put Hannah to bed at 7.30pm and wake her up at 6.00am every day. From the assessment information, 6.00am was Hannah's average morning wake time. On average, Hannah fell asleep at 7.30pm, thus her bedtime was set at this time to increase sleep pressure. A Gro-

Clock™ was also introduced to provide a discriminative stimulus for sleep/wake times. A Gro-Clock™ is a digital clock on which the face lights up blue showing a large star for bedtime, and switches to yellow with a large sun for wake time. Hannah's parents were able to adjust the settings of the Gro-Clock™ to match her sleep schedule. Table 3 provides the days that Hannah's sleep/wake schedule was modified.

Phase Two (P2). In order to begin fading external cues for social attention or tangible items (e.g., milk), Hannah's mother was instructed to start a faded parental presence procedure. For the first seven nights, Hannah's mother would put Hannah to bed and then prompt her to grab the bottle of milk from her bedside table by pointing at it. Hannah's mother was asked to engage nonverbally (e.g., no verbal interactions), sit beside her bed, leave the room after Hannah fell asleep, and place a bottle of milk on her bedside table for her to drink if she woke up. When Hannah woke up, Hannah's mother was instructed to go into the room and nonverbally prompt her to grab the bottle. If Hannah did not respond to the nonverbal prompt, Hannah's mother could use a hand over hand prompt. Following this step, Hannah's mother started sitting 1 to 2 metres further away from Hannah's bed when Hannah was falling asleep. Then, Hannah's mother sat in the doorway, outside the doorway with her legs visible, and outside the doorway with her legs not visible to Hannah. Table 3 provides the days that the steps in this procedure were introduced.

A social story was also introduced to prepare Hannah and help her to understand the procedure. Social stories were written by the researchers according to guidelines by Gray (2010). Hannah's parents provided photos of Hannah and her mother modelling the appropriate behaviour. Social stories were updated throughout the intervention to communicate information pertaining to the current and next steps of the treatment plan.

Hannah's parents reported that Hannah was tired, and it was difficult to keep her awake until 7.30pm. Following completion of Phase 2, Phase 1 was revisited, and Hannah's bedtime was faded earlier to 7.15pm on night 77 and then to 7.00pm on night 85.

Phase Three (P3). Hannah's mother was asked to start fading the amount of nighttime milk given to Hannah and implement a reward system. The researchers provided Hannah's mother with a milk schedule that reduced the milk by 10% every third night, until it was eliminated (see Table 3). Hannah's mother was asked to give Hannah the specified amount of milk before bed and during night wakings, and then leave the room. Hannah was given verbal praise and a sticker on her chart when she slept through the night independently. Hannah was able to choose a tangible reward when she had five stickers.

Phase Four (P4). Antecedent-based modifications were revisited due to the occurrence of nightmares during P2 and P3. To decrease sleep-interfering thoughts and possible nightmares, Hannah was taught a relaxation exercise (diaphragmatic breathing) and positive coping self-statements (e.g., "I am a big girl", "I am brave"). Hannah was instructed to use these tools when she woke up in the night and go back to sleep independently. These strategies were used proactively to target Hannah calling out or leaving her bedroom during the night. A night light was also introduced to help with Hannah's fear of the dark following nightmares.

Follow-up. STFU was completed four weeks post-treatment, and LTFU was completed 12 weeks post-treatment.

Luna

Luna was a 12 year, 7-month-old girl. Luna's age-equivalent receptive and expressive domain scores on the Vineland-3 were not collected. Luna had been prescribed melatonin but only took it occasionally.

Presenting Problems. Luna's parents expressed concern about Luna's difficulty falling asleep at night and early morning wakings. Information about the nature and history of Luna's sleep problems was obtained from her parents. From the assessment sleep diaries, Luna took an average of approximately 1 hour to fall asleep and the longest duration was 120 minutes. However, no early morning wakings were reported over the week by Luna's parents.

Goals. Luna's parents' goals were for Luna to: (1) fall asleep within 15 to 20 minutes, and (2) feel refreshed when waking in the morning.

FBA. Luna's primary sleep problem was sleep onset delay. There were several factors precipitating and maintaining this sleep problem, as seen in Table 2. Luna had an inconsistent sleep/wake schedule, and thereby a disrupted circadian rhythm and decreased sleep pressure to fall asleep. When Luna went to bed earlier on a weeknight, she had insufficient motivation to fall asleep (i.e., inadequate sleep pressure) following a delayed bedtime and wake time in the weekend. Luna also had difficulty falling asleep during warmer temperatures. It was hypothesised that the function of Luna's SOD was access to tangible items or activities.

Baseline. Luna was randomly assigned a baseline length of two weeks.

Intervention. Luna's individualised intervention involved modification to sleep/wake times.

Modification to Sleep/wake Times: In order to reset Luna's circadian rhythm and increase sleep pressure, Luna's parents were asked to implement a set bedtime and wake time. Luna's parents were instructed to put Luna to bed at 9.30pm and wake her up at 7.00am every day. From the assessment information, Luna typically fell asleep at around 9.30pm. Luna's wake time was set to 7.00am to allow her to achieve 9.5 hours of sleep, which was within the appropriate range for her age. Table 3 provides the days that Luna's sleep/wake schedule was modified.

Follow-up. STFU was completed 7 weeks post-treatment, and LTFU was completed 13 weeks post-treatment. Both STFU and LTFU were delayed to suit the family's availability.

Mason

Mason was a 13 year, 5-month-old adolescent male. Mason's age-equivalent scores on the receptive and expressive domains of the Vineland-3 were 11:0 and 11:6, respectively. Mason had been prescribed melatonin but did not regularly take it during the study.

Presenting Problems. Mason's parents reported that he would take 1-2 hours to fall asleep each night. Information about the nature and history of Mason's sleep problems was obtained from Mason and his parents.

Goals. Mason's parents' goals were for Mason to: (1) to fall asleep within 20 to 30 minutes, (2) feel refreshed when waking in the morning, and (3) fall asleep without the use of medication. Mason's goals were to: (1) fall asleep within 30 minutes, and (2) have good quality sleep and not wake up at night.

FBA. Mason's sleep problems included sleep onset delay and night wakings. There were several factors precipitating and maintaining these sleep problems, as seen in Table 2. Mason had an inconsistent sleep/wake schedule on weekdays compared to the weekend. This disrupted his circadian rhythm and thereby decreased sleep pressure. When Mason went to bed on weeknights following a delayed wake time on the weekend, this decreased the reinforcement value of sleep and in turn, Mason's motivation to sleep i.e., insufficient sleep pressure. In addition to insufficient sleep pressure, Mason experienced sleep interfering thoughts. It was hypothesised that the function of Mason's sleep problems was access to tangible items or preferred activities.

Baseline. Mason was randomly assigned to a baseline length of two weeks.**Intervention.** Mason's individualised intervention involved modification to sleep/wake times.

Modification to Sleep/wake Times: In order to reset Mason's circadian rhythm and increase sleep pressure, a set bedtime and wake time was implemented. At first, Mason was asked to go to bed at 10pm and wake up at 7.30am every day. From the assessment information, 10pm was the time Mason appeared to be falling asleep on average. The sleep diaries also indicated that Mason woke up at 7.30am on weekdays and around 10.30am on the weekend. Mason's wake time was set to 7.30am throughout the week to increase sleep pressure and allow him to achieve appropriate sleep for his age. Mason's bedtime was faded earlier to 9.45pm once sleep onset was stable. Table 3 provides the days that Mason's sleep/wake schedule was modified.

Follow-up. STFU was completed 6 weeks post-treatment, and LTFU data was not collected due to difficulty contacting the family.

Liam

Liam was an 8 year, 1-month-old boy. Liam had an age-equivalent score on the receptive and expressive domains of the Vineland-3 of 1:10 and 1:9, respectively. Liam took melatonin (6mg) for the duration of the study.

Presenting Problems. Liam's parents expressed concern about his sleep onset delay. Information about the nature and history of Liam's sleep problems was obtained from Liam's parents. Before starting the study, Liam's sleep onset latency had improved significantly, reducing to 30 minutes since following a new melatonin regime. When Liam was asleep, he would typically sleep through the night and wake up between 7.30am to 8.00am.

Goals. Liam's parents' goals were for Liam to: (1) fall asleep within 30 minutes, and (2) go to bed at 8.30pm and wake up at 7am.

FBA. Liam's primary sleep problem related to sleep onset delay. Liam had an inconsistent sleep/wake schedule resulting in a disrupted circadian rhythm and insufficient sleep pressure (see Table 2). Liam also used electronic devices before bedtime which increased his wakefulness. Results also showed that Liam lacked a consistent discriminative stimulus for sleep onset. Liam did not have a consistent bedtime routine to cue bedtime approaching and he was lying awake in bed for a long duration (engaging in vocal stereotypy). This may have led to an association of bed with wakefulness rather than with sleeping. It was hypothesised that the function of Liam's SOD was access to tangible items or preferred activities, though vocal stereotypy was automatic (i.e., non-functional).

Baseline. Liam was randomly assigned to a baseline length of one week.

Intervention. Liam's individualised intervention was comprised of three distinct sub-phases: (P1) modification to sleep/wake times, (P2) sleep hygiene modifications, and (P3) white noise. Table 3 provides the days that Liam's sleep/wake schedule was modified, as well as the days that sleep hygiene modifications and white noise were introduced.

Phase One (P1). In order to reset Liam's circadian rhythm and increase sleep pressure, Liam's parents were asked to implement a set bedtime and wake time. Initially, Liam's parents were instructed to put him to bed at 9.45pm and wake him up at 7.15am. From the assessment information, Liam appeared to be falling asleep around 9.45pm. Liam's wake time was set to 7.15am to increase sleep pressure and allow him to achieve appropriate sleep for his age. Liam's bedtime was faded earlier to 9.30pm and then to 9.20pm once sleep onset was stable. At the end of intervention, Liam's wake time was faded earlier from 7.15am to 7.00am to provide more time to get him ready in the morning. Table 3 provides the days Liam's sleep/wake schedule was modified.

Phase Two (P2). To facilitate sleep onset, a consistent bedtime routine was implemented, and screen time was reduced. Liam was allowed to access his electronic device

until 8.45pm, then he watched his favourite TV show, and then there was no screen time, and he could engage in stereotypy, calming sensory activities, and have cuddle time, before going to bed. After 1 month, an adjustment was made to the plan as Liam's stereotypy was stimulating him after he was calm. Parents reduced the no screen time before bed to 15 minutes, made it non-stimulating, and redirected Liam away from engaging in stereotypy (e.g., running back and forth) where possible.

Phase Three (P3). White noise had previously been discussed as a potential strategy to implement; Liam's parents spontaneously initiated the use of white noise in response to Liam having difficulty falling asleep. White noise was introduced consistently from night 34 onwards to avoid it being used reactively, provide Liam with a consistent discriminative stimulus for sleep onset, and target his vocal stereotypy.

Follow-up. STFU was completed 4 weeks post-treatment, and LTFU was completed 10 weeks post-treatment.

Ethan

Ethan was a 3 year, 9-month-old boy. Ethan's age-equivalent score on the receptive and expressive domains of the Vineland-3 were both 0:3. Ethan was prescribed 6mg melatonin and took it every night for the duration of the study.

Presenting Problems. Ethan's parents expressed concern about his sleep/wake schedule as Ethan was sleeping multiple times per day. Ethan's sleep onset and wake times significantly varied every night. In order to achieve sleep onset, his parents would drive him around in the car. Ethan's day typically consisted of varying combinations, durations, and timings of playing, napping, and then having a long sleep. Ethan's sleep/wake schedule was highly variable and did not follow a consistent pattern over a 24-hour period. For example, he slept from 9.00am to 3.00pm, 9.30pm to 12.00am, and 5.45am to 9.00am in one 24-hour period, and then 7.00pm to 10.30 and 4.30 to 7.30am in the next 24-hour period. Ethan also

co-slept with his parents though this was not a target for intervention. During the study, Ethan was typically given melatonin between 12 to 2am. After Ethan was given melatonin, he would go to sleep within 1-2.5 hours. Ethan's sleep duration was 5 to 9.5 hours at night in addition to a 45 minute to 3.5 hour nap.

Goals. Ethan's parents' goals were for Ethan to: (1) maintain a consistent sleep/wake schedule each day, and (2) achieve 10 hours of sleep at night and nap for 1 hour.

FBA. Ethan had a highly irregular sleep schedule (sleep onset and wake time) with two periods of sleep over 24-hours. There were several factors precipitating and maintaining this sleep problem, as seen in Table 2. Ethan had an inconsistent sleep/wake schedule and thereby, a disrupted circadian rhythm and decreased sleep pressure. In addition to insufficient MO for sleep, Ethan had an inconsistent discriminative stimulus for sleep, as he often fell asleep in the car. It was hypothesised that the functions of this sleep problem were access to tangible items or social attention.

Baseline. Ethan was randomly assigned to a baseline length of two weeks.

Intervention. Ethan's individualised intervention involved modification to sleep/wake times.

Modification to Sleep/wake Times: In order to reset Ethan's circadian rhythm and increase sleep pressure, Ethan's parents were asked to implement a set sleep/wake schedule (bedtime, wake time, nap time). Ethan's parents were instructed to put Ethan to bed at 9pm for the night, wake him up at 7.00am, and allow him to nap in the morning at 10am for a maximum of 1 hour. If Ethan did not fall asleep at 10am, his parents were asked to attempt this again, however, the researchers asked his parent's to not allow him to sleep at all between 1pm and 9pm. Ethan's set sleep/wake schedule would allow him to achieve 11 hours of asleep, within the appropriate range for his age. The family did not want support

addressing Ethan falling asleep in the car. Table 3 provides the days that Ethan's sleep/wake schedule was modified.

Follow-up. Follow-up was completed 18 weeks post-treatment due to difficulty getting in contact with the family.

Table 2

Sleep Problems, Factors Precipitating and/or Maintaining the Sleep Problems, Hypothesised Function(s), and Treatment Component(s)

	Sleep problems	Factors precipitating and/or maintaining the sleep problems	Hypothesised function(s)	Sub-phase treatment component(s)
Jack	SOD, TST, irregular sleep/wake schedule	(1) Insufficient sleep pressure, inconsistent sleep/wake schedule, (2) Device use, bedroom temperature, (3) Sleep-interfering thoughts	Access to tangible items or preferred activities	*Antecedent-based modifications:* (1) Modification to sleep/wake times, (2) Sleep hygiene modifications (e.g., eliminated electronic device use before bed, modified bedroom temperature), (3) Relaxation (diaphragmatic breathing exercises)
Hannah	Freq. and duration of NWs, sleep dependencies (milk, parental presence), irregular sleep/wake schedule	(1) Insufficient sleep pressure, inconsistent sleep/wake schedule, (2) Social attention (3) Access to bottle (4) Sleep-interfering thoughts	Access to social attention or tangible	*Antecedent-based modifications:* (1) Modification to sleep/wake times, Gro-Clock™ (4) Relaxation (diaphragmatic breathing exercises in response to nightmares), positive coping self-statements, night light *Consequence-based modifications:* (2) Systematic fading of parental presence, (3) Systematic fading of nighttime milk, rewards
Luna	SOD, irregular sleep/wake schedule	(1) Insufficient sleep pressure, inconsistent sleep/wake schedule, (2) Sleep environment (e.g., bedroom temperature)	Access to tangible items or preferred activities	*Antecedent-based modifications:* (1) Modification to sleep/wake times

Mason	SOD, freq. and duration of NWs, irregular sleep/wake schedule	(1) Insufficient sleep pressure, inconsistent sleep/wake schedule, (2) Sleep interfering thoughts	Access to tangible items or preferred activities	*Antecedent-based modifications:* (1) Modification to sleep/wake times
Liam	SOD, irregular sleep/wake schedule	(1) Inadequate sleep pressure, inconsistent sleep schedule, (2) Device use, (3) Lack of consistent discriminative stimulus for bedtime	Access to tangible items or preferred activities	*Antecedent-based modifications:* (1) Modification to sleep/wake times (2) Sleep hygiene modifications (e.g., consistent bedtime routine, reduced screen time) (3) White noise
Ethan	Irregular sleep/wake schedule	(1) Inadequate sleep pressure, inconsistent sleep schedule	Access to social attention or tangible	*Antecedent-based modifications:* (1) Modification to sleep/wake times

Note. Freq.= frequency, SOD= sleep onset delay, TST= total sleep time, NWs= night wakings.

Table 3

Days of Sub-phase Changes and Treatment Modifications

	Sleep/wake schedule	Parental presence faded	Nighttime bottle faded	Sleep hygiene modifications	Relaxation exercises	White noise
Jack	P1; 15 (1.00am/7.00am), 21 (12.30am/7.00am), 28 (12.00am/7.00am), 33 (11.45pm//7.00am). 49 (11.30pm/7.00am), 68 (11.15pm/7.00am), 84 (11.00pm/7.00am), 91 (11.15pm/7.00am)			P2; 102	P3; 125	
Hannah	P1; 15 (7.30pm/6.00am), 77 (7.15pm/6.00am), 85 (7.00pm/6.00am)	P2; 39 (nonverbal prompting), 46 (moved 1-2m), 55 (moved to doorway), 61 (outside door legs visible), 66 (outside door not visible)	P3; 95 (75mL), 98 (67mL), 102 (59mL), 107 (51mL), 110 (43mL), 113 (35mL), 119 (19mL), 123 (11mL), 127 (0mL)		P4; 146	
Luna	P1; 15 (9.30pm/7.00am), 57 (9.45pm/7.00am)					
Mason	P1; 15 (10.00pm/7.30am), 50 (9.45pm/7.30am)					

Liam	P1; 8 (9.45pm/7.15am), 33 (9.35pm/7.15am), 63 (9.20pm/7.15am), 150 (9.20pm/7.00am)	P2; 19	P3; 34
Ethan	P1; 15 (9.00pm/7.00am)		

Note. P=phase.

Chapter Four: Results (Part Two)

In this chapter, sleep outcome data derived from sleep diaries and psychometric measures are presented. Pre- and post-treatment measures of child wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and measures of parental mental health, sleep, and relationship quality are also presented. Finally, post-treatment measures of treatment acceptability and reliability are provided.

Quality of Sleep Data

For all participants, there was missing or incomplete data recorded in sleep diaries, psychometric measures, or technological devices (i.e., actigraphs), as outlined below.

Sleep Diaries

For Jack, there was missing data pertaining to TST for nights 23 and 74, and sleep/wake times for nights 13, 23, 28, 64, 74, 86, 88, 90, 114, 122, 124, 125, and 138. Hannah's frequency of NWs data for night 38, and duration of NWs data for nights 38, 40, 51, 123, 135, and 146 were missing. Luna's SOL data was missing for nights 62 and 105. Data for Mason's SOL, frequency and duration of NWs was missing for nights 1, 2, 52, 53, 54, 55, and 56. For Liam, there was missing data pertaining to SOL for nights 50 and 51, and sleep/wake times for nights 2, 50, 51, 73-77, and 84. Ethan's sleep/wake times were missing for nights 36-41. Mason completed STFU but did not complete LTFU, and Ethan completed LTFU only.

Psychometric Measures

There were inconsistencies between participants in the psychometric measures completed at baseline and post-treatment. For Jack and Hannah, the RQI was not completed by parents at baseline and post-treatment. For Mason, the GARS-3 was not completed post-treatment. For Ethan, only the GARS-3 and CBCL were completed by parents at baseline and these measures were not completed post-treatment.

Actigraphs

There were technological issues with two of the actigraphs. As a result, there was usable data for 10 nights for Jack and 3 nights for Mason. Therefore, IOA was calculated for only 7% and 3% of nights, respectively. Further, Ethan's parents did not consent to video recording or actigraph use. Thus, IOA was not able to be calculated for his data.

Primary Treatment Outcomes

The data presented in the graphs and described below was collected from the sleep diaries. Figures 1, 3, 5, 7, 9, and 11 were modelled from Piazza et al. (1998) and depict sleep/wake schedules across baseline, intervention, and follow-up phases. Time of day is plotted on the y-axis, which represents a 24-hour period. The day is plotted on the x-axis. Bars that are shaded in black represent "appropriate" sleep, defined as sleep occurring within the participant's scheduled bedtime and wake time during that phase (Piazza et al., 1998, p. 363). Bars that are shaded red, represent "inappropriate" sleep, defined as sleep occurring outside of the participant's scheduled bedtime and waketime during that phase (Piazza et al., 1998, p. 363). The white gaps within the bars reflect a period of waking. The solid blue line represents the participant's target bedtime and wake time during each study phase. The blue dotted line was used to differentiate between baseline, treatment, STFU, and LTFU. Figures 2, 4, 6, 8, and 10 depict SOL (mins), frequency and duration of NWs (mins), and TST (mins) for each participant for whom this was an intervention target. The black dotted line is used to indicate the baseline medians. The black solid lines were used to differentiate between baseline, treatment sub-phases, STFU, and LTFU.

Dependent Variables

Jack. For Jack, his sleep/wake schedule and TST were identified as being problematic in baseline and were targeted during intervention. The outcome of the intervention for Jack's

sleep/wake schedule is presented in Figure 1, and SOL and TST are presented together in Figure 2.

Sleep/wake Schedule. Jack's sleep/wake schedule was highly variable during baseline (see Figure 1); Jack fell asleep between 12.15am to 7.15am and woke between 6.40am to 12.50pm. There was an immediate increase in appropriate sleep and reduction in inappropriate sleep following the introduction of a consistent bedtime and wake time (P1). Jack adhered to the set bedtime and wake time though SOL varied and impacted his sleep onset time. There was also an increase in the longest period of consolidated sleep, as evidenced by longer black bars representing sleep particularly in STFU. The outliers of inappropriate sleep on nights 55, 56, 79, 128, and 136 were due to external circumstances (e.g., travelling) or Jack choosing to go to bed later (e.g., sleepovers). By the final week of intervention, Jack was falling asleep between 11.45pm to 12.00am and waking up between 6.40am to 7.00am. Improvements were maintained at STFU (i.e., Jack fell asleep between 11.52pm to 12.20am and woke between 7.00 to 7.25am), though there was a small increase in inappropriate sleep at LTFU in which Jack went to sleep between 12.20am to 1.10am and woke between 6.45am to 8.30am. Overall, there was an increase in the consistency of Jack's sleep/wake schedule (time awake/asleep) and duration of consolidated sleep following intervention.

SOL. Jack's SOL was variable in baseline, ranging from 10 to 30 mins (M = 14.29), and remained within this range during the first 28 nights of P1 (modification to sleep/wake schedules). Jack's SOL ranged from 5 to 80 mins across P1 (M = 22.76 min), with an increase in duration and variability evident towards the end of this phase (see Figure 2). Jack's SOL remained variable across P2 (sleep hygiene modifications) though there was a progressive decrease and stabilisation by the end of intervention (i.e., SOL in P3; relaxation exercises, ranged from 10 to 45 mins, M = 29.52 mins). It should be noted that Jack's SOL

was non-problematic in baseline and was monitored due to his problematic sleep/wake schedule, and then increased as a result of shifting his bedtime earlier during P1. There was a slight increase from the end of intervention in SOL at STFU, and it remained variable (range= 20 to 60 mins; *M* = 38.57). There was a slight reduction in SOL at LTFU compared to STFU (range from 25 to 30 mins; *M* = 30.71 mins) though there was one outliner in which SOL was 40 mins. The PEM value may not be helpful for this dependent variable; the baseline median was low (i.e., 10 mins) thus, there was only one value below the baseline median. Overall, intervention was effective in improving the duration, variability, and consistency of Jack's SOL; Jack's SOL did increase from baseline though his bedtime was significantly more age-appropriate and SOL was more consistent.

Figure 1

Jack's Sleep/wake Schedule (Time Awake/asleep) across Treatment Phases

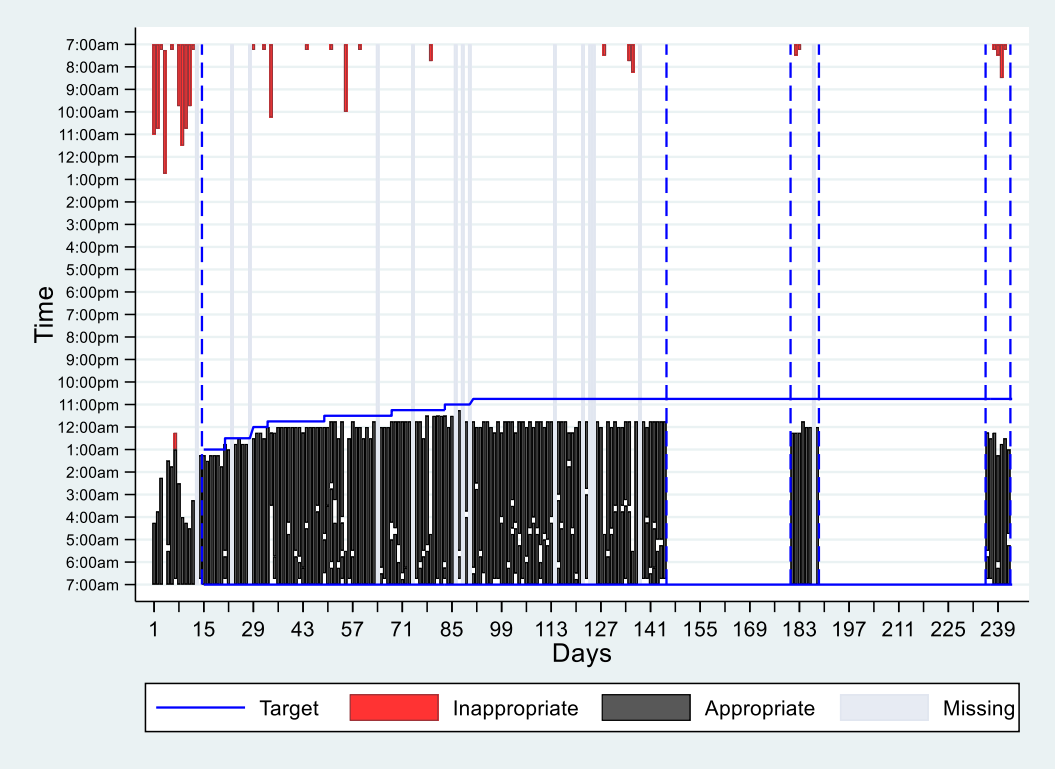
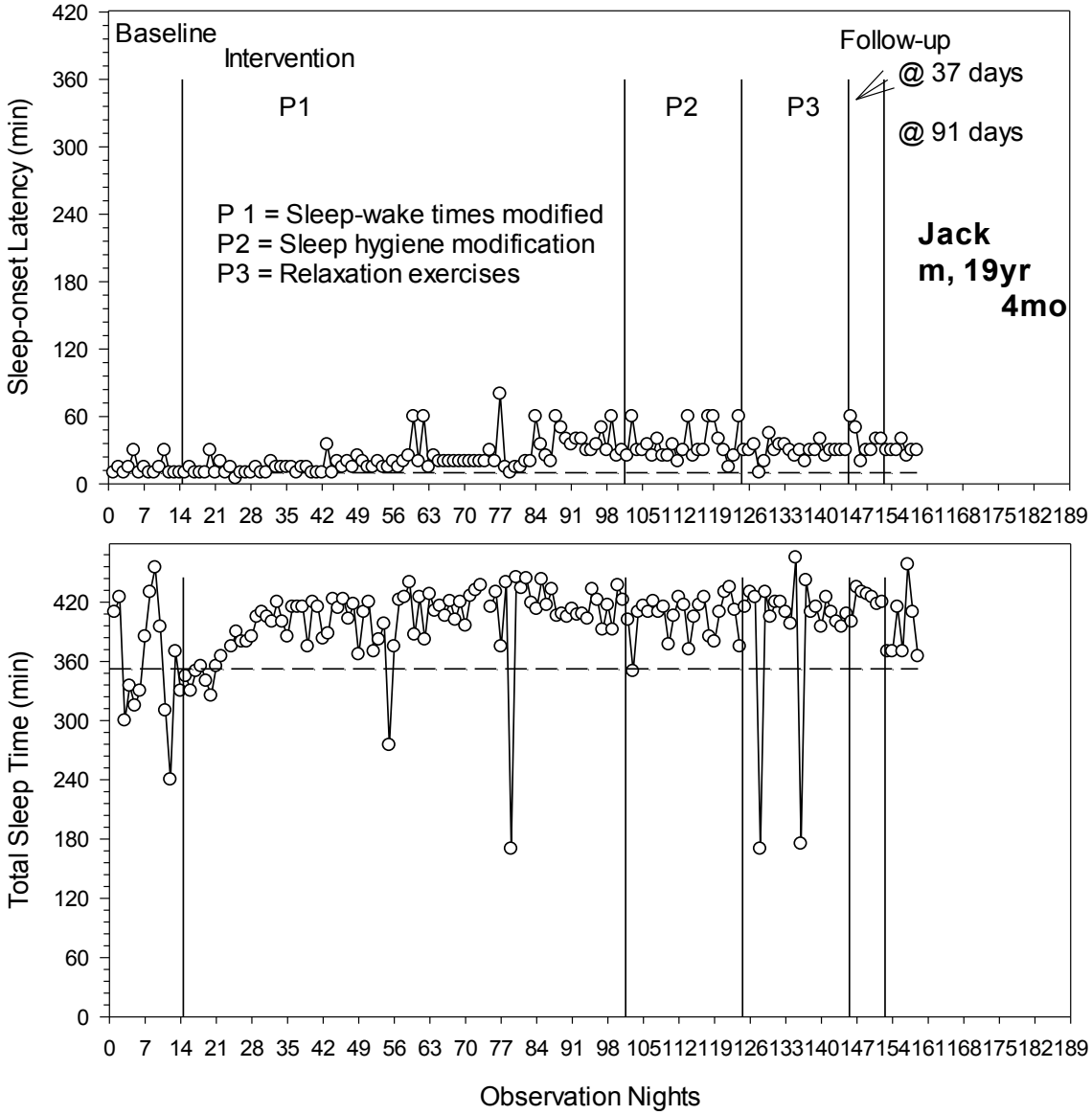


Figure 2

Duration of Sleep Onset Latency and Total Sleep Time per night (mins) across Treatment Phases for Jack



Note. M= male, mo= months, P= phase, yr= years.

TST. Jack's TST was problematic and below the clinical cut-off (i.e., 420 minutes; Hirshkowitz et al., 2015) at baseline. Following the initiation of treatment, a gradual increasing trend in the duration and stability of TST was observed (see Figure 2). Jack's TST ranged from 240-455 mins (M = 359.29) at baseline and 395-425 (M = 406.86) during the final week of intervention. Sleep improved mostly during the introduction of P1, where there

was an immediate increase in TST. The outliers evident on the graph (i.e., nights 55, 79, 128, 136) were associated with external circumstances that resulted in Jack going to bed late (e.g., travelling, sleepovers with friends or family). Further improvements were evident at STFU; TST was stable and ranged from 400-435 mins (M = 422.29). Jack's mean TST at STFU was above the clinical cut-off, indicating a clinically substantive change between baseline and post-treatment. There was a decrease in the duration and stability of TST at LTFU though improvements were maintained from baseline (M = 394.0; range= 365-458). The PEM value (i.e., percentage *above* the baseline median) was 0.92 indicating the intervention was highly effective. These treatment effects were maintained at STFU and LTFU for Jack (follow-up PBM= 1). Overall, intervention was effective in improving the duration and decreasing the variability of Jack's TST.

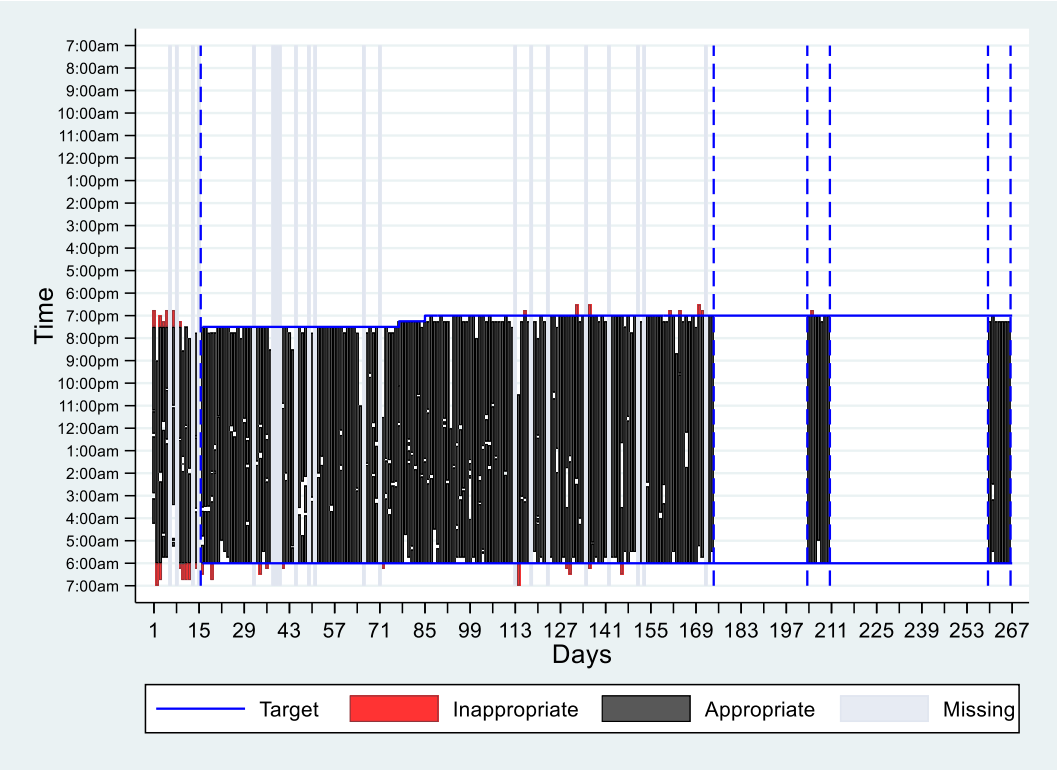
Hannah. For Hannah, sleep/wake times, frequency of NWs, and duration of NWs were problematic in baseline and measured across study phases. The outcome of the intervention for Hannah's sleep/wake schedule is presented in Figure 3, and frequency and duration of NWs is presented in Figure 4.

Sleep/wake Schedule. During baseline, Hannah's sleep/wake schedule was highly variable; Hannah fell asleep between 6.38pm and 9.05pm and woke up between 4.08am and 6.54am (see Figure 3). Following the introduction of P1 (i.e., modification to sleep/wake schedules), there was an almost immediate reduction in inappropriate sleep, and immediate increase in appropriate sleep. As Hannah's modified sleep/wake schedule was introduced (i.e., 7.30pm bedtime and 6.00am wake time), Hannah was sleeping and waking at appropriate and consistent times. There was also an increase in the longest period of consolidated sleep, as evident by longer black bars representing sleep particularly in P1, P4**,** and follow-up. It should be noted that frequency and duration of NWs were also problematic for Hannah and accounted for disruptions in consolidated sleep. During the final week of

intervention, Hannah fell asleep between 6.32pm to 7.24pm and woke up 5.09am to 6.00am, and the longest period of consolidated sleep increased due to an absence of night wakings. Improvements were maintained and the consistency of time asleep improved at STFU and LTFU; Hannah was falling asleep between 6.41pm to 7.10pm at STFU and between 7.06pm to 7.21pm at LTFU. Hannah also woke up between 5.00am to 6.00am at STFU and between 5.30am to 6.00am at LTFU. Overall, there was an increase in the consistency and consolidation of Hannah's sleep (time awake/asleep) following intervention.

Figure 3

Hannah's Sleep/wake Schedule (Time Awake/asleep) across Treatment Phases

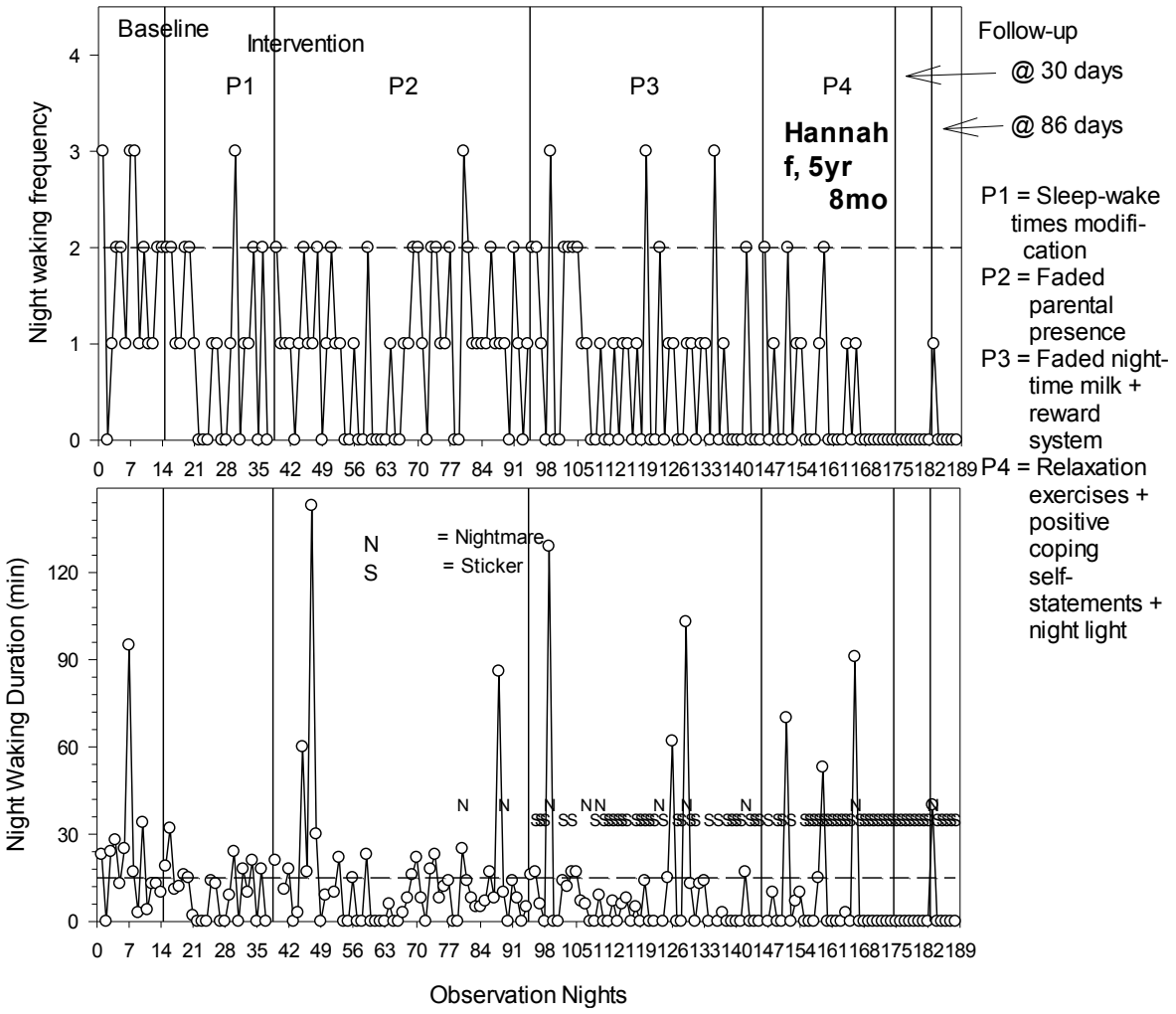


Frequency of NWs. Hannah's frequency of NWs was problematic though below the clinical cut-off (i.e., ≥ 4; Ohayon et al., 2017) at baseline. Following the commencement of treatment, Hannah showed a progressive reduction in frequency of NWs overtime (see Figure 4). For Hannah, frequency of NWs were variable and ranged from 0-3 (M = 1.71) at baseline. This frequency remained variable throughout intervention until P4 (i.e., relaxation exercises, positive coping self-statements, night light) at which point the frequency of NWs were stable

and reduced to zero for the final week of intervention. It appears that sleep improved mostly in P4 though this may be due to the combined and cumulative effect of all intervention components. Improvements were maintained in the follow-up phase during which time the frequency of NWs remained at zero aside from one NW at LTFU, however, this may have been because Hannah had a babysitter. The PEM value (i.e., percentage *below* the baseline median) during intervention was 0.79 indicating a moderate effect. This treatment effect was maintained and improved at STFU and LTFU for Hannah (follow-up PBM= 1). Overall, intervention was effective in reducing the frequency of NWs for Hannah.

Figure 4

Frequency and Duration of NWs per night across Treatment Phases for Hannah



Note. F= female, mo= months, N= nightmare, P= phase, S= sticker, yr= years.

Duration of NWs. Hannah exhibited an immediate reduction in the duration of NWs following treatment (see Figure 4). For Hannah, duration of NWs ranged from 0-95 mins (*M* = 21.57) at baseline. Sleep improved the most following the introduction of P1, where Hannah's sleep/wake schedule was modified. As noted above, Hannah was not waking by the end of intervention. As evident on the graph (Figure 4), nightmares were often present on nights with longer NWs and reduced between P3 and P4. During P4, the duration of NWs reduced and stabilised. Improvements were maintained at STFU as NWs had been eliminated, though there was one NW 40 mins in duration at LTFU as detailed above. The PEM value (i.e., percentage *below* the baseline median) was 0.76 indicating moderate effect of intervention on NW duration. These treatment effects were maintained and improved at STFU and LTFU for Hannah (follow-up PBM= 0.93). Overall, intervention was effective in reducing the duration and variability of NWs for Hannah.

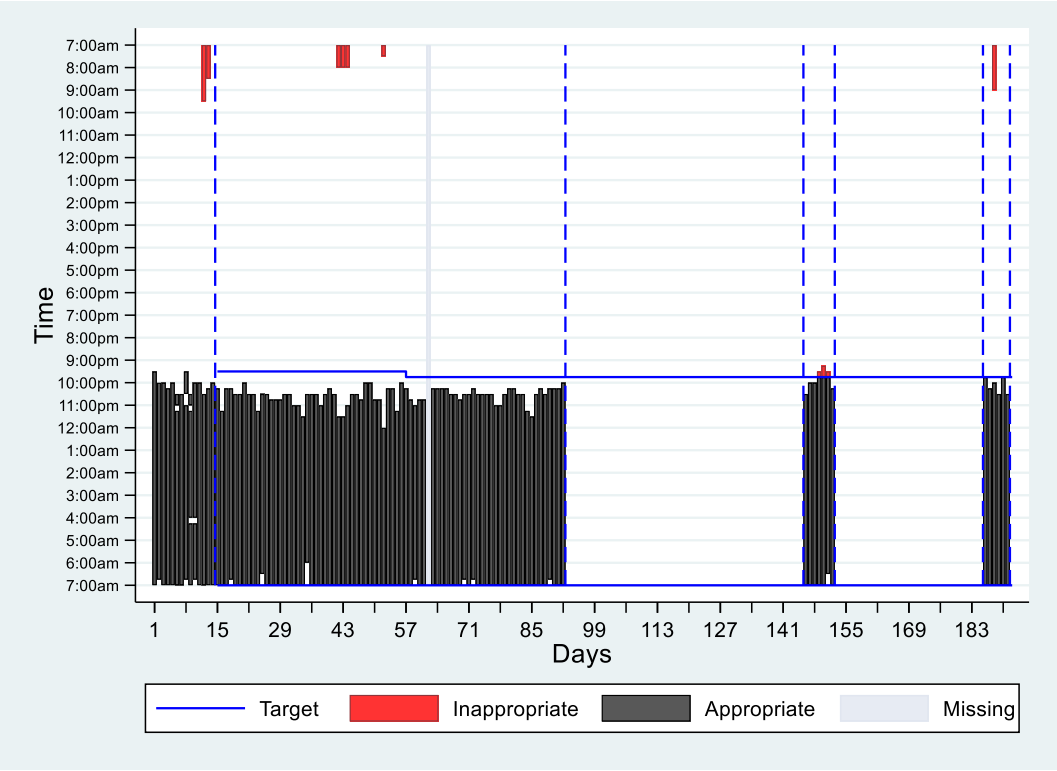
Luna. For Luna, sleep/wake times and SOL were problematic in baseline and measured across study phases. The outcome of the intervention for Luna's sleep/wake schedule is presented in Figure 5, and SOL is presented in Figure 6.

Sleep/wake Schedule. Luna's sleep/wake schedule was variable during baseline (see Figure 5); Luna fell asleep between 9.30pm and 10.30pm and woke between 6.50am and 9.30am. There was an immediate reduction in inappropriate sleep following the introduction of intervention. Luna adhered to the set bedtime and wake time, however, her SOL varied; this impacted her sleep onset time and the longest period of consolidated sleep. By the final week of intervention, there was an increase in the longest period of consolidated sleep as seen by longer black bars representing sleep; Luna was falling asleep between 9.55pm and 10.30pm and waking up between 6.45am and 7.00am. Improvements in appropriate and consolidated sleep were maintained at STFU and LTFU, though there was a small increase in inappropriate sleep at STFU and LTFU. Luna was falling asleep between 9.15pm to 10.30pm

at STFU and between 9.40pm to 10.30pm at LTFU, and woke up between 6.30am to 7.00am at STFU and between 7.00am to 9.00am at LTFU. Overall, there was an increase in the consistency of Luna's sleep/wake schedule (time awake/asleep).

Figure 5

Luna's Sleep/wake Schedule (Time Awake/asleep) across Treatment Phases

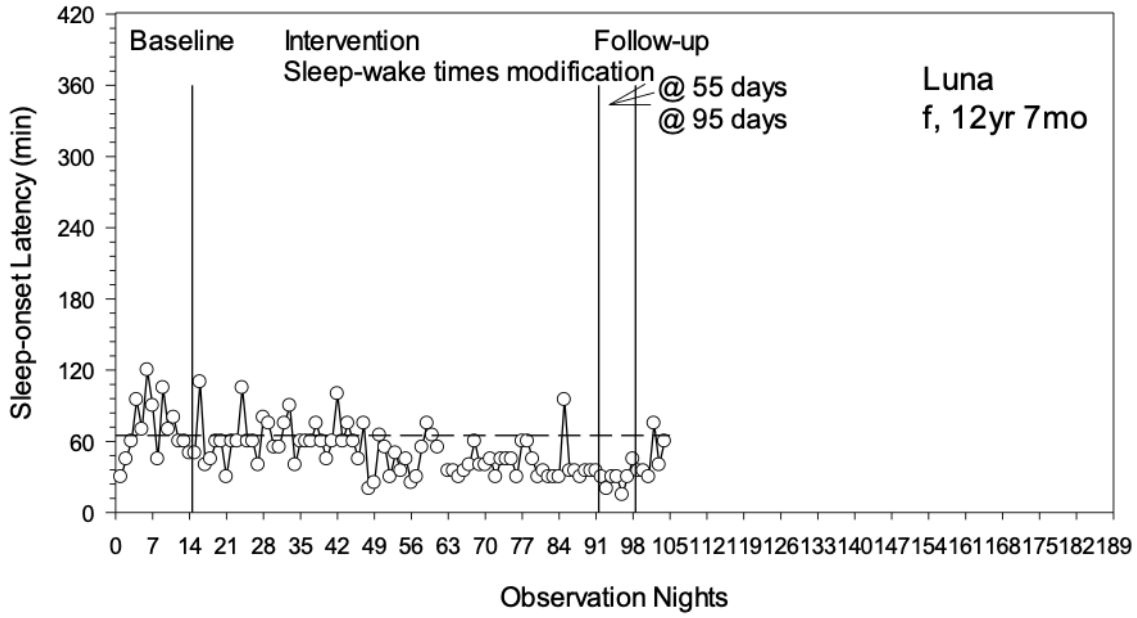


SOL. For Luna, SOL was highly variable and above the clinical cut-off (i.e., >45 mins; Ohayon et al., 2017) in baseline, ranging from 30 to 120 mins (M = 70.0). Following treatment initiation, Luna's SOL progressively reduced in duration. On night 57, Luna's bedtime was shifted 15 mins later, and SOL was observed to almost immediately reduce and stabilise over time i.e., sleep improved the most following this modification. Luna's SOL was relatively low and stable by the end of intervention, with further improvements evident at STFU (M = 28.57; range= 15-45). Luna's SOL (mean and range) at STFU was below the clinical cut-off, indicating a clinically substantive change between baseline and post-treatment. There was an increase in duration and variability of SOL at LTFU (range = 30 to

75 mins; *M* = 45.83 mins). The PEM value (i.e., percentage *below* the baseline median) was 0.83 indicating a moderate effect. This was maintained and enhanced at STFU and LTFU for Luna (follow-up PBM= 0.92). Overall, intervention was effective in improving the duration and variability of Luna's SOL.

Figure 6

Duration of SOL per night across Treatment Phases for Luna



Note. F= female, mo= months, yr= years.

Mason. For Mason, sleep/wake times, SOL, frequency of NWs, and duration of NWs were problematic in baseline. These dependent variables were measured across baseline, intervention, and STFU; however, LTFU data was not provided by the family. The outcome of the intervention for Mason's sleep/wake schedule is presented in Figure 7, and SOL, frequency and duration of NWs are presented together in Figure 8.

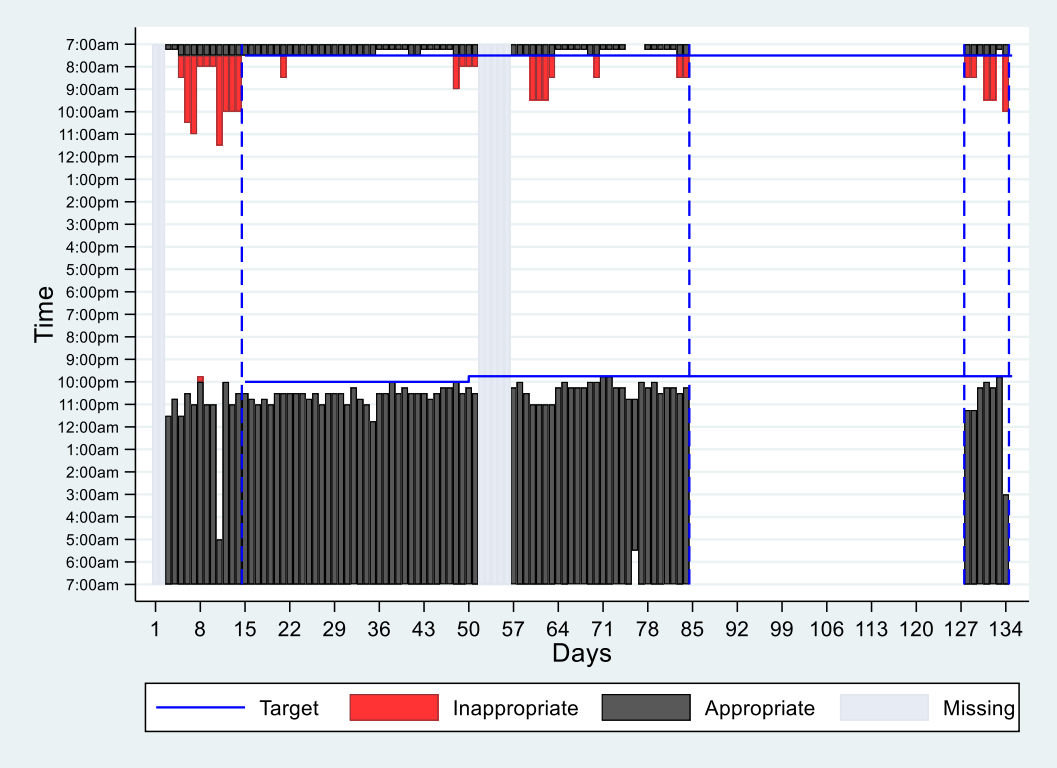
Sleep/wake Schedule. It should be noted that there was missing data pertaining to the timing of night wakings for Mason. As such, NW data has been omitted from Figure 7 and only the sleep/wake schedule data (i.e., sleep onset time and wake time) can be shown. Mason's sleep/wake schedule was highly variable during baseline (see Figure 7); Mason fell

asleep between 9.40pm and 5.00am and woke between 7.15am and 11.30am. There was an immediate reduction in inappropriate sleep and increase in appropriate sleep following the introduction of intervention. As seen by longer black bars representing sleep, there was also an increase in the longest period of consolidated sleep following treatment initiation. Mason's modified sleep/wake schedule was introduced and adhered by Mason, therefore, he was sleeping and waking at appropriate and consistent times. During the final week of intervention, Mason was falling asleep between 10.00pm to 10.30pm and waking up between 7.15am to 8.30am. Some improvements were maintained at STFU; there was an increase in inappropriate sleep and decrease in the longest period of consolidated sleep particularly on the last night due to a significant SOD (SOL= 300 mins). At STFU, Mason was falling asleep between 9.50pm to 3.00am and woke up between 7.15am to 10.00pm.

SOL. Mason's SOL was highly variable and above the clinical cut-off in baseline, ranging from 30 to 420 mins (M = 103.33). Following treatment initiation, there was an immediate reduction in SOL duration and variability; Mason's SOL ranged from 30 to 60 mins (M = 41.43) during the first week of intervention. SOL was observed to decrease and stabilise the most following modification to his sleep/wake schedule on night 50. Mason's SOL was low and stable relative to baseline, ranging from 30 to 45 mins (M = 35.71) during the last week of intervention. Further improvement was observed at STFU; SOL ranged from 10 to 40 mins (M = 30.0) excluding an outlier of 300 mins on night 91. Mason's SOL (mean and range) was below the clinical cut-off at STFU, indicating a clinically substantive change between baseline and post-treatment. The PEM value (i.e., percentage *below* the baseline median) was 1 indicating intervention was highly effective. The treatment effect reduced slightly at STFU for Mason (follow-up PBM= 0.86). Overall, intervention was effective in improving the duration and variability of Mason's SOL.

Figure 7

Mason's Sleep/wake Schedule (Time Awake/asleep) across Treatment Phases



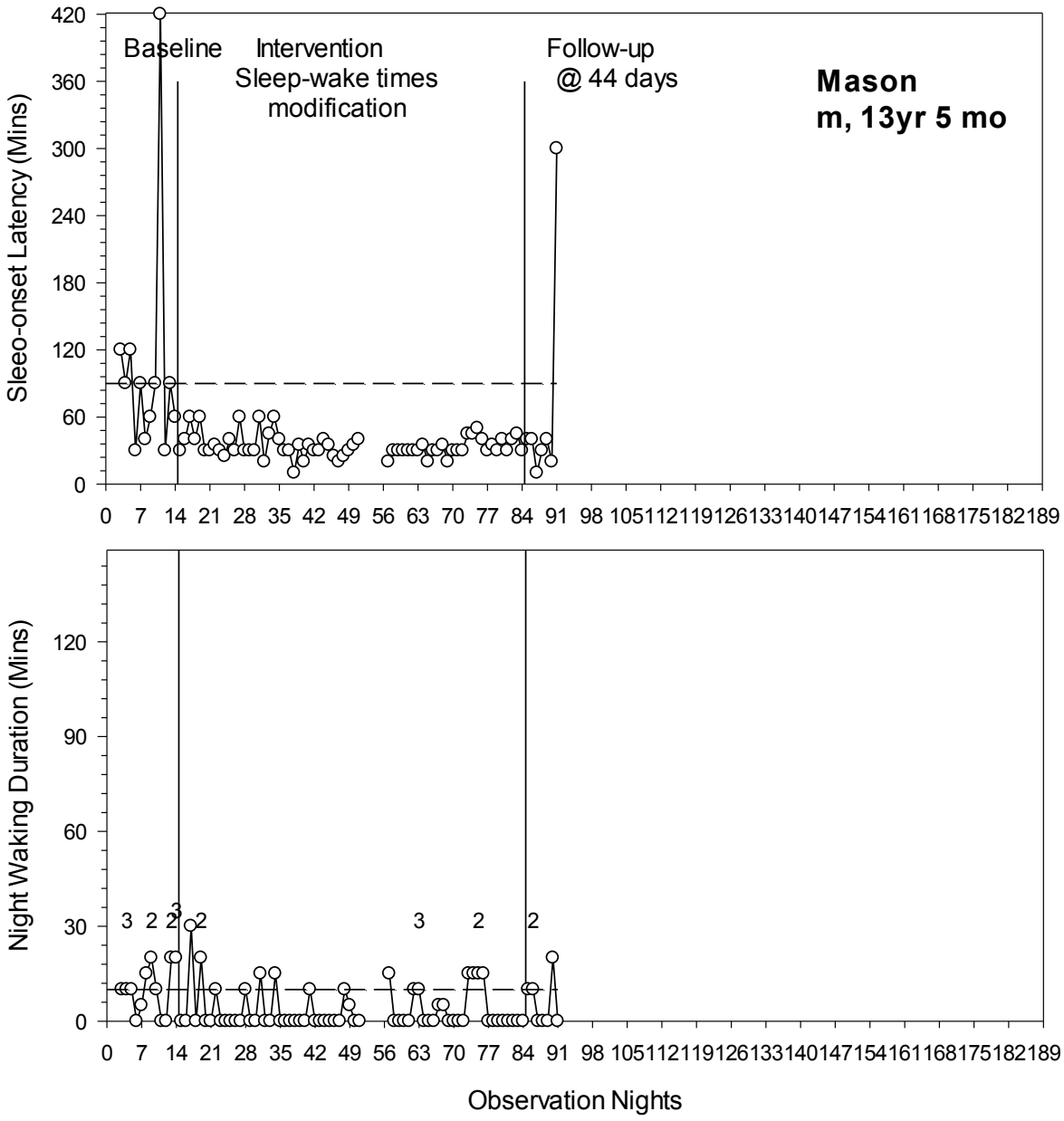
Duration of NWs. Mason exhibited an almost immediate reduction in NW duration and variability following treatment initiation (see Figure 8). For Mason, duration of NWs ranged from 0-20 (M = 10.0) at baseline. By the final week of intervention, Mason's duration of NWs reduced to zero. Mason exhibited an increase in NW duration at STFU (range 0-20, M = 5.71) however, this was a reduction in duration relative to baseline. The PEM value (i.e., percentage *below* the baseline median) was 0.77 indicating moderate treatment effect. These treatment effects reduced at STFU for Mason (follow-up PBM= 0.57). Overall, intervention was effective in reducing the duration of NWs for Mason.

Frequency of NWs. Following treatment initiation, Mason showed an immediate reduction in frequency NWs (see Figure 8). For Mason, frequency of NWs was variable and problematic though above the clinical cut-off ranging from 0-3 (M= 1.25) at baseline. This frequency remained low and stable throughout intervention relative to baseline. Mason exhibited an increase in the frequency of NWs at STFU (M = 0.57; range= 0-2), however,

this still reflected a reduction in frequency relative to baseline. The PEM value (i.e., percentage *below* the baseline median) was 0.72 indicating moderate effect. These treatment effects reduced at STFU (PBM= 0.57). Overall, intervention was effective in reducing frequency of NWs for Mason.

Figure 8

Frequency of NWs, Duration of NWs, and SOL per night across Treatment Phases for Mason



Note. M= male, mo= months, yr= years. Numbers 2 and 3 represent frequency of NWs.

Liam. For Liam, sleep/wake times and SOL were problematic in baseline and measured across study phases. The outcome of the intervention for Liam's sleep/wake schedule is presented in Figure 9, and SOL is presented in Figure 10.

Sleep/wake Schedule. During baseline, Liam's sleep/wake schedule was highly variable; Liam fell asleep between 9.30pm and 12.00am and woke up between 4.00am and 8.45am (see Figure 9). Following the introduction of a set bedtime and waketime (P1), there was an immediate decrease in inappropriate sleep and increase in appropriate sleep. Liam's modified sleep/wake schedule was introduced and adhered to thus, Liam was going to sleep and waking at appropriate and consistent times. There was also an increase in the longest period of consolidated sleep following treatment initiation, as seen by longer black bars representing sleep. This improved further following two modifications to Liam's sleep/wake schedule (i.e., bedtime faded earlier to 9.35pm on night 33 and 9.20pm on night 63). During the final week of intervention, Liam was falling asleep between 9.20pm to 10.05pm and waking up between 6.50am to 7.20am. Improvements were maintained at STFU, and further improvement was observed at LTFU i.e., Liam was falling asleep between 9.25pm and 10.40pm at STFU and 9.20pm and 9.45pm at LTFU, and waking up between 7.00am to 7.05 am at STFU and between 7.00am to 7.15am at LTFU. Overall, there was an increase in the consistency and consolidation of Liam's sleep (time awake/asleep) following intervention.

SOL. For Liam, SOL was highly variable and above the clinical cut-off in baseline, ranging from 60 to 195 mins (M = 116.43). There was an immediate reduction in Liam's SOL following treatment initiation; Liam's SOL ranged from 10 to 125 mins (M= 46.43) during the first week of intervention. Sleep improved mostly following the introduction of P1; on night 13 the SOL of 125 mins occurred due to incorrect melatonin dosage. SOL was observed to progressively decrease and then increase in P3, peaking on night 47 (145 mins) This coincided with a temporary reduction in melatonin for five nights (nights 47 to 51).

Following melatonin being reinstated, Liam's SOL continued to progressively decrease until the end of intervention. The outlier evident on the graph at night 78 was due to a storm. Liam's SOL was low and stable relative to baseline, ranging from 0 to 45 mins (*M* = 22.86) during the last week of intervention. Improvements were maintained at STFU (*M* = 25.71; range 5-80) and further improvement was observed at LTFU (*M* = 7.14; range= 0-25). Liam's mean TST was below the clinical cut-off at STFU and LTFU, indicating a clinically substantive change between baseline and post-treatment. The PEM value (i.e., percentage *below* the baseline median) was 0.97 indicating intervention was highly effective. These treatment effects were maintained and enhanced at STFU and LTFU for Liam (PBM= 1). Overall, intervention was effective in improving the duration and consistency of Liam's SOL.

Figure 9

Liam's Sleep/wake Schedule (Time Awake/asleep) across Treatment Phases

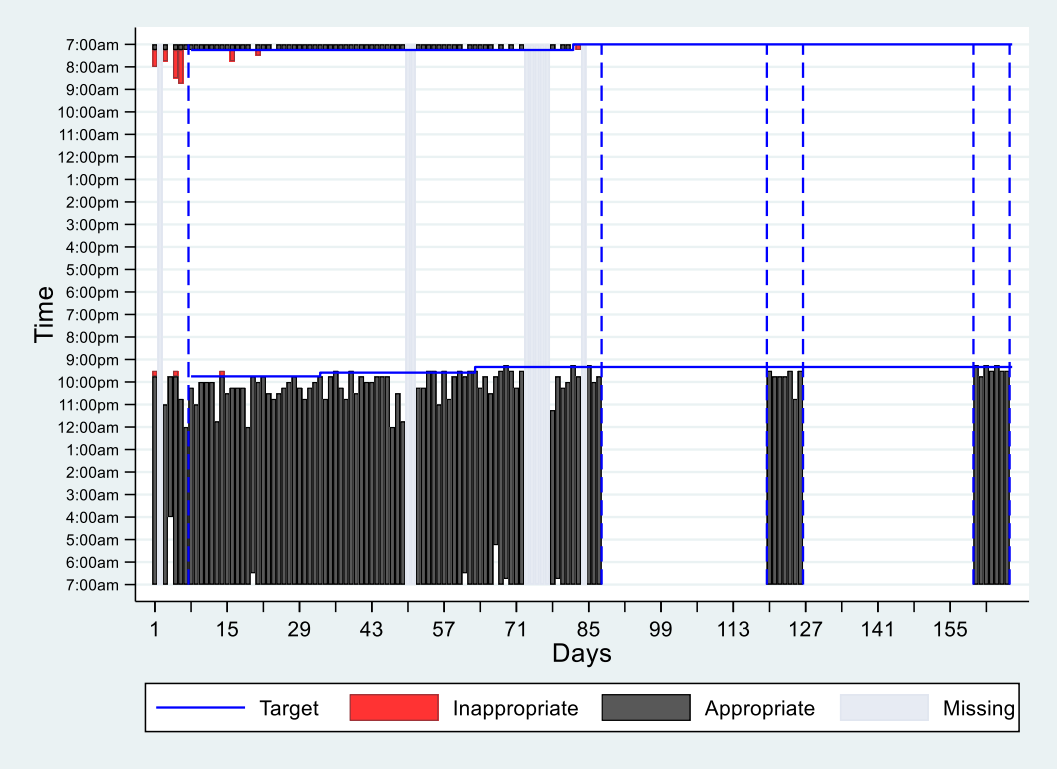
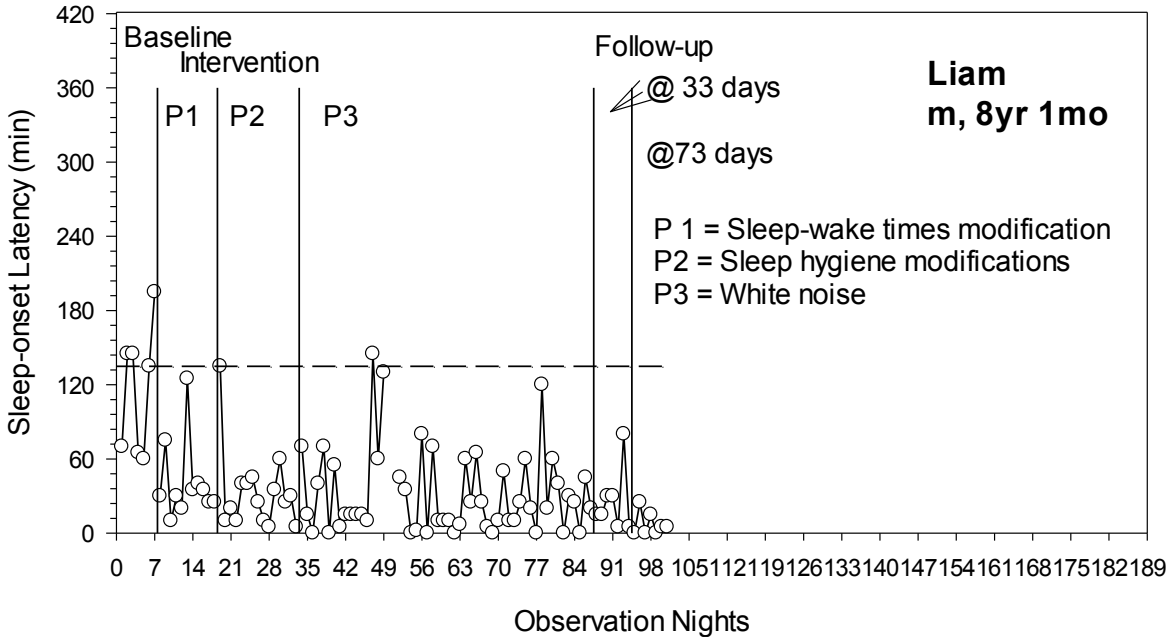


Figure 10

Duration of SOL per night across Treatment Phases for Liam



Note. M= male, mo= months, P= phase, yr= years.

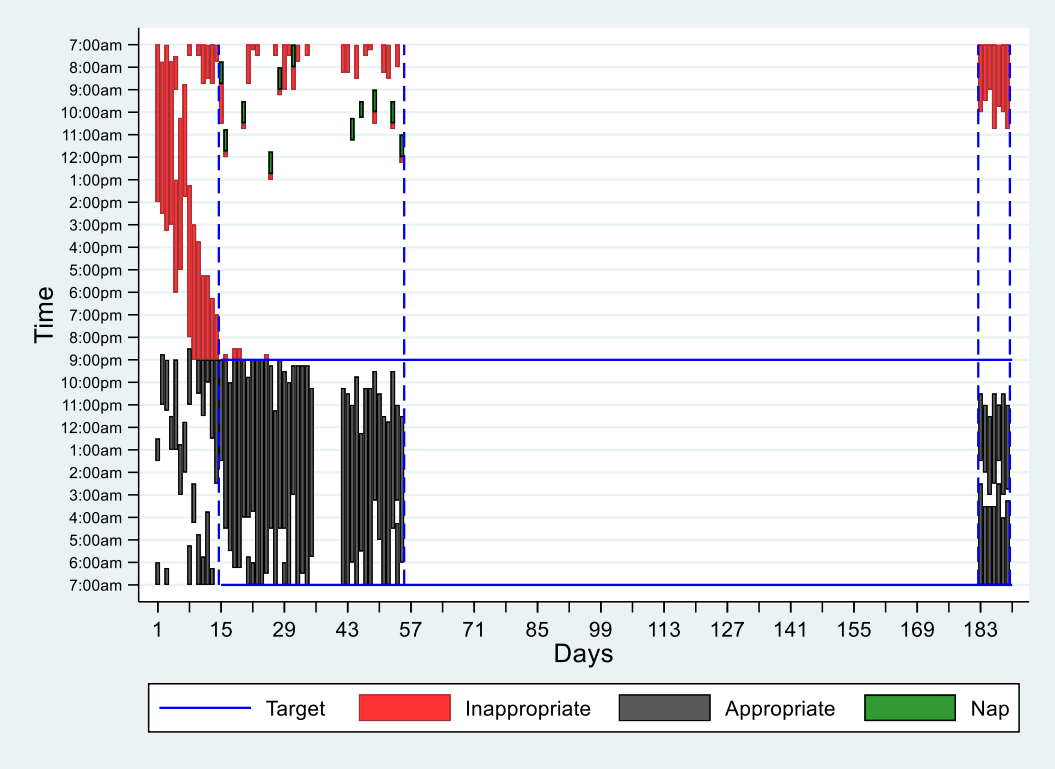
Ethan. For Ethan, sleep/wake times were problematic in baseline. This was measured across baseline, intervention, and LTFU; however, STFU is not reported. The outcome of the intervention for Ethan's sleep/wake schedule is presented in Figure 11.

Sleep/wake Schedule. Ethan's sleep/wake schedule was the most variable in baseline; he had one to three sleeps over a 24-hour period and his longest sleep period often occurred during the daytime (see Figure 11). Green bars were added to Figure 11 to represent naps, defined as age-appropriate daytime sleeping within the scheduled window of time. There was a significant shift in Ethan's sleep/wake schedule following treatment initiation; there was a reduction in inappropriate sleep and an increase in appropriate sleep, naps, and the longest period of consolidated sleep. During the final week of intervention, Ethan fell asleep at nighttime between 10.05pm and 11.45pm and woke up from this sleep between 3.20am and 8.30am though he napped if he woke 4.30am or earlier. Improvements were maintained at follow-up relative to baseline; Ethan was achieving more appropriate and consistent sleep at

conventional times though there was an increase in inappropriate sleep and a decrease in the longest period of consolidated sleep relative to the end of intervention. Overall, there was an increase in the consistency of Ethan's sleep/wake schedule (time awake/asleep) and amount of consolidated sleep following intervention.

Figure 11

Ethan's Sleep/wake Schedule (time awake/asleep) across Treatment Phases



Effects of Antecedent-based Modifications

Table 4 summarises each participant's sleep problems, treatment component(s) differentiated into antecedent- and consequence-based modifications, and the effectiveness of each treatment component. Results show that antecedent-based modifications were effective for treating sleep problems in the participants. For three participants, modification to sleep/wake times alone were effective for improving sleep problems.

Table 4

Sleep Problems, Treatment Component(s), and the Effectiveness of Treatment Component(s)

	Sleep problems	Sub-phase treatment component(s)	Effectiveness of treatment component(s)
Jack	SOD, TST, irregular sleep/wake schedule	*Antecedent-based modifications:* (1) Modification to sleep/wake times, (2) Sleep hygiene modifications (3) Relaxation exercises	*Antecedent-based modifications:* Improvements in TST and sleep/wake schedule, according to visual analysis and the PEM. Sleep problems improved immediately with (1). Further improvements with (2) and (3).
Hannah	Frequency and duration of NWs, sleep dependencies (milk, parental presence), irregular sleep/wake schedule	*Antecedent-based modifications:* (1) Modification to sleep/wake times, Gro-Clock™ (4) Relaxation exercises (positive coping self-statements, night light) *Consequence-based modifications:* (2) Systematic fading of parental presence (3) Systematic fading of nighttime milk, rewards	*Antecedent-based modifications:* Improvements in freq. and duration of NWs, according to visual analysis and the PEM. Sleep problems reduced immediately with (1) and were eliminated by the end of (4). *Consequence-based modifications:* Parental presence and nighttime milk were successfully eliminated with (2) and (3).
Luna	SOD, irregular sleep/wake schedule	*Antecedent-based modifications:* (1) Modification to sleep/wake times	*Antecedent-based modifications:* Improvements in SOD, according to visual analysis and the PEM. SOD improved sufficiently with (1).
Mason	SOD, frequency and duration of NWs, irregular sleep/wake schedule	*Antecedent-based modifications:* (1) Modification to sleep/wake times	*Antecedent-based modifications:* Improvements in SOD and freq. and duration of NWs, according to visual analysis and the PEM. Sleep problems improved immediately and sufficiently with (1).
Liam	SOD, irregular sleep/wake schedule	*Antecedent-based modifications:* (1) Modification to sleep/wake times (2) Sleep hygiene modifications (3) White noise	*Antecedent-based modifications:* Improvements in SOD, according to visual analysis and the PEM. SOD improved immediately with (1). Further improvements with (2) and (3).

Ethan	Irregular sleep/wake schedule	*Antecedent-based modifications:* (1) Modification to sleep/wake times	*Antecedent-based modifications:* Improvements in sleep/wake schedule, according to visual analysis. Sleep/wake schedule improved sufficiently with (1).

Sleep Outcome Measures

Table 5 presents pre- and post-treatment scores on the Children's Sleep Habits Questionnaire (CSHQ), Sleep Self-Report (SSR), Adolescent Sleep Hygiene Scale (ASHS), and Adolescent Sleep Wake Scale – short version (ASWS) as applicable for each participant.

Table 5

Pre- and Post-treatment Psychometric Total Scores for Sleep

	CSHQ		**SSR**		**ASHS**		**ASWS**	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Jack					4.36	5.21	4.25	4.15
Hannah	60	45						
Luna	53	44	30	26				
Mason	56	60	41	31	4.83	5.49		
Liam	46	41						
Ethan	-	-	-	-	-	-	-	-

Note. ASHS= Adolescent Sleep Hygiene Scale, ASWS= Adolescent Sleep Wake Scale, CSHQ= Children's Sleep Habits Questionnaire, SSR= Sleep Self-Report.

CSHQ. Parents completed the CSHQ at baseline and post-treatment for four of the six participants (Hannah, Luna, Liam, and Mason). All CSHQ scores were in the clinical range at baseline (≥41), ranging from 46-60. CSHQ scores reduced from 60 to 45 for Hannah, 53 to 44 for Luna, and 46 to 41 for Liam though remained within the clinical range. The other participant's (Mason) CSHQ score increased from 56 at baseline to 60 post-treatment and remained within the clinical range.

SSR. Two participants (Luna and Mason) also completed the SSR at baseline and post-treatment. It should be noted that there is no clinical cut-off score for the SSR, however,

reductions in SSR scores were evident between pre- and post-treatment from 30 to 26 for Luna and 41 to 31 for Mason.

ASHS and ASWS. Two participants (Jack and Mason) completed the ASHS, and one participant (Jack) also completed the ASWS at baseline and post-treatment. Jack and Mason's ASHS scores improved from 4.36 to 5.21 and 4.83 to 5.49, respectively. Jack's ASWS score reduced from 4.25 to 4.15 between pre- and post-treatment, indicating a decrease in subjective sleep quality. The researcher could not assess if these ASHS and ASWS scores were clinically significant as there is no reported clinical cut-off score for these measures.

Collateral Treatment Outcomes

Child Outcomes

Pre- and post-treatment scores on the Gilliam Autism Rating Scale, Third Edition (GARS-3), the Child Behavior Checklist (CBCL), the Youth Self-Report (YSR), and the Paediatric Quality of Life Inventory (PedsQL) are presented in Table 6 for five participants. The Multidimensional Anxiety Scale for Children, Second Edition (MASC 2) and the Repetitive Behaviour Scale- Revised (RBS-R) are also described for one participant.

GARS-3. Parents completed the GARS-3 at baseline for all six of the participants and post-treatment for four participants. Jack, Hannah, Luna, Liam, and Ethan's Autism Index Scores were Level 2 or 3 at baseline, indicating the probability of ASD is Very Likely. Mason's Autism Index score was classified as Level 1 suggesting ASD is Probable. Both Jack, Hannah, and Liam's Autism Index remained within Level 2 though Jack's Autism Index score reduced from 87 to 83 and Liam's increased from 78 to 83. Luna's Autism Index score reduced from 78 (Level 2) to 66 (Level 1). In terms of subscales, reductions were evident in restrictive/repetitive behaviours for Jack (9 to 8) and Hannah (11 to 9), social interaction for Jack (10 to 9), Luna (6 to 4), and Liam (10 to 9), and social communication for

Jack (7 to 5), Hannah (9 to 8), Luna (7 to 5), and Liam (7 to 6). Luna also showed reductions in emotional responses (11 to 6) and cognitive style (11 to 10). There were increases in restrictive/repetitive behaviours for Luna (4 to 5) and Liam (7 to 8), social interaction for Hannah (7 to 9), and emotional responses for Liam (4 to 6). Interestingly, there were increases in maladaptive speech for all four children, from baseline (6 to 10) to post-treatment (7 to 11). Table 6 provides an overview of the six subscale scores in addition to ASD Index scores.

CBCL and YSR. Parents completed the CBCL at baseline and post-treatment for all but one participant (Ethan). It should be noted that in Table 6 the Emotionally Reactive and Sleep Problems subscales were reported in place of the Social Problems and Thought Problems subscales for Hannah, as the CBCL 1.5-5 form was used. There was a reduction in internalising problems for two participants (Hannah and Luna), though there was also an increase in internalising problems for one participant (Mason) and externalising problems for three participants (Luna, Mason, and Liam). Of note, Mason's internalising score and Luna's externalising score moved from the Borderline range to the Clinical range. In terms of total scores, Jack's total score reduced from 59 to 58, Hannah's total score reduced from 85 to 73, Liam's total score reduced from 57 to 55, Mason's total score increased from 56 to 59, and Luna's total score increased from 62 to 66 moving from Borderline to Clinical. Three participants (Jack, Luna, and Mason) also completed the YSR. Jack's internalising problems and externalising problems reduced following intervention. Both Luna and Mason's internalising scores increased, and Mason's externalising score also increased. Jack's total score reduced from 58 to 54, Luna's total score reduced from 31 to 28, and Mason's total score increased from 48 to 49. Overall, there were mixed findings across participants and raters on the CBCL and YSR. An overview of the syndrome scale scores in addition to broadband and total scores is provided in Table 6.

PedsQL. The parents of five participants completed the PedsQL at baseline and post-treatment. In addition, Jack, Luna, and Mason also completed the self-report version of the PedsQL post-treatment. There was an increase in parent-reported quality of life total scores for Jack (62 to 71.7), Hannah (60.9 to 73.9), and Mason (father-reported; 71.7 to 77.2), and a decrease in parent-reported quality of life total scores for Liam (75 to 69.6), Mason (mother-reported; 69.6 to 64.1), and Luna (79.4 to 69.6). Interestingly, all three adolescents self-reported an increase in quality of life, with total scores from 71.7 to 72.8 for Jack, 65.2 to 73.9 for Mason, and 98.9 to100 for Luna. Table 6 provides an overview of child- and parent-reported dimension and total scores.

MASC 2. Parent-and self-report forms of the MASC 2 were completed at baseline and post-treatment by Jack and his parents. Jack's self-reported Total Score was 76 (Very Elevated) at baseline and reduced to 66 (Elevated) post-treatment. Jack's parent-reported Total Score was 78 (Very Elevated) at baseline and 77 (Very Elevated) post-treatment, suggesting little change based on parent report.

RBS-R. Liam's parents completed the RBS-R at baseline and post-treatment. Liam's parents reported an increase in stereotyped behaviour, compulsive behaviour, and sameness behaviour post-treatment, and no changes in other subscale scores.

Table 6

Pre- and Post-treatment Psychometric Syndrome Scale and Total Scores for Collateral Outcomes

	Jack		**Hannah**		**Luna**		**Mason**		**Liam**		**Ethan**	
CBCL (Parent) /YSR (Child)	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Anxious/Depressed,	*10*/*13*	9/*12*	*12*	8	*11*/0	4/0	3/5	8/7	2	1	-	-
Withdrawn/Depressed	4/8	5/7	*9*	*6*	5/0	2/1	*10*/6	*10*/7	1	1	-	-
Somatic Complaints	5/3	4/1	*8*	6	0/0	1/0	0/4	4/4	1	2	-	-
Social Problems/Emotionally Reactive*	3/6	3/6	**15*	**11*	*13*/2	7/1	4/0	2/1	3	4	-	-
Thought Problems/Sleep Problems*	*8*/7	4/3	**10*	*3	*7*/0	6/0	6/4	6/2	*8*	4	-	-
Attention Problems	7/5	9/6	*5*	6	*12*/2	*11*/0	8/4	5/5	*13*	*13*	-	-
Rule Breaking Behaviour	0/3	0/2			7/0	4/0	0/0	0/0	0	0	-	-
Aggressive Behaviour	0/0	0/0	*29*	*28*	*21*/1	13/1	0/1	4/0	2	3	-	-
Internalising	*69*/*69*	*69*/*66*	*82*	*74*	*65*/27	53/32	63/61	*70*/64	50	50	-	-
Externalising	34/40	34/37	*74*	*74*	*70*/34	*65*/34	34/34	48/37	44	46	-	-
Total	59/58	58/54	*85*	*73*	*71*/31	*66*/28	56/48	59/49	57	55	-	-
GARS-3												
Restrictive/Repetitive Behaviours	9	8	11	9	4	5	6	-	7	8	14	-
Social Interaction	10	9	7	9	6	4	6	-	10	9	13	-
Social Communication	7	5	9	8	7	5	2	-	7	6	12	-
Emotional Responses	6	6	13	13	11	6	3	-	4	6	10	-
Cognitive Style	10	10	13	13	11	10	12	-	8	8	7	-
Maladaptive Speech	9	10	10	11	6	7	6	-	9	11	7	-
ASD Index (6 scores)	87	83	105	105	78	66	63	-	78	83	105	-

Table 6

(Continued)

	Jack		Hannah		Luna		Mason		Liam		Ethan	
PedsQL (Child-reported)	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Physical Functioning	90.6	90.6	-	-	100	100	56.3	68.8	-	-	-	-
Emotional Functioning	45.0	65.0	-	-	95.0	100	70.0	65.0	-	-	-	-
Social Functioning	65.0	65.0	-	-	100	100	75.0	90.0	-	-	-	-
School Functioning	75.0	60.0	-	-	100	100	65.0	75.0	-	-	-	-
Total Score	71.7	72.8	-	-	98.9	100	65.2	73.9	-	-	-	-
PedsQL (Parent-report)												
Physical Functioning	84.4	84.4	78.1	93.8	93.8	84.4	81.25/81.25*	78.1/81.3*	100	90.6	-	-
Emotional Functioning	35.0	65.0	35.0	50.0	55.0	70.0	75.0/85.0*	50.0/65.0*	80.0	80.0	-	-
Social Functioning	55.0	55.0	55.0	60.0	65.0	50.0	55.0/50.0*	45.0/70.0*	45.0	50.0	-	-
School Functioning	60.0	75.0	65.0	80.0	95.0	65.0	60.0/65.0*	75.0/90.0*	60.0	45.0	-	-
Total Score	62.0	71.7	60.9	73.9	79.4	69.6	69.6/71.7*	64.1/77.2*	75.0	69.6	-	-

Note. CBCL and YSR scores reported in italics are above the clinical cut-off. ASD= autism spectrum disorder, CBCL= Child Behavior Checklist, GARS-3= Gilliam Autism Rating Scale, Third Edition, YSR= Youth Self-Report, PedsQL= Pediatric Quality of Life Inventory. *Mother-/father-reported.

Parent Outcomes

Table 7 below presents comparisons of pre- and post-treatment Depression, Anxiety and Stress Scale (DASS-21), Relationship Quality Index (RQI), and Pittsburgh Sleep Quality Index (PSQI) scores for parents of five participants.

Table 7

Pre- and Post-treatment Psychometric Total Scores for Parent Outcomes

	DASS-21												**RQI**				**PSQI**			
	Mother-report						Father-report						Mother-report		Father-report		Mother-report		Father-report	
	Pre			Post			Pre			Post			Pre	Post	Pre	Post	Pre	Post	Pre	Post
	D	A	S	D	A	S	D	A	S	D	A	S								
Jack	1	2	6	0	4	8	-	-	-	-	-	-	-	-	-	-	4	7	-	-
Hannah	1	3	5	0	0	2	1	4	4	2	1	3	-	-	-	-	10	3	4	5
Luna	2	0	7	1	1	6	2	0	5	8	3	10	36	36	36	38	10	6	5	7
Mason	1	1	3	0	1	1	2	1	1	0	0	5	45	44	39	37	12	13	5	6
Liam	1	3	3	3	2	2	3	6	12	1	2	8	29	29	37	45	12	9	4	4
Ethan	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-

Note. A= anxiety, D=depression, DASS-21= Depression Anxiety Stress Scale, PSQI= Pittsburgh Sleep Quality Index, RQI= Relationship Quality Index, S= stress.

DASS-21. Nine parents (five mothers; four fathers) completed the DASS-21 at baseline and post-treatment. All scores on the Depression axes were in the Normal range at baseline, ranging from 1-3. Depression scores reduced and remained within the Normal range for most (4/5) mothers and half (2/4) of the fathers. Two fathers experienced an increase in their depression scores from 1-2 to 2-8, with one score remaining within the Normal range and one score moving to the Moderate range post-treatment. Most (7/9) scores on the Anxiety axes were in the Normal range at baseline, ranging from 0-6, and almost all remained within the Normal range at post-treatment. However, one mother's score was in the Normal range at baseline and increased from 2 (Normal) to 4 (Mild) post-treatment. Anxiety scores for two fathers were in the Mild or Moderate range at baseline (4 and 6) and reduced to the Normal

range post-treatment (1 and 2). Scores on the Stress axes were variable: five scores were within the Normal range and reduced post-treatment, one score reduced from Moderate to Mild (12 to 8) post-treatment, one increased from Normal to Mild (6-8), one increased from Normal to Moderate (5 to 10), and one increased but remained in the Normal range (1 to 5).

RQI. Three sets of parents completed the RQI at baseline and post-treatment. Five out of six RQI scores were above the cut-off indicative of good relationship quality, at baseline (36-45) and post-treatment (36-45). For the other parent, the RQI score was equal to the cut-off at baseline and post-treatment (29), indicating poor relationship quality.

PSQI. Nine parents (five mothers; four fathers) completed the PSQI at baseline and post-treatment. Global PSQI scores reduced from 10-12 to 3-9 for three parents indicating better sleep quality following the intervention. There was, however, an increase in PSQI scores for five parents from 4-12 to 5-13 indicating sleep quality slightly deteriorated post-treatment.

Treatment Acceptability

Treatment Acceptability Rating Form-Revised (TARF-R)

Parents for all six participants (five mothers; four fathers) and one adolescent (Jack) completed the TARF-R post-treatment. Post-treatment TARF-R subscale and total scores are presented in Table 8. Maximum scores for Reasonableness, Effectiveness, Side-Effects, Disruption/Time Consuming, and Willingness were 21, Cost and Problem Severity were 14, Understanding was 7, and Total Acceptability was 119. Parent-reported Total Acceptability scores ranged from 80-108, and Jack's total acceptability score was 103. Mean parent ratings were high for Reasonableness (*M* = 19.22; range = 16-21), Effectiveness (*M* = 18.11; range = 13-21), Willingness (*M* = 19.78; range = 16-21), Cost (M = 12; range = 8-14) and Understanding (*M* = 6.56; range = 6-7), and lower for Side Effects (*M* = 16.11; range = 7-21), and Disruptive/Time Consuming (*M* = 13; range = 7-18). Overall, parent ratings suggest that

the intervention was experienced as acceptable, reasonable, effective, low cost, they had a clear understanding of the intervention, and were willing to carry it out. But for some parents, it was costly, there were side-effects, and it was disruptive to the family routine or time-consuming. It should be noted that for the one participant (Hannah) for whom consequence-based modifications were implemented, the TARF-R did not differentiate between antecedent- and consequence-based modifications. This information was provided in the post-treatment interview by Hannah's parents.

Post-treatment Interview

Post-treatment interviews were conducted with five children's parents (Hannah, Luna, Liam, Mason, and Ethan) and two adolescents (Jack and Mason). Several common themes emerged from the responses provided by the parents and adolescents. All parents and adolescents reported that the intervention was challenging at times, particularly at the start with modification to sleep/wake schedules. Parents noted that they found this treatment component difficult when their child was tired and difficult to wake up. Another common challenge with modification to sleep/wake schedules was implementing the set schedule within the child or family's normal routine and adhering to it on the weekend. Despite these challenges, all parents and adolescents reported that it was worthwhile for the positive change. Further, parents reported that the continual support and encouragement from the researchers and sleep psychoeducation were helpful when faced with these challenges. Another theme was the collateral benefits for the child, their parents, and the family. Parents reported improvements in their child's school functioning, attributions about sleep, coping skills, sleep independence, self-awareness of sleep, energy levels, talking, eye contact, focus, following of instructions, and/or activity levels. Some parents also reported improvements in their own sleep, energy, and productivity, and that there were benefits for the family unit including better evening and morning routines, and less stress before bedtime. Jack reported

during the post-treatment interview that he noticed improvements in his alertness, concentration, memory, and mood. Only Hannah's parents were able to comment on their experience implementing consequence-based modifications. Hannah's parents reported that these modifications were the biggest challenge for the family. More specifically, fading nighttime milk was the most difficult though it was helpful that this was completed gradually to suit the family's needs.

Reliability

Inter-observer Agreement (IOA)

IOA data was collected for 5/6 participants. IOA was calculated as planned for Hannah and Liam using video recording against sleep diaries for 25% of nights across study phases. Using actigraph data against sleep diaries, IOA was able to be calculated for 10 nights (7%) for Jack's bedtime, 29 nights (28%) for Luna's SOL, bedtime, and wake time, and 3 nights (3%) for Mason's SOL and bedtime. Mean IOA was 92% for video data (range= 87-98), 78% for actigraph data (range= 60-100%), and 84% overall.

Treatment Fidelity

Treatment fidelity was calculated for 5/6 participants for 25% of nights across intervention. For consistency across participants, treatment fidelity was based on adherence to P1 i.e., modification to sleep/wake schedules. Treatment fidelity was not calculated for Ethan as there was ambiguity with the age-appropriate daytime sleep (i.e., naps) that occurred outside of the set sleep/wake schedule. Treatment fidelity for P2 was calculated as planned for Jack, though P2 and P3 were not calculated for Hannah as there was ambiguity with the parent progressing through each step to suit their needs. Mean treatment fidelity was 95% (range= 91-99).

Table 8

Post-treatment Scores on the TARF-R

	Jack		Hannah		Luna		Mason		Liam		Ethan		Maximum Score
	Mother	Self	Mother	Father	Mother	Father	Mother	Father	Mother	Father	Mother	Father	
Reasonableness	19	18	20	21	17	16	20	18	21	21	-	21	21
Effectiveness	20	20	21	21	17	15	17	18	21	21	-	13	21
Side Effects	17	16	14	18	7	12	17	19	17	21	-	21	21
Disruptive/Time Consuming	16	16	15	13	9	10	12	18	7	12	-	18	21
Cost	12	14	8	14	14	14	14	14	14	10	-	8	14
Willingness	19	19	21	20	16	18	20	21	21	21	-	21	21
Problem Severity*	10	5	11	11	8	8	7	9	10	5	-	14	14
Understanding*	7	6	7	7	6	6	6	6	7	7	-	7	7
Total Acceptability Score	103	103	99	107	80	85	100	108	101	106	-	102	119

Note. *Subscales not included in the Total Acceptability Score

Chapter Five: Discussion

Research Questions

The overall aim of this research was to identify whether modification to sleep/wake schedules and other antecedent-based modifications are effective for treating sleep problems among children on the autism spectrum. This research was divided into two parts. Part one involved a systematic review of the literature that has investigated the effectiveness of modification to antecedent variables for treating sleep problems in children on the autism spectrum (presented in Chapter Two), and Part Two used a single-case AB design to separately investigate the effectiveness of modification to sleep/wake schedules and other antecedent-based modifications as well as consequence-based modifications if necessary. The following research aims were also addressed in Part two: (1) to explore how parents of children on the autism spectrum perceive the acceptability of modification to sleep/wake schedules and other antecedent-based modifications, (2) to examine the effect of an improvement in children's sleep on child wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parental mental health, sleep, and relationship quality, and (3) examine whether reductions in sleep problems are maintained at STFU and LTFU. The key research question guiding both Part one and two was: Are modifications to sleep/wake schedules and/or other antecedent-based modifications, effective in reducing sleep problems in children on the autism spectrum?

The Effect of Modification to Antecedent Variables including Sleep/wake Schedules

In the present study, all six participants started intervention with modification to sleep/wake schedules. This procedure was introduced first as it was considered to be less restrictive. Each participant's set bedtime and wake time was selected from the assessment information, noting the average sleep onset and wake time. These set times were modified throughout the intervention as needed until the family's goal sleep/wake times had been met.

All participants demonstrated an immediate improvement in the consistency of their sleep/wake schedule (time awake/asleep) and the longest period of consolidated nighttime sleep. Sleep onset latency also reduced for all participants (3/3) for whom this was problematic in baseline. For Jack, SOL was not a target for treatment as it was non-problematic in baseline. Evidently, his SOL increased during treatment. There was an increase in TST for the one participant (Jack) for whom this was problematic in baseline. Both frequency and duration of NWs reduced for both participants who demonstrated difficulties in this area (Hannah and Mason). Following intervention, three out of four participants (Hannah, Luna, and Liam) showed a reduction in parent-reported sleep problems on the CSHQ. Two adolescents (Luna and Mason) completed the SSR and self-reported a reduction in sleep problems. In addition, each of the participants who completed the ASHS (Jack and Mason) self-reported improved sleep hygiene practices though one participant (Jack) also completed the ASWS and reported reduced sleep quality.

Further antecedent-based modifications were required to be implemented with three participants (Jack, Hannah, and Liam). These included sleep hygiene modifications (e.g., removing screens and preferred activities in bed before bedtime, modifying bedroom temperature, consistent bedtime routine), stimulus control (e.g., white noise), and relaxation exercises (e.g., diaphragmatic breathing). However, no further treatment components were necessary to implement for the three remaining participants (Luna, Mason, and Ethan) as sleep problems had reduced and the family's goals had been achieved. All three of these participants (Jack, Hannah, and Liam) demonstrated improvements following the introduction of these modifications. Improvements were evident in Jack's TST relative to baseline, though there was a slight decrease compared to modification to sleep/wake schedules alone. Further, Liam's SOL reduced with sleep hygiene modifications relative to baseline and modification to sleep/wake times. For one participant (Hannah) it was necessary

to implementation other antecedent-based modifications (relaxation exercises, positive coping self-statements, night light) as well as consequence-based modifications. Whilst modification to sleep/wake schedules were effective for reducing sleep problems, Hannah's sleep dependencies (i.e., parental presence, nighttime milk) interfered with her ability to settle independently. Consequence-based modifications (faded parental presence, fading nighttime milk) were used to fade these sleep dependencies and achieve the family's goal of Hannah sleeping independently through the night. As a result of these antecedent- and consequence-based modifications, the frequency and duration of Hannah's NWs reduced to zero by the end of intervention. Overall, modification to sleep/wake schedules alone were sufficient for improving Luna, Mason, and Ethan's sleep problems. Other antecedent-based modifications were also implemented for Jack, Hannah, and Liam, and consequence-based modifications were needed to fade Hannah's sleep dependencies. Thus, modification to sleep/wake schedules and other antecedent-based modifications were sufficient alone or in combination for all participant's sleep problems though further modifications were needed for Hannah's sleep dependencies.

These findings are consistent with the systematic review (Part one; Ford et al., 2021) in which five studies were identified that investigated the effectiveness of modification to sleep/wake schedules using similar procedures to the present study (i.e., sleep/wake scheduling, faded bedtime). Findings from Ford et al. (2021) showed that these modifications were effective for improving SOL, TST, and frequency of NWs, across participants aged 2 to 18 years (for e.g., Delemere & Dounavi, 2018; DeLeon, 2004; Luiselli et al., 2020; Luiselli et al., 2021; van Deurs et al., 2019). Ford et al. (2021) also highlighted that modification to sleep/wake schedules alone, are not necessarily sufficient to treat sleep problems in all children, and further antecedent-based modifications such as positive bedtime routines (with visual schedules), sleep restriction and positive bedtime routines together, white noise,

stimulus control, relaxation exercises, and social stories can be necessary to effectively resolve sleep problems. Further, some cases also needed consequence-based modifications.

Findings from Part one and two are consistent with a wider base of existing research. Previous research has demonstrated that modification to sleep/wake schedules (e.g., faded bedtime) is effective for treating sleep problems in children with other types of developmental disabilities, including ADHD and intellectual disability (e.g., Piazza & Fisher, 1991). Research has also demonstrated the effectiveness of this procedure with younger typically developing children aged 1.5 to 4 years, using a parent education approach (Cooney et al., 2018). Further, previous research has demonstrated the utility of other antecedent-based modifications for treating sleep problems such as sleep hygiene modifications (Vriend et al., 2011). Indeed, there is a large body of research supporting the utility of sleep hygiene as an essential component for treating sleep problems in children (Hall & Nethery, 2019), though it may not be sufficient alone (Stepanski & Wyatt, 2003). In the present study, sleep hygiene modifications were implemented for Jack and Liam following modification to sleep/wake schedules. For Jack and Liam, sleep hygiene modifications were not sufficient as standalone treatment components. It was necessary to implement a further treatment component (i.e., relaxation exercises or white noise) for both participants following sleep hygiene modifications.

With the addition of white noise, there was a small improvement in Liam's SOL by the end of intervention. This finding is also consistent with previous research demonstrating the effectiveness of white noise for reducing sleep problems in infants and toddlers (e.g., Forquer & Johnson, 2005). Further, relaxation exercises were implemented for both Jack and Hannah as the final component of their individualised intervention. There was a small improvement in Jack's TST and Hannah's NWs were eliminated following the introduction of relaxation exercises. However, it should be noted that relaxation exercises were used in

addition to positive coping self-statements and a night light for Hannah. Previous research also supports the effectiveness of relaxation exercises for reducing sleep problems, as part of a cognitive behavioural intervention for adolescents with and without autism (e.g., Hendricks et al., 2014; van Deurs et al., 2021). This present study adds to preliminary research that documents the effectiveness of these procedures individually.

Parental Ratings of Treatment Acceptability

Findings from the post-treatment interviews indicated that the parents and adolescents experienced intervention as acceptable despite the challenges. A theme emerged that the intervention was challenging at times, but the positive change made it worthwhile. Another common theme was the collateral benefits for the child (e.g., daytime behaviour and functioning), their parents (e.g., sleep, energy), and the family unit (e.g., better routines, less stress before bedtime). Hannah's parents reported that consequence-based modifications (i.e., fading nighttime milk, faded parental presence) were the most difficult treatment components for the family to implement. These outcomes from the post-treatment interviews were also evident in the TARF-R results. Overall, parents and Jack all experienced the intervention as acceptable, reasonable, effective, and low cost. Also, they reported having a clear understanding and willingness to implement the intervention. There were, however, some parents that indicated it was costly, disruptive or time consuming, and there were side-effects.

The findings from the post-treatment interview and TARF-R are consistent with previous research demonstrating high levels for reasonable, effective, willingness, cost and understanding on the TARF-R for FBA-informed, sleep interventions for children on the autism spectrum (e.g., McLay et al., 2017; van Deurs et al., 2019). In addition, van Deurs et al. (2019) also reported lower or mixed scores for side-effects and disruptive or time-consuming aspects. Given that modification to antecedent variables provides a less restrictive, minimally sufficient approach, parents' perception that it was acceptable and

effective though there were mixed reports for cost, side-effects and disruption was a noteworthy finding. This finding may link to the definition of least restrictive introduced in Chapter One of this thesis (i.e., least restrictive refers to the least restrictive of the available alternative treatment components). While antecedent-based modifications may be less restrictive than consequence-based modifications, there may be aspects of these procedures that are perceived as restrictive by some families.

Collateral Outcomes for Children

Findings from this study demonstrated that improved sleep resulted in some improvements in the children's wellbeing, daytime behaviour, ASD symptomatology and quality of life, though not across all participants. ASD Index scores indicated that overall ASD symptomatology reduced or remained unchanged for three of the four participants whose parents completed the GARS-3 at pre- and post-treatment. This finding is consistent with previous research which suggests that improvements in ASD symptoms can be a collateral benefit of behavioural sleep interventions (Malow et al., 2014; McCrae et al., 2020; McLay, France, Blampied, van Deurs, et al., 2021; Reed et al., 2009). Further, research also suggests a bidirectional relationship between the severity of ASD symptoms and sleep problems (Adams, Matson, Cervantes, et al., 2014). In the present study, one participant (Liam) would lie in bed and engage in stereotypy for a long period of time. Liam's opportunity to engage in stereotypy was reduced with the implementation of a structured bedtime routine and subsequent improvements were evident in his sleep. The findings of the present study provide evidence for the relationship between ASD symptoms and sleep problems.

Mixed results were reported across participants and raters on the CBCL and the YSR. CBCL results indicated that internalising problems reduced or remained constant for four participants and increased for one participant, externalising problems remained constant for

two participants and increased for three participants, and total scores reduced for three participants and increased for two participants. YSR results indicated that internalising problems reduced for one participant and increased for two participants, externalising problems reduced or remained constant for two participants and increased for one participant, and total scores reduced for two participants and increased for one participant. This finding of mixed results across participants, raters and scales is inconsistent with existing research. Previous research in children on the autism spectrum has shown improvements on internalising and externalising subscales of the CBCL (e.g., anxious/depressed, withdrawn/depressed, attention) post-treatment at the group level (Malow et al., 2014; McLay, France, Blampied, van Deurs, et al., 2021), and improvements in CBCL total scores (i.e., internalising and externalising problems) across participants (Moon et al., 2011). There are several possible reasons for this inconsistency between the findings of the present study and existing research. For example, there were a small number of participants in the present study relative to other studies (e.g., Malow et al., 2014). Further, Malow et al. (2014) analysed the mean score of all participants and this study presented results for individuals.

The MASC 2 was also completed by Jack and his parents. Improvements in the total score were parent- and self-reported between pre- and post-treatment. This finding is consistent with previous research demonstrating improved sleep following behavioural intervention also resulted in a reduction in anxiety for children on the autism spectrum (Loring et al., 2018). It should be noted that this finding is for one participant and limited conclusions can be drawn, despite the consistency with previous research. Jack's individualised intervention included relaxation exercises (i.e., diaphragmatic breathing) to alleviate anxiety before bedtime. Therefore, the present study may provide evidence for a bidirectional relationship between anxiety and sleep problems. A wider body of research has

also investigated integrating sleep and anxiety interventions for children due to the bidirectional relationship between these problems (for e.g., Clementi & Alfano, 2020).

PedsQL parent-report scores indicated an increase in quality of life for two participants, a decrease for two participants, and mixed results for one participant. Three out of three adolescents whom completed the self-report form reported an increase in quality of life. These findings suggest that adolescents on the autism spectrum may experience an increase in their quality of life with improved sleep though their parents may not share this attribution. This finding is consistent with previous research that has demonstrated a negative association between parent-reported sleep problems and quality of life among children on the autism spectrum (Delahaye et al., 2014), and that behavioural sleep interventions can improve quality of life for these children (Malow et al., 2014; Papadopoulos et al., 2019) and children with other developmental disabilities (e.g., ADHD; Hiscock et al., 2015). This suggests that effective sleep interventions can also improve children's quality of life, and adolescents perceive their quality of life as poorer when experiencing sleep problems. These findings fit within a wider research base showing the significance of sleep problems for quality of life, across childhood to adolescence and adulthood (for e.g., Williamson et al., 2020; Deserno et al., 2019).

Collateral Outcomes for Parents

Findings suggest that improvements in the children's sleep problems can also have an effect on parental mental health. Most parents experienced a reduction in depression or stress and around half experienced a reduction in anxiety, according to DASS-21 axes scores. This finding is consistent with previous research showing an association between parental mental health and sleep problems in children on the autism spectrum (Hodge et al., 2013; Tilford et al., 2015), and improved parental mental health following behavioural intervention for sleep problems in typically developing infants (Hiscock et al., 2011). These findings fit within a

wider research base showing that parenting a child on the autism spectrum is associated with more mental health difficulties than parents of typically developing children or children with other disabilities (Da Paz & Wallander, 2017; Hodge et al., 2013), and adds to the research base for interventions that can improve parental mental health (McLay, France, Blampied, van Deurs, et al., 2021).

As mentioned above, relationship quality reduced or remained unchanged for most parents with improvements only seen for two out of six parents on the RQI. There is a paucity of research examining the effect of behavioural sleep interventions on parental relationship quality though one study has investigated this relationship and reported a small improvement in relationship quality (McLay, France, Blampied, van Deurs, et al., 2021). It is possible that relationship quality may have been unchanged or deteriorated as the relationship between children's sleep and parent's relationship quality is influenced by another variable, such as parental mental health, stress levels, or the stress associated with implementing an intervention. There is a complex relationship between these variables, for example, Weitlauf et al. (2014) investigated relationships between children's problem behaviours, relationship quality, parenting stress, and depression symptoms in mothers of children on the autism spectrum. They found that relationship quality needs to be considered in the interaction between children's problem behaviours and mother's depression, and that relationship quality moderates the relationship between parenting stress and depression symptoms (Weitlauf et al., 2014).

Likewise, three out of nine parents reported improvements in subjective sleep quality on the PSQI. Given that parents of children on the autism spectrum with sleep problems have poorer sleep (Meltzer, 2008; Meltzer & Mindell, 2007), it would follow that an effective behavioural sleep intervention would also improve parental sleep. However, it is possible that implementing a sleep intervention may initially result in sleep loss for parents. Further, the

post-treatment psychometric measures were implemented immediately following the intervention. It may take time for parents to re-establish their own sleep habits and patterns once their child's sleep has improved. Very few studies have investigated the effect of improvements in children's sleep on parental sleep, though one study has found small change in parental sleep quality following sleep intervention (McLay, France, Blampied, van Deurs, et al., 2021).

Maintenance of Treatment Effects

In the present study, most improvements in the children's sleep were maintained at STFU and LTFU. There was a small increase in inappropriate sleep at LTFU for Jack and Ethan, and an increase in the frequency and duration of NWs for Mason though improvements were evident compared to baseline. This is noteworthy as it implies that families were willing and able to implement the intervention without the support of the researchers. Therefore, it suggests that intervention is not only effective, but was well understood by the parents. These are important considerations for treating sleep problems in this group. This is particularly important given the impact and the pervasiveness of sleep problems in children on the autism spectrum. This finding is consistent with previous research demonstrating maintenance of treatment effects at follow-up for children on the autism spectrum following behavioural sleep interventions (McCrae et al., 2020; Moon et al., 2011).

Strengths and Limitations of the Present Study

The current study had noteworthy strengths including the published systematic review, the use of instrumented sleep measures, the assessment of social validity, IOA, and treatment fidelity, the single-case AB design, the use of FBA to inform intervention, and the use of PEM for analysing dependent variables. The completion of a systematic review allowed the researcher to appraise the evidence base pertaining to modification to sleep/wake

schedules and other antecedent-based modifications, while conducting research with participants implementing these procedures. Aside from allowing evidence to inform practice, this also allowed the research questions to be answered using both existing research and results from the present study. The use of instrumented sleep measures (i.e., actigraphs and video recording) also strengthened the study design as it enabled the calculation of IOA. This allowed the researcher to measure and thereby draw conclusions on the reliability of the data. In addition, the researcher calculated treatment fidelity for most participants. This reliability measure strengthened the researcher's ability to attribute a reduction in sleep problems to the intervention itself. Treatment fidelity was high for all participants for whom this was calculated (i.e., 91-99%). The single-case AB research design was a strength in the context of evaluating an individualised, FBA-based intervention as it allowed for the staggered introduction of individualised interventions i.e., starting with modification to sleep/wake schedules as a less restrictive treatment component relative to subsequent treatment components. Other study designs may implement a placebo or procedures not necessary or helpful for treating the participant's specific sleep problems. Further, the use of FBA provided each participant with an individualised intervention that was able to be modified to suit their needs. This approach also aligns with the principles of least restriction and minimal sufficiency in treatment. Finally, another strength is the use of PEM as a single-case measure of effect size for the dependent variables (i.e., SOL, TST, NWs). This was a strength as it allowed the researcher to evaluate the magnitude of treatment effects.

Although this study had many strengths, there were also limitations that future research should take into consideration. The researcher intended to employ a multiple baseline across participants design though the research is a single-case AB design as the same procedure was not implemented across participants with different baseline lengths. The use of a single-case AB design has a number of limitations including a lack of replication of

baseline (A) or intervention (B) study phases and a lack of control for confounding variables or internal validity, that is provided with using a multiple baseline design (Backman et al., 1997; Kratochwill & Levin, 2010). Given that this study collected pre- and post-treatment data and there was no control condition, improvements may be due to the intervention or the result of positive change accumulating over time.

A further limitation of the current study is the quantity of missing data. Firstly, there were technological issues with two out of the three actigraph devices. As a result, IOA was only calculated for 7% of nights for Jack and 3% of nights for Mason which is significantly less than the planned 25% of nights across study phases. This means that the researcher was unable to measure with confidence the reliability of Jack and Mason's reporting in sleep diaries. In addition, Mason did not complete LTFU, and Ethan only completed LTFU and not STFU. This limited the researcher's ability to assess the maintenance of treatment effects for these participants, and the family's implementation of intervention without support. Similarly, the researcher was the lead clinician on two cases and there were different lead clinicians on the other four cases. This means that it is possible that there was not a uniform approach as evident through differences between participants in the psychometric measures completed at pre- and post-treatment. However, the study team did have team meetings for all of these cases and some differences in the chosen psychometrics were due to the child's age, developmental stage, and/or cognitive abilities.

Another limitation is the reliance on non-instrumented sleep measures such as parent- and self-report sleep diaries and psychometric measures. Whilst instrumented measures were used in the current study, this was for a small number of nights to calculate IOA and non-instrumented methods were primarily used to collect data. As a result, the researcher was reliant on self-report by the child and/or parent which may be less reliable than an instrumented measure. Further, treatment fidelity was calculated for modification to

sleep/wake schedules alone for most participants rather than across all treatment components. Finally, the small number of participants in the current study means there is a lack of a foundation to support generalisation to other children on the autism spectrum with sleep problems. Each participant's intervention was unique and individualised based upon sleep problems, precipitating and/or maintaining factors, and the hypothesised function(s). These individualised interventions may not necessarily generalise to other children on the autism spectrum for whom do not share these unique formulations.

Future Research

The strengths and limitations of the current study have a number of implications for future research. Firstly, the reliability of the data collected could be enhanced by including both instrumented and non-instrumented measures (i.e., video recordings and actigraphs alongside sleep diaries). In addition to enhanced reliability, there are advantages of using both subjective and objective measures. For example, sleep diaries are useful tools for collecting information from the parents' perspective about parent-child interactions and reporting of events occurring outside the capacity of video recording, such as challenging behaviour before bedtime outside the child's bedroom. Also, there were technological issues with actigraph devices in the present study. Thus, using both instrumented and non-instrumented measures may reduce missing data.

Further, a multiple baseline across participants design could be employed to increase experimental control (i.e., to demonstrate behaviour change when and only when the independent variable is introduced) and increase the researcher's ability to attribute the findings to intervention alone. It would also be recommended to calculate treatment fidelity for a greater percentage of nights across all cases for this reason.

In the current study, the effectiveness of modification to sleep/wake schedules and other antecedent-based modifications was examined as a pool of interventions sharing the

common feature of being antecedents to the problem behaviour. Future research could aim to investigate the effectiveness of individual procedures within these categories, and create a hierarchy of minimally sufficient, less restrictive interventions. More specifically, a hierarchy could be created to provide recommendations by type of sleep problem (i.e., sleep onset delay, night wakings), antecedent and consequence variables, and hypothesised function(s) of sleep behaviour (i.e., attention, escape, tangible). This would require a larger sample size and replication across a wider range of participants with varying characteristics. In addition, future research could assess the feasibility and acceptability of individual antecedent-based modifications, to inform our knowledge of less restrictive interventions.

Clinical Implications

The current study presents evidence for the utility of modification to sleep/wake schedules and other antecedent-based modifications for treating sleep problems among children on the autism spectrum. Behavioural interventions are commonly used to treat sleep problems in children on the autism spectrum and with typically developing children. These interventions are often comprised of both antecedent- and consequence-based modifications, or the use of extinction procedures in isolation (e.g., Knight & Johnston, 2014; McLay et al., 2017; Weiskop et al., 2005). This study provides evidence for effective FBA-informed sleep interventions that avoid consequence-based modifications and potential child and parental distress associated with such procedures (i.e., post-extinction response bursts). The findings also suggest that consequence-based modifications may be necessary at times when the sleep-interfering behaviour is maintained by external reinforcement including attention or tangible reinforcers. However, they suggest that less restrictive treatment components can justifiably be introduced before consequence-based modifications, as outlined in this study.

Overall, parents experienced the intervention as acceptable. Social validity is important to consider when planning an intervention programme as this may increase the

family's adherence to intervention (i.e., treatment fidelity) and therefore the likelihood of the intervention being successful. An effective intervention can also lead to collateral benefits for the child and family. Among collateral benefits, this study found some improvements for the children's wellbeing, daytime behaviour, ASD symptomatology and quality of life, and parental mental health and sleep. Therefore, implementation of these interventions may also provide other positive outcomes for the child and their parents. Further, this intervention may be considered for the purpose of improving problems other than the child's sleep such as parental mental health.

Conclusion

The present study suggests that treatments involving modification to antecedent variables including modification to sleep/wake schedules are effective sleep interventions for children on the autism spectrum. There were some positive findings for children's wellbeing, daytime behaviour, ASD symptomatology, and quality of life, and parental mental health and sleep, though these findings were variable and future research is warranted in this area. Parents experienced the intervention as acceptable which suggests that antecedent-based modifications provide a less restrictive intervention approach. Overall, this study adds to the research base on the utility of antecedent-based modifications as standalone interventions for treating sleep problems in children on the autism spectrum, and provides evidence for a less restrictive, minimally sufficient intervention for this group.

References

Achenbach, T. M., & Rescorla, L. A. (2000). *Manual for the ASEBA preschool forms and profiles*. ASEBA.

Achenbach, T. M., & Rescorla, L. A. (2001). *Manual for the ASEBA school-age forms & profiles*. ASEBA.

Adams, H. L., Matson, J. L., Cervantes, P. E., & Goldin, R. L. (2014). The relationship between autism symptom severity and sleep problems: Should bidirectionality be considered?. *Research in Autism Spectrum Disorders, 8*(3), 193-199. https://doi.org/10.1016/j.rasd.2013.11.008

Adams, H. L., Matson, J. L., & Jang, J. (2014). The relationship between sleep problems and challenging behavior among children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *8*(9), 1024-1030. https://doi.org/10.1016/j.rasd.2014.05.008

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). https://doi.org/10.1176/appi.books.9780890425596

Atladóttir, H. Ó., Thorsen, P., Østergaard, L., Schendel, D. E., Lemcke, S., Abdallah, M., & Parner, E. T. (2010). Maternal infection requiring hospitalization during pregnancy and autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *40*, 1423-1430. https://doi.org/10.1007/s10803-010-1006-y

Backman, C. L., Harris, S. R., Chisholm, J. A. M., & Monette, A. D. (1997). Single-subject research in rehabilitation: A review of studies using AB, withdrawal, multiple baseline, and alternating treatments designs. *Archives of Physical Medicine and Rehabilitation, 78*(10), 1145-1153. https://doi.org/10.1016/S0003-9993(97)90142-8

Bathory, E., & Tomopoulos, S. (2017). Sleep regulation, physiology and development, sleep duration and patterns, and sleep hygiene in infants, toddlers, and preschool-age

children. *Current Problems in Pediatric and Adolescent Health Care*, *47*(2), 29-42. https://doi.org/10.1016/j.cppeds.2016.12.001

Bell, B. G., & Belsky, J. (2008). Parents, parenting, and children's sleep problems: Exploring reciprocal effects. *British Journal of Developmental Psychology*, *26*(4), 579-593. https://doi.org/10.1348/026151008X285651

Blackmer, A. B., & Feinstein, J. A. (2016). Management of sleep disorders in children with neurodevelopmental disorders: A review. *Pharmacotherapy: The Journal of Human Pharmacology and Drug Therapy*, *36*(1), 84-98. https://doi.org/ 10.1002/phar.1686

Blampied, N. M. (2013). Functional behavioural analysis of sleep in infants and children. In A. R. Wolfson & H. E. Montgomery-Downs (Eds.), *The Oxford handbook of infant, child and adolescent sleep and behavior* (pp. 169-188). Oxford University Press. https://doi.org/10.1093/oxfordhb/9780199873630.013.0013

Blampied, N. M., & France, K. G. (1993). A behavioral model of infant sleep disturbance. *Journal of Applied Behavior Analysis*, *26*(4), 477-492. https://doi.org/10.1901/jaba.1993.26-477

Bodfish, J. W., Symons, F. J. & Lewis, M. H. (1998). *The Repetitive Behavior Scale: A test manual.* Western Carolina Center Research Reports.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30,* 237–243. https://doi.org/10.1023/A:1005596502855

Borbély, A.A. (1982). A two-process model of sleep regulation. *Human Neurobiology, 1*(3), 195-204.

Borbély, A. A., & Achermann, P. (1992). Concepts and models of sleep regulation: An overview. *Journal of Sleep Research*, *1*(2), 63-79. https://doi.org/10.1111/j.1365-2869.1992.tb00013.x

Borbély, A. A., Daan, S., Wirz-Justice, A., & Deboer, T. (2016). The two-process model of sleep regulation: A reappraisal. *Journal of Sleep Research*, *25*(2), 131-143. https://doi.org/10.1111/jsr.12371

Bourgeron, T. (2007). The possible interplay of synaptic and clock genes in autism spectrum disorders. *Cold Spring Harbor Symposia on Quantitative Biology*, *72*, 645-654. https://doi.org/10.1101/sqb.2007.72.020

Bowden, N., Thabrew, H., Kokaua, J., Audas, R., Milne, B., Smiler, K., Stace, H., Taylor, B., & Gibb, S. (2020). Autism spectrum disorder/takiwātanga: An integrated data infrastructure-based approach to autism spectrum disorder research in New Zealand. *Autism*, *24*(8), 2213-2227. https://doi.org/10.1177/1362361320939329

Braam, W., Smits, M. G., Didden, R., Korzilius, H., Van Geijlswijk, I. M., & Curfs, L. M. G. (2009). Exogenous melatonin for sleep problems in individuals with intellectual disability: A meta-analysis. *Developmental Medicine and Child Neurology*, *51*, 340–349. https://doi.org/10.1111/j.1469-8749.2008.03244.x

Brown, K. A., & Piazza, C. C. (1999). Commentary: Enhancing the effectiveness of sleep treatments: Developing a functional approach. *Journal of Pediatric Psychology*, *24*(6), 487-489. https://doi.org/10.1093/jpepsy/24.6.487

Buysse, D. J., Reynolds III, C. F., Monk, T. H., Berman, S. R., & Kupfer, D. J. (1989). The Pittsburgh Sleep Quality Index: A new instrument for psychiatric practice and research. *Psychiatry research, 28*(2), 193-213. https://doi.org/10.1016/0165-1781(89)90047-4

Carnett, A., McLay, L., Hansen, S., France, K., & Blampied, N. (2021). Sleep problems in children and adolescents with autism: Type, severity and impact. *Journal of Developmental and Physical Disabilities*, *33*(6), 1-15. https://doi.org/10.1007/s10882-020-09783-5

Christensen, D. L., Braun, K. V. N., Baio, J., Bilder, D., Charles, J., Constantino, J. N., Daniels, J., Durkin, M. S., Fitzgerald, R, T., Kurzius-Spencer, M., Lee, L., Pettygrove, S., Robinson, C., Schulz, E., Wells, C., Wingate, M. S., Zahorodny, W., & Yeargin-Allsopp, M. (2018). Prevalence and characteristics of autism spectrum disorder among children aged 8 years—autism and developmental disabilities monitoring network, 11 sites, United States, 2012. *MMWR Surveillance Summaries*, *65*(13), 1. https://doi.org/10.15585/mmwr.ss6513a1

Christodulu, K. V., & Durand, V. M. (2004). Reducing bedtime disturbance and night waking using positive bedtime routines and sleep restriction. *Focus on Autism and Other Developmental Disabilities*, *19*(3), 130-139. https://doi.org/10.1177/10883576040190030101

Clementi, M. A., & Alfano, C. A. (2020). An integrated sleep and anxiety intervention for anxious children: A pilot randomized controlled trial. *Clinical Child Psychology and Psychiatry, 25*(4), 945-957. https://doi.org/10.1177/1359104520933936

Cohen, S., Conduit, R., Lockley, S. W., Rajaratnam, S. M., & Cornish, K. M. (2014). The relationship between sleep and behavior in autism spectrum disorder (ASD): A review. *Journal of Neurodevelopmental Disorders*, *6*(44), 1-10. https://doi.org/10.1186/1866-1955-6-44

Cohen, L. L., Feinstein, A., Masuda, A., & Vowles, K. E. (2014). Single-case research design in pediatric psychology: Considerations regarding data analysis. *Journal of Pediatric Psychology, 39*(2), 124-137. https://doi.org/10.1093/jpepsy/jst065

Cooney, M. R., Short, M. A., & Gradisar, M. (2018). An open trial of bedtime fading for sleep disturbances in preschool children: A parent group education approach. *Sleep Medicine*, *46*, 98-106. https://doi.org/10.1016/j.sleep.2018.03.003

Cooper, J. O., Heron, T. E., & Heward, W. L. (2020). *Applied behavior analysis* (3rd ed.). Pearson.

Cortesi, F., Giannotti, F., Ivanenko, A., & Johnson, K. (2010). Sleep in children with autistic spectrum disorder. *Sleep Medicine*, *11*(7), 659-664. https://doi.org/10.1016/j.sleep.2010.01.010

Cotton, S., & Richdale, A. (2006). Brief report: Parental descriptions of sleep problems in children with autism, Down syndrome, and Prader–Willi syndrome. *Research in Developmental Disabilities*, *27*(2), 151-161. https://doi.org/10.1016/j.ridd.2004.12.003

Croen, L. A., Najjar, D. V., Fireman, B., & Grether, J. K. (2007). Maternal and paternal age and risk of autism spectrum disorders. *Archives of Pediatrics and Adolescent Medicine*, *161*(4), 334-340. https://doi.org/10.1001/archpedi.161.4.334

Da Paz, N. S., & Wallander, J. L. (2017). Interventions that target improvements in mental health for parents of children with autism spectrum disorders: A narrative review. *Clinical Psychology Review, 51*, 1-14. https://doi.org/10.1016/j.cpr.2016.10.006

Davis, N. O., & Carter, A. S. (2008). Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism and Developmental Disorders*, *38*(7), 1278-1291. https://doi.org/10.1007/s10803-007-0512-z

DeFilippis, M. (2018). Depression in children and adolescents with autism spectrum disorder. *Children*, *5*(9), 112. https://doi.org/10.3390/children5090112

Delahaye, J., Kovacs, E., Sikora, D., Hall, T. A., Orlich, F., Clemons, T. E., van der Weerd, E., Glick, L., & Kuhlthau, K. (2014). The relationship between health-related quality of life and sleep problems in children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, *8*(3), 292-303. https://doi.org/10.1016/j.rasd.2013.12.015

Delemere, E., & Dounavi, K. (2018). Parent-implemented bedtime fading and positive routines for children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *48*(4), 1002-1019. https://doi.org/10.1007/s10803-017-3398-4

DeLeon, I. G., Fisher, W. W., & Marhefka, J.-M. (2004). Decreasing self-injurious behavior associated with awakening in a child with autism and developmental delays. *Behavioral Interventions*, *19*(2), 111-119. https://doi.org/10.1002/bin.154

Deliens, G., Leproult, R., Schmitz, R., Destrebecqz, A., & Peigneux, P. (2015). Sleep disturbances in autism spectrum disorders. *Review Journal of Autism and Developmental Disorders*, *2*, 343-356. https://doi.org/10.1007/s40489-015-0057-6

Deserno, M. K., Borsboom, D., Begeer, S., Agelink Van Rentergem, J. A., Mataw, K., & Geurts, H. M. (2019). Sleep determines quality of life in autistic adults: A longitudinal study. *Autism Research, 12*(5), 794-801. https://doi.org/10.1002/aur.2103

DeStefano, F., & Shimabukuro, T. T. (2019). The MMR vaccine and autism. *Annual Review of Virology*, *6*, 585-600. https://doi.org/10.1146/annurev-virology-092818-015515

Didden, R., Curfs, L. M., van Driel, S., & de Moor, J. M. (2002). Sleep problems in children and young adults with developmental disabilities: Home-based functional assessment and treatment. *Journal of Behavior Therapy and Experimental Psychiatry*, *33*(1), 49-58. https://doi.org/10.1016/S0005-7916(02)00012-5

Didden, R., & Sigafoos, J. (2001). A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. *Research in Developmental Disabilities*, *22*(4), 255-272. https://doi.org/10.1016/s0891-4222(01)00071-3

Durand, V. M. (2002). Treating sleep terrors in children with autism. *Journal of Positive Behavior Interventions*, *4*(2), 66-72. https://doi.org/10.1177/109830070200400201

Durand, V. M., & Christodulu, K. V. (2004). Description of a sleep-restriction program to reduce bedtime disturbances and night waking. *Journal of Positive Behavior Interventions*, *6*(2), 83-91. https://doi.org/10.1177/10983007040060020301

Durand, V. M., Gernert-Dott, P., & Mapstone, E. (1996). Treatment of sleep disorders in children with developmental disabilities. *Journal of the Association for Persons with Severe Handicaps*, *21*(3), 114-122. https://doi.org/10.1177/154079699602100302

Dziuk, M. A., Larson, J. G., Apostu, A., Mahone, E. M., Denckla, M. B., & Mostofsky, S. H. (2007). Dyspraxia in autism: Association with motor, social, and communicative deficits. *Developmental Medicine and Child Neurology*, *49*(10), 734-739. https://doi.org/10.1111/j.1469-8749.2007.00734.x

El-Sheikh, M., & Kelly, R. J. (2017). Family functioning and children's sleep. *Child Development Perspectives*, *11*(4), 264-269. https://doi.org/10.1111/cdep.12243

Essner, B., Noel, M., Myrvik, M., & Palermo, T. (2015). Examination of the factor structure of the Adolescent Sleep–Wake Scale (ASWS). *Behavioral Sleep Medicine*, *13*(4), 296-307. https://doi.org/10.1080/15402002.2014.896253

Ford, K. A., McLay, L. K., France, K. G., Blampied, N. M., & Gibbs, R. M. (2021). Systematic review of the effect of modification of antecedents in the treatment of sleep problems among children on the autism spectrum. *Advances in Neurodevelopmental Disorders,* 1-17. https://doi.org/10.1007/s41252-021-00224-8

Forquer, L. M., & Johnson, C. M. (2005). Continuous white noise to reduce resistance going to sleep and night wakings in toddlers. *Child and Family Behavior Therapy*, *27*(2), 1-10. https://doi.org/10.1300/J019v27n02_01

France, K., & Blampied, N. (1999). Infant sleep disturbance: Description of a problem behaviour process. *Sleep Medicine Reviews*, *3*(4), 265-280. https://doi.org/10.1053/smrv.1999.0071

France, K. G., & Hudson, S. M. (1990). Behavior management of infant sleep disturbance. *Journal of Applied Behavior Analysis*, *23*(1), 91-98. https://doi.org/10.1901/jaba.1990.23-91

Gabriele, S., Sacco, R., & Persico, A. M. (2014). Blood serotonin levels in autism spectrum disorder: A systematic review and meta-analysis. *European Neuropsychopharmacology*, *24*(6), 919-929. https://doi.org/10.1016/j.euroneuro.2014.02.004

Galland, B. C., & Mitchell, E. A. (2010). Helping children sleep. *Archives of Disease in Childhood*, *95*(10), 850-853. http://dx.doi.org/10.1136/adc.2009.162974

Geoffray, M. M., Nicolas, A., Speranza, M., & Georgieff, N. (2016). Are circadian rhythms new pathways to understand Autism Spectrum Disorder?. *Journal of Physiology-Paris, 110*(4), 434-438. https://doi.org/10.1016/j.jphysparis.2017.06.002

Giannotti, F., Cortesi, F., Cerquiglini, A., & Bernabei, P. (2006). An open-label study of controlled-release melatonin in treatment of sleep disorders in children with autism. *Journal of Autism and Developmental Disorders*, *36*(6), 741-752. https://doi.org/10.1007/s10803-006-0116-z

Gilliam, J. E. (2013). *Gilliam autism rating scale-third edition.* Pro-Ed.

Glasson, E. J., Bower, C., Petterson, B., de Klerk, N., Chaney, G., & Hallmayer, J. F. (2004). Perinatal factors and the development of autism: A population study. *Archives of General Psychiatry*, *61*(6), 618-627. https://doi.org/10.1001/archpsyc.61.6.618

Glickman, G. (2010). Circadian rhythms and sleep in children with autism. *Neuroscience and Biobehavioral Review*, *34*(5), 755-768. https://doi.org/10.1016/j.neubiorev.2009.11.017

Goldman, S. E., Alder, M. L., Burgess, H. J., Corbett, B. A., Hundley, R., Wofford, D., Fawkes, D. B., Wang, M. L., & Malow, B. A. (2017). Characterizing sleep in adolescents and adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *47*(6), 1682-1695. https://doi.org/10.1007/s10803-017-3089-1

Goldman, S. E., McGrew, S., Johnson, K. P., Richdale, A. L., Clemons, T., & Malow, B. A. (2011). Sleep is associated with problem behaviors in children and adolescents with autism spectrum

disorders. *Research in Autism Spectrum Disorders*, *5*(3), 1223-1229. https://doi.org/10.1016/j.rasd.2011.01.010

Goldman, S. E., Richdale, A. L., Clemons, T., & Malow, B. A. (2012). Parental sleep concerns in autism spectrum disorders: Variations from childhood to adolescence. *Journal of Autism and Developmental Disorders*, *42*(4), 531-538. https://doi.org/10.1007/s10803-011-1270-5

Gradisar, M., Jackson, K., Spurrier, N. J., Gibson, J., Whitham, J., Williams, A. S., Dolby, R., & Kennaway, D. J. (2016). Behavioral interventions for infant sleep problems: A randomized controlled trial. *Pediatrics*, *137*(6) e20151486. https://doi.org/10.1542/peds.2015-1486

Gray, C. (2010). *The new social story book.* Future Horizons.

Gray, C. A., & Garand, J. D. (1993). Social stories: Improving response of students with autism with accurate social information. *Focus on Autistic Behavior, 8*(1), 1-10.

Greimel, E., Nehrkorn, B., Schulte-Rüther, M., Fink, G. R., Nickl-Jockschat, T., Herpertz-Dahlmann, B., Konrad, K., & Eickhoff, S. B. (2013). Changes in grey matter development in autism spectrum disorder. *Brain Structure and Function*, *218*(4), 929-942. https://doi.org/10.1007/s00429-012-0439-9

Gringras, P., Nir, T., Breddy, J., Frydman-Marom, A., & Findling, R. L. (2017). Efficacy and safety of pediatric prolonged-release melatonin for insomnia in children with autism spectrum disorder. *Journal of the American Academy of Child and Adolescent Psychiatry*, *56*(11), 948-957. https://doi.org/10.1016/j.jaac.2017.09.414

Hall, W. A., & Nethery, E. (2019). What does sleep hygiene have to offer children's sleep problems?. *Paediatric Respiratory Reviews*, *31*, 64-74. https://doi.org/10.1016/j.prrv.2018.10.005

Hanley, G. P. (2005). *Sleep assessment and treatment tool*. https://practicalfunctionalassessment.files.wordpress.com/2015/06/satt.pdf

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behavior: A review. *Journal of Applied Behavior Analysis, 36*(2), 147-185. https://doi.org/10.1901/jaba.2003.36-147

Hanley, G. P., Jin, C. S., Vanselow, N. R., & Hanratty, L. A. (2014). Producing meaningful improvements in problem behavior of children with autism via synthesized analyses and treatments. *Journal of Applied Behavior Analysis*, *47*(1), 16-36. https://doi.org/10.1002/jaba.106

Healy, O., Brett, D., & Leader, G. (2013). A comparison of experimental functional analysis and the Questions About Behavioral Function (QABF) in the assessment of challenging behavior of individuals with autism. *Research in Autism Spectrum Disorders*, *7*(1), 66-81. https://doi.org/10.1016/j.rasd.2012.05.006

Hendricks, M. C., Ward, C. M., Grodin, L. K., & Slifer, K. J. (2014). Multicomponent cognitive-behavioural intervention to improve sleep in adolescents: A multiple baseline design. *Behavioural and Cognitive Psychotherapy*, *42*(3), 368-373. https://doi.org/10.1017/S1352465813000623

Heussler, H., Chan, P., Price, A. M., Waters, K., Davey, M. J., & Hiscock, H. (2013). Pharmacological and non-pharmacological management of sleep disturbance in children: An Australian Paediatric Research Network survey. Sleep Medicine, 14(2), 189-194. http://dx.doi.org/10.1016/j.sleep.2012.09.023

Hirshkowitz, M., Whiton, K., Albert, S. M., Alessi, C., Bruni, O., DonCarlos, L., Hazen, N., Herman, J., Katz, E., Kheirandish-Gozal, L., Neubauer, D., O'Donnell, A., Ohayon, M., Peever, J., Rawding, R., Sachdeva, R., Setters, B., Vitiello, M., ... & Hillard, P. J. A. (2015). National Sleep Foundation's sleep time duration recommendations: Methodology and results summary. *Sleep Health*, *1*(1), 40-43. https://doi.org/10.1016/j.sleh.2014.12.010

Hiscock, H., Sciberras, E., Mensah, F., Gerner, B., Efron, D., Khano, S., & Oberklaid, F. (2015). Impact of a behavioural sleep intervention on symptoms and sleep in children with attention deficit hyperactivity disorder, and parental mental health: Randomised controlled trial. *BMJ*, *350*. https://doi.org/10.1136/bmj.h68

Hiscock, H., & Wake, M. (2002). Randomised controlled trial of behavioural infant sleep intervention to improve infant sleep and maternal mood. *BMJ, 324*(7345), 1062. https://doi.org/10.1136/bmj.324.7345.1062

Hodge, D., Carollo, T. M., Lewin, M., Hoffman, C. D., & Sweeney, D. P. (2014). Sleep patterns in children with and without autism spectrum disorders: Developmental comparisons. *Research in Developmental Disabilities*, *35*(7), 1631-1638. https://doi.org/10.1016/j.ridd.2014.03.037

Hodge, D., Hoffman, C. D., Sweeney, D. P., & Riggs, M. L. (2013). Relationship between children's sleep and mental health in mothers of children with and without autism. *Journal of Autism and Developmental Disorders*, *43*(4), 956-963. https://doi.org/10.1007/s10803-012-1639-0

Hoffman, C. D., Sweeney, D. P., Gilliam, J. E., & Lopez-Wagner, M. C. (2006). Sleep problems in children with autism and in typically developing children. *Focus on Autism and Other Developmental Disabilities*, *21*(3), 146-152. https://doi.org/10.1177/10883576060210030301

Hoffman, C. D., Sweeney, D. P., Lopez-Wagner, M. C., Hodge, D., Nam, C. Y., & Botts, B. H. (2008). Children with autism: Sleep problems and mothers' stress. *Focus on Autism and Other Developmental Disabilities*, *23*(3), 155-165. https://doi.org/10.1177/1088357608316271

Hollway, J. A., & Aman, M. G. (2011). Sleep correlates of pervasive developmental disorders: A review of the literature. *Research in Developmental Disabilities*, *32*(5), 1399-1421. https://doi.org/10.1016/j.ridd.2011.04.001

Hollway, J. A., Aman, M. G., & Butter, E. (2013). Correlates and risk markers for sleep disturbance in participants of the autism treatment network. *Journal of Autism and Developmental Disorders*, *43*(12), 2830-2843. https://doi.org/10.1007/s10803-013-1830-y

Hrdlicka, M., Komarek, V., Propper, L., Kulisek, R., Zumrova, A., Faladova, L., Havlovicova, M., Sedlacek, Z., Blatny, M., & Urbanek, T. (2004). Not EEG abnormalities but epilepsy is associated with autistic regression and mental functioning in childhood autism. *European Child and Adolescent Psychiatry*, *13*(4), 209-213. https://doi.org/10.1007/s00787-004-0353-7

Jang, J., Dixon, D. R., Tarbox, J., & Granpeesheh, D. (2011). Symptom severity and challenging behavior in children on the autism spectrum. *Research in Autism Spectrum Disorders*, *5*(3), 1028-1032. https://doi.org/10.1016/j.rasd.2010.11.008

Jin, C. S., Hanley, G. P., & Beaulieu, L. (2013). An individualized and comprehensive approach to treating sleep problems in young children. *Journal of Applied Behavior Analysis*, *46*(1), 161-180. https://doi.org/10.1002/jaba.16

Johnson, K. P., & Malow, B. A. (2008). Sleep in children with autism spectrum disorders. *Current Treatment Options in Neurology*, *10*(5), 350-359. https://doi.org/10.1007/s11940-008-0038-5

Johnston, J. M., & Sherman, R. A. (1993). Applying the least restrictive alternative principle to treatment decisions: A legal and behavioral analysis. *The Behavior Analyst*, *16*(1), 103-115. https://doi.org/10.1007/BF03392615

Kaat, A. J., & Lecavalier, L. (2015). Reliability and validity of parent-and child-rated anxiety measures in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(10), 3219-3231. https://doi.org/10.1007/s10803-015-2481-y

Kahn, M., Juda-Hanael, M., Livne-Karp, E., Tikotzky, L., Anders, T. F., & Sadeh, A. (2020). Behavioral interventions for pediatric insomnia: One treatment may not fit all. *Sleep*, *43*(4), zsz268. https://doi.org/10.1093/sleep/zsz268

Keil, A., Daniels, J. L., Forssen, U., Hultman, C., Cnattingius, S., Söderberg, K. C., Feychting, M., & Sparen, P. (2010). Parental autoimmune diseases associated with autism spectrum disorders in offspring. *Epidemiology, 21*(6), 805-808. https://doi.org/10.1097/EDE.0b013e3181f26e3f

Keil, K. P., & Lein, P. J. (2016). DNA methylation: A mechanism linking environmental chemical exposures to risk of autism spectrum disorders?. *Environmental epigenetics*, *2*(1), dvv012. https://doi.org/10.1093/eep/dvv012

Kodak, T., & Piazza, C. C. (2011). Bedtime fading with response cost for children with multiple sleep problems. In M. L. Perlis, M. Aloia & B. Kuhn (Eds.), *Behavioral Treatments for Sleep Disorders* (pp. 285-292). Academic Press. https://doi.org/10.1016/B978-0-12-381522-4.00029-8

Kokina, A., & Kern, L. (2010). Social story interventions for students with autism spectrum disorders: A meta-analysis. *Journal of autism and developmental disorders*, *40*(7), 812-826. https://doi.org/10.1007/s10803-009-0931-0

Kotagal, S., & Broomall, E. (2012). Sleep in children with autism spectrum disorder. *Pediatric Neurology, 47*(4), 242-251. https://doi.org/10.1016/j.pediatrneurol.2012.05.007

Kratochwill, T. R., Hitchcock, J., Horner, R. H., Levin, J. R., Odom, S. L., Rindskopf, D. M., & Shadish, W. R. (2010). *Single-case designs technical documentation.* What Works Clearinghouse. https://files.eric.ed.gov/fulltext/ED510743.pdf

La Malfa, G., Lassi, S., Bertelli, M., Salvini, R., & Placidi, G. F. (2004). Autism and intellectual disability: A study of prevalence on a sample of the Italian population. *Journal of Intellectual Disability Research*, *48*(3), 262-267. https://doi.org/10.1111/j.1365-2788.2003.00567.x

Lambert, A., Tessier, S., Rochette, A. C., Scherzer, P., Mottron, L., & Godbout, R. (2016). Poor sleep affects daytime functioning in typically developing and autistic children not complaining of sleep problems: A questionnaire-based and polysomnographic study. *Research in Autism Spectrum Disorders*, *23*, 94-106. https://doi.org/10.1016/j.rasd.2015.11.010

Lawton, C., France, K. G., & Blampied, N. M. (1991). Treatment of infant sleep disturbance by graduated extinction. *Child and Family Behavior Therapy, 13*(1), 39-56. https://doi.org/10.1300/J019v13n01_03

LeBourgeois, M. K., Giannotti, F., Cortesi, F., Wolfson, A. R., & Harsh, J. (2005). The relationship between reported sleep quality and sleep hygiene in Italian and American adolescents. Pediatrics, 115(Supplement 1), 257-265. https://doi.org/10.1542/peds.2004-0815H

Lee, B. K., Magnusson, C., Gardner, R. M., Blomström, Å., Newschaffer, C. J., Burstyn, I., Karlsson, H., & Dalman, C. (2015). Maternal hospitalization with infection during pregnancy and risk of autism spectrum disorders. *Brain, Behavior, and Immunity, 44*, 100-105. https://doi.org/10.1016/j.bbi.2014.09.001

Lerman, D. C., & Iwata, B. A. (1995). Prevalence of the extinction burst and its attenuation during treatment. *Journal of Applied Behavior Analysis*, *28*(1), 93-94. https://doi.org/10.1901/jaba.1995.28-93

Lewandowski, A. S., Toliver-Sokol, M., & Palermo, T. M. (2011). Evidence-based review of subjective pediatric sleep measures. *Journal of Pediatric Psychology*, *36*(7), 780-793. https://doi.org/10.1093/jpepsy/jsq119

Lewis, M. L., Kesler, M., Candy, S. A., Rho, J. M., & Pittman, Q. J. (2018). Comorbid epilepsy in autism spectrum disorder: Implications of postnatal inflammation for brain excitability. *Epilepsia*, *59*(7), 1316-1326. https://doi.org/10.1111/epi.14440

Leyfer, O. T., Folstein, S. E., Bacalman, S., Davis, N. O., Dinh, E., Morgan, J., Tager-Flusberg, H., & Lainhart, J. E. (2006). Comorbid psychiatric disorders in children with autism: Interview development and rates of disorders. *Journal of Autism and Developmental Disorders*, *36*(7), 849-861. https://doi.org/10.1007/s10803-006-0123-0

Libbey, J. E., Sweeten, T. L., McMahon, W. M., & Fujinami, R. S. (2005). Autistic disorder and viral infections. *Journal of NeuroVirology, 11*(1), 1-10. https://doi.org/10.1080/13550280590900553

Liu, R., Dong, H., Wang, Y., Lu, X., Li, Y., Xun, G., Ou, J., Shen, Y., & Zhao, J. (2021). Sleep problems of children with autism may independently affect parental quality of life. *Child Psychiatry and Human Development, 52*(3), 488-499. https://doi.org/10.1007/s10578-020-01035-z

Liu, X., Hubbard, J. A., Fabes, R. A., & Adam, J. B. (2006). Sleep disturbances and correlates of children with autism spectrum disorders. *Child Psychiatry and Human Development*, *37*(2), 179-191. https://doi.org/10.1007/s10578-006-0028-3

Lodge R. E. (2001). Functional behavioral assessment and children with autism: Working as a team. *Focus on Autism and Other Developmental Disabilities*, *16*(4), 228-231. https://doi.org/10.1177/108835760101600405

Lora, C. C., Kisamore, A. N., Reeve, K. F., & Townsend, D. B. (2020). Effects of a problem-solving strategy on the independent completion of vocational tasks by adolescents with autism spectrum disorder. *Journal of Applied Behavior Analysis*, *53*(1), 175-187.

Loring, W. A., Johnston, R. L., Shui, A. M., & Malow, B. A. (2018). Impact of a brief behavioral intervention for insomnia on daytime behaviors in adolescents with autism spectrum disorders. *Journal of Contemporary Psychotherapy*, *48*(3), 165-177. https://doi.org/10.1007/s10879-018-9381-3

Lovibond, P. F., & Lovibond, S. H. (1995). The structure of negative emotional states: Comparison of the Depression Anxiety Stress Scales (DASS) with the Beck Depression and Anxiety Inventories. *Behaviour Research and Therapy, 33*(3), 335-343. https://doi.org/10.1016/0005-7967(94)00075-U

Luiselli, J. K., Harper, J. M., Murphy, K. J., Leach, M., & Luke, K. (2021). Case report of faded bedtime intervention on delayed sleep-onset and sleep duration in an adolescent with autism spectrum disorder. *Child and Family Behavior Therapy*, *43*(2), 114-122. https://doi.org/10.1080/07317107.2021.1894712

Luiselli, J. K., Harper, J. M., Shlesinger, A., Murphy, K. J., & Luke, K. (2020). Faded bedtime intervention for delayed sleep onset in an adolescent with autism spectrum disorder. *Clinical Case Studies*, *19*(3), 180-188. https://doi.org/10.1177/1534650119897964

Ma, H. (2006). An alternative method for quantitative synthesis of single-subject researches: Percentage of data points exceeding the median. *Behavior Modification, 30*(5), 598-617. https://doi.org/10.1177/0145445504272974

Malow, B., Adkins, K. W., McGrew, S. G., Wang, L., Goldman, S. E., Fawkes, D., & Burnette, C. (2012). Melatonin for sleep in children with autism: A controlled trial examining dose, tolerability, and outcomes. *Journal of Autism and Developmental Disorders, 42*(8), 1729-1737. https://doi.org/10.1007/s10803-011-1418-3

Malow, B. A., Adkins, K. W., Reynolds, A., Weiss, S. K., Loh, A., Fawkes, D., Katz, T., Goldman, S., Madduri, N., Hundley, R., & Clemons, T. (2014). Parent-based sleep education for children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *44*(1), 216-228. https://doi.org/10.1007/s10803-013-1866-z

Malow, B. A., Katz, T., Reynolds, A. M., Shui, A., Carno, M., Connolly, H. V., Coury, D., & Bennett, A. E. (2016). Sleep difficulties and medications in children with autism spectrum disorders: A registry study. *Pediatrics*, *137*(Supplement 2), S98-S104. https://doi.org/10.1542/peds.2015-2851H

Malow, B. A., Marzec, M. L., McGrew, S. G., Wang, L., Henderson, L. M., & Stone, W. L. (2006). Characterizing sleep in children with autism spectrum disorders: A multidimensional approach. *Sleep, 29*(12), 1563-1571. https://doi.org/10.1093/sleep/29.12.1563

Mannion, A., Leader, G., & Healy, O. (2013). An investigation of comorbid psychological disorders, sleep problems, gastrointestinal symptoms and epilepsy in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *7*(1), 35-42. https://doi.org/10.1016/j.rasd.2012.05.002

Maras, A., Schroder, C. M., Malow, B. A., Findling, R. L., Breddy, J., Nir, T., Shahmoon, S., Zisapel, N., & Gringras, P. (2018). Long-term efficacy and safety of pediatric prolonged-release melatonin for insomnia in children with autism spectrum disorder. *Journal of Child and Adolescent Psychopharmacology*, *28*(10), 699-710. https://doi.org/10.1089/cap.2018.0020

March, J.S. (2012). *The Multidimensional Anxiety Scale for Children – Second Edition (MASC-2)*. Multi-Health Systems.

Matson, J. L., & Vollmer, T. R. (1995). *User's guide: Questions about behavioral function (QABF)*. Scientific Publishers.

Matson, J. L., Wilkins, J., & Macken, J. (2008). The relationship of challenging behaviors to severity and symptoms of autism spectrum disorders. *Journal of Mental Health Research in Intellectual Disabilities*, *2*(1), 29-44. https://doi.org/10.1080/19315860802611415

Mazurek, M. O., & Sohl, K. (2016). Sleep and behavioral problems in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *46*(6), 1906-1915. https://doi.org/10.1007/s10803-016-2723-7

Mazzone, L., Postorino, V., Siracusano, M., Riccioni, A., & Curatolo, P. (2018). The relationship between sleep problems, neurobiological alterations, core symptoms of autism spectrum disorder, and psychiatric comorbidities. *Journal of Clinical Medicine, 7*(5), 102. https://doi.org/10.3390/jcm7050102

Mbadiwe, T., & Millis, R. M. (2013). Epigenetics and autism. *Autism Research and Treatment*, *2013*, 1-9. https://doi.org/10.1155/2013/826156

McAlonan, G. M., Suckling, J., Wong, N., Cheung, V., Lienenkaemper, N., Cheung, C., & Chua, S. E. (2008). Distinct patterns of grey matter abnormality in high-functioning autism and Asperger's syndrome. *Journal of Child Psychology and Psychiatry*, *49*(12), 1287-1295. https://doi.org/10.1111/j.1469-7610.2008.01933.x

McCrae, C. S., Chan, W. S., Curtis, A. F., Deroche, C. B., Munoz, M., Takamatsu, S., Muckerman, J. E., Takahashi, N., McCann, D., McGovney, K., Sahota, P., & Mazurek, M. O. (2020). Cognitive behavioral treatment of insomnia in school-aged

children with autism spectrum disorder: A pilot feasibility study. *Autism Research*, *13*(1), 167-176. https://doi.org/10.1002/aur.2204

McLay, L., France, K., Blampied, N., & Hunter, J. (2017). Using functional behavioral assessment to treat sleep problems in two children with autism and vocal stereotypy. *International Journal of Developmental Disabilities*, *65*(3), 175-184. https://doi.org/10.1080/20473869.2017.1376411

McLay, L. L., France, K. G., Blampied, N. M., Hunter, J. E., van Deurs, J. R., Woodford, E. C., Gibbs, R., & Lang, R. (2021). Collateral child and parent effects of function-based behavioral interventions for sleep problems in children and adolescents with autism. Journal of Autism and Developmental Disorders, 1-16. https://doi.org/10.1007/s10803-021-05116-3

McLay, L., France, K., Blampied, N., van Deurs, J., Hunter, J., Knight, J., Hastie, B., Carnett, A., Woodford, E., Gibbs, R., & Lang, R. (2021). Function-based behavioral interventions for sleep problems in children and adolescents with autism: Summary of 41 clinical cases. *Journal of Autism and Developmental Disorders, 51*(2), 418-432. https://doi.org/10.1007/s10803-020-04548-7

McLay, L. K., France, K. G., Knight, J., Blampied, N. M., & Hastie, B. (2019). The effectiveness of function-based interventions to treat sleep problems, including unwanted co-sleeping, in children with autism. *Behavioral Interventions*, *34*(1), 30-51. https://doi.org/10.1002/bin.1651

McLay, L. K., Schluter, P. J., Eggleston, M. J. F., Woodford, E. C., & Bowden, N. (2021). Melatonin dispensing among New Zealand children aged 0–18 years with autism: A nationwide cross-sectional study. *Sleep Medicine*, *80*, 184-192. https://doi.org/10.1016/j.sleep.2021.01.028

Melke, J., Goubran Botros, H., Chaste, P., Betancur, C., Nygren, G., Anckarsater, H., Rastam, M., Stahlberg, O., Gillberg, I. C., Delorme, R., Chabane, N., Mouren-

Simeoni, M-C., Fauchereau, F., Durand, C. M., Chevalier, F., Drouot, X., Collet, C., Launay, J-M., Leboyer, M., . . . Bourgeron, T. (2008). Abnormal melatonin synthesis in autism spectrum disorders. *Molecular Psychiatry*, *13*(1), 90-98. https://doi.org/10.1038/sj.mp.4002016

Meltzer, L. J. (2008). Brief report: Sleep in parents of children with autism spectrum disorders. *Journal of Pediatric Psychology, 33*(4), 380-386. https://doi.org/10.1093/jpepsy/jsn005

Meltzer, L. J., & Mindell, J. A. (2007). Relationship between child sleep disturbances and maternal sleep, mood, and parenting stress: A pilot study. *Journal of Family Psychology*, *21*(1), 67-73. https://doi.org/10.1037/0893-3200.21.1.67

Meltzer, L. J., & Mindell, J. A. (2014). Systematic review and meta-analysis of behavioral interventions for pediatric insomnia. Journal of Pediatric Psychology, 39(8), 932-948. https://doi.org/10.1093/jpepsy/jsu041

Michael, J. (1982). Distinguishing between discriminative and motivational functions of stimuli. *Journal of the Experimental Analysis of Behavior*, *37*(1), 149-155. https://doi.org/10.1901/jeab.1982.37-149

Mindell, J. A., Telofski, L. S., Wiegand, B., & Kurtz, E. S. (2009). A nightly bedtime routine: Impact on sleep in young children and maternal mood. *Sleep*, *32*(5), 599-606. https://doi.org/10.1093/sleep/32.5.599

Ming, X., Sun, Y. M., Nachajon, R. V., Brimacombe, M., & Walters, A. S. (2009). Prevalence of parasomnia in autistic children with sleep disorders. *Clinical Medicine: Pediatrics*, *2009*(3), 1-10. https://doi.org/10.4137/CMPed.S1139

Ministry of Health. (2019). *Annual data explorer 2018/19: New Zealand Health Survey*. https://minhealthnz.shinyapps.io/nz-health-survey-2018-19-annual-data-explorer/_w_fc21d8ce/#!/explore-topics

Mirenda, P., Smith, I. M., Vaillancourt, T., Georgiades, S., Duku, E., Szatmari, P., Bryson, S., Fombonne, E., Roberts, W., Volden, J., Waddell, C., & Zwaigenbaum, L. (2010). Validating the repetitive behavior scale-revised in young children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(12), 1521-1530. https://doi.org/10.1007/s10803-010-1012-0

Moon, E. C., Corkum, P., & Smith, I. M. (2011). Case study: A case-series evaluation of a behavioral sleep intervention for three children with autism and primary insomnia. *Journal of Pediatric Psychology, 36*(1), 47-54. https://doi.org/10.1093/jpepsy/jsq057

Morgenthaler, T. I., Owens, J., Alessi, C., Boehlecke, B., Brown, T. M., Coleman, J., Friedman, L., Kapur, V. K., Lee-Chiong, T., Pancer, J., & Swick, T. J. (2006). Practice parameters for behavioural treatment of bedtime problems and night wakings in infants and young children. *Sleep, 29*(10), 1277-1281. https://doi.org/10.1093/sleep/29.10.1277

Muhle, R., Trentacoste, S. V., & Rapin, I. (2004). The genetics of autism. *Pediatrics*, *113*(5), e472-e486. https://doi.org/10.1542/peds.113.5.e472

Nicholas, B., Rudrasingham, V., Nash, S., Kirov, G., Owen, M. J., & Wimpory, D. C. (2007). Association of Per1 and Npas2 with autistic disorder: Support for the clock genes/social timing hypothesis. *Molecular psychiatry*, *12*(6), 581-592. https://doi.org/10.1038/sj.mp.4001953

Norton, P. J. (2007). Depression Anxiety and Stress Scales (DASS-21): Psychometric analysis across four racial groups. *Anxiety, Stress, and Coping, 20*(3), 253-265. https://doi.org/10.1080/10615800701309279

Norton, R. (1983). Measuring marital quality: A critical look at the dependent variable. *Journal of Marriage and the Family 45*(1), 141-151. https://doi.org/10.2307/351302

Ohayon, M., Wickwire, E. M., Hirshkowitz, M., Albert, S. M., Avidan, A., Daly, F. J., Dauvilliers, Y., Ferri, R., Fung, C., Gozal, D., Hazen, N., Krystal, A., Lichstein, K., Mallampalli, M., Plazzi, G., Rawding, R., Scheer, F. A., Somers, V., & Vitiello, M. V. (2017). National Sleep Foundation's sleep quality recommendations: First report. *Sleep health, 3*(1), 6-19. https://doi.org/10.1016/j.sleh.2016.11.006

Okawa, M., Uchiyama, M., Ozaki, S., Shibui, K., & Ichikawa, H. (1998). Circadian rhythm sleep disorders in adolescents: Clinical trials of combined treatments based on chronobiology. *Psychiatry and Clinical Neurosciences*, *52*(5), 483-490. https://doi.org/10.1046/j.1440-1819.1998.00449.x

O'Reilly, M., Rispoli, M., Davis, T., Machalicek, W., Lang, R., Sigafoos, J., Kang, S., Lancioni, G., Green, V., & Didden, R. (2010). Functional analysis of challenging behavior in children with autism spectrum disorders: A summary of 10 cases. Research in Autism Spectrum Disorders, 4(1), 1-10. https://doi.org/10.1016/j.rasd.2009.07.001

Owens, L. J., France, K. G., & Wiggs, L. (1999). Behavioural and cognitive-behavioural interventions for sleep disorders in infants and children: A review. *Sleep Medicine Reviews*, *3*(4), 281-302. https://doi.org/10.1053/smrv.1999.0082

Owens, J. A., Spirito, A., & McGuinn, M. (2000). The Children's Sleep Habits Questionnaire (CSHQ): Psychometric properties of a survey instrument for school-aged children. *Sleep, 23*(8), 1043-1052.

Owens, J. A., Spirito, A., McGuinn, M., & Nobile, C. (2000). Sleep habits and sleep disturbance in elementary school-aged children. *Journal of Developmental and Behavioral Pediatrics*, *21*(1), 27-36. https://doi.org/10.1097/00004703-200002000-00005

Paavonen, E. J., Vehkalahti, K., Vanhala, R., von Wendt, L., Nieminen-von Wendt, T., & Aronen, E. T. (2008). Sleep in children with Asperger syndrome. *Journal of Autism and Developmental Disorders*, *38*(1), 41-51. https://doi.org/10.1007/s10803-007-0360-x

Paclawskyj, T. R., Matson, J. L., Rush, K. S., Smalls, Y., & Vollmer, T. R. (2000). Questions about behavioral function (QABF): A behavioral checklist for functional assessment of aberrant behavior. *Research in Developmental Disabilities*, *21*(3), 223-229. https://doi.org/10.1016/S0891-4222(00)00036-6

Palermo, T. M., Fonareva, I., & Janosy, N. R. (2008). Sleep quality and efficiency in adolescents with chronic pain: Relationship with activity limitations and health-related quality of life. *Behavioral Sleep Medicine*, *6*(4), 234-250. https://doi.org/10.1080/15402000802371353

Pandolfi, V., Magyar, C. I., & Dill, C. A. (2012). An initial psychometric evaluation of the CBCL 6–18 in a sample of youth with autism spectrum disorders. *Research in Autism Spectrum Disorders, 6*(1), 96-108. https://doi.org/10.1016/j.rasd.2011.03.009

Papadopoulos, N., Sciberras, E., Hiscock, H., Mulraney, M., McGillivray, J., & Rinehart, N. (2019). The efficacy of a brief behavioral sleep intervention in school-aged children with ADHD and comorbid autism spectrum disorder. *Journal of Attention Disorders*, *23*(4), 341-350. https://doi.org/10.1177/1087054714568565

Park, S., Cho, S. C., Cho, I. H., Kim, B. N., Kim, J. W., Shin, M. S., Chung, U., Park, T., Son, J., & Yoo, H. J. (2012). Sleep problems and their correlates and comorbid psychopathology of children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, *6*(3), 1068-1072. https://doi.org/10.1016/j.rasd.2012.02.004

Pattison, E., Papadopoulos, N., Marks, D., McGillivray, J., & Rinehart, N. (2020). Behavioural treatments for sleep problems in children with Autism Spectrum

Disorder: A review of the recent literature. *Current Psychiatry Reports*, *22*(9), 1-10. https://doi.org/10.1007/s11920-020-01172-1

Pfaender, S., Sauer, A. K., Hagmeyer, S., Mangus, K., Linta, L., Liebau, S., Bockmann, J., Huguet, G., Bourgeron, T., Boeckers, T. M., & Grabrucker, A. M. (2017). Zinc deficiency and low enterocyte zinc transporter expression in human patients with autism related mutations in SHANK3. *Scientific Reports*, *7*, 1-15. https://doi.org/10.1038/srep45190

Piazza, C. C., & Fisher, W. W. (1991). Bedtime fading in the treatment of pediatric insomnia. *Journal of Behavior Therapy and Experimental Psychiatry*, *22*(1), 53-56. https://doi.org/10.1016/0005-7916(91)90034-3

Piazza, C. C., Fisher, W. W., & Sherer, M. (1997). Treatment of multiple sleep problems in children with developmental disabilities: Faded bedtime with response cost versus bedtime scheduling. *Developmental Medicine and Child Neurology*, *39*(6), 414-418. https://doi.org/10.1111/j.1469-8749.1997.tb07456.x

Piazza, C. C., Hagopian, L. P., Hughes, C. R., & Fisher, W. W. (1998). Using chronotherapy to treat severe sleep problems: A case study. *American Journal of Mental Retardation*, *102*(4), 358-366. https://doi.org/10.1352/0895-8017(1998)102<0358:ucttss>2.0.co;2

Price, A. M., Wake, M., Ukoumunne, O. C., & Hiscock, H. (2012). Five-year follow-up of harms and benefits of behavioral infant sleep intervention: Randomized trial. *Pediatrics*, *130*(4), 643-651. https://doi.org/10.1542/peds.2011-3467

Quach, J. L., Nguyen, C. D., Williams, K. E., & Sciberras, E. (2018). Bidirectional associations between child sleep problems and internalizing and externalizing difficulties from preschool to early adolescence. *JAMA pediatrics, 172*(2), e174363. https://doi.org/10.1001/jamapediatrics.2017.4363

Rafihi-Ferreira, R. E., Pires, M. L. N., & Silvares, E. F. D. M. (2019). Behavioral intervention for sleep problems in childhood: A Brazilian randomized controlled trial. *Psicologia: Reflexão e Crítica*, *32*(5), 1-13. https://doi.org/10.1186/s41155-019-0118-3

Reed, H. E., McGrew, S. G., Artibee, K., Surdkya, K., Goldman, S. E., Frank, K., Wang, L., & Malow, B. A. (2009). Parent-based sleep education workshops in autism. *Journal of Child Neurology*, *24*(8), 936-945. https://doi.org/10.1177/0883073808331348

Reimers, T. M., Wacker, D. P., Cooper, L. J., & De Raad, A. O. (1992). Acceptability of behavioral treatments for children: Analog and naturalistic evaluations by parents. *School Psychology Review, 21*(4), 628-643.

Reynolds, A. M., & Malow, B. A. (2011). Sleep and autism spectrum disorders. *Pediatric Clinics, 58*(3), 685-698. https://doi.org/10.1016/j.pcl.2011.03.009

Richdale, A. L. (2013). Autism and other developmental disabilities. In A. R. Wolfson & H. Montgomery-Downs. *The Oxford Handbook of Infant, Child, and Adolescent Sleep and Behavior* (pp. 471-494). Oxford, UK: Oxford University Press.

Richdale, A. L., & Baglin, C. L. (2015). Self-report and caregiver-report of sleep and psychopathology in children with high-functioning autism spectrum disorder: A pilot study. *Developmental Neurorehabilitation*, *18*(4), 272-279. https://doi.org/10.3109/17518423.2013.829534

Richdale, A. L., & Prior, M. R. (1995). The sleep/wake rhythm in children with autism. *European child & adolescent psychiatry*, *4*(3), 175-186. https://doi.org/10.1007/BF01980456

Richdale, A. L., & Schreck, K. A. (2009). Sleep problems in autism spectrum disorders: Prevalence, nature, & possible biopsychosocial aetiologies. *Sleep Medicine Reviews*, *13*(6), 403-411. https://doi.org/10.1016/j.smrv.2009.02.003

Richdale, A. L., & Schreck, K. A. (2019). Examining sleep hygiene factors and sleep in young children with and without autism spectrum disorder. *Research in Autism Spectrum Disorders*, *57*, 154-162. https://doi.org/10.1016/j.rasd.2018.10.008

Richdale, A., & Wiggs, L. (2005). Behavioral approaches to the treatment of sleep problems in children with developmental disorders: What is the state of the art?. *International Journal of Behavioral Consultation and Therapy, 1*(3), 165. http://dx.doi.org/10.1037/h0100743

Rickert, V. I., & Johnson, C. M. (1988). Reducing nocturnal awakening and crying episodes in infants and young children: A comparison between scheduled awakenings and systematic ignoring. *Pediatrics*, *81*(2), 203-212. https://doi.org/10.1542/peds.81.2.203

Rolf, L. H., Haarmann, F. Y., Grotemeyer, K. H., & Kehrer, H. (1993). Serotonin and amino acid content in platelets of autistic children. *Acta Psychiatrica Scandinavica*, *87*(5), 312-316. https://doi.org/10.1111/j.1600-0447.1993.tb03378.x

Rosen, T. E., Mazefsky, C. A., Vasa, R. A., & Lerner, M. D. (2018). Co-occurring psychiatric conditions in autism spectrum disorder. *International Review of Psychiatry*, *30*(1), 40-61. https://doi.org/10.1080/09540261.2018.1450229

Rossignol, D. A., & Frye, R. E. (2011). Melatonin in autism spectrum disorders: A systematic review and meta-analysis. *Developmental Medicine and Child Neurology*, *53*(9), 783-792. https://doi.org/10.1111/j.1469-8749.2011.03980.x

Rossignol, D. A., & Frye, R. E. (2014). Melatonin in autism spectrum disorders. *Current Clinical Pharmacology*, *9*(4), 326-334. https://doi.org/10.2174/1574884711308666007 2

Sanders, M. R., Kirby, J. N., Tellegen, C. L., & Day, J. J. (2014). The Triple P-Positive Parenting Program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clinical Psychology Review*, *34*(4), 337-357. https://doi.org/10.1016/j.cpr.2014.04.003.

Sandin, S., Hultman, C. M., Kolevzon, A., Gross, R., MacCabe, J. H., & Reichenberg, A. (2012). Advancing maternal age is associated with increasing risk for autism: A review and meta-analysis. *Journal of the American Academy of Child and Adolescent Psychiatry*, *51*(5), 477-486. https://doi.org/10.1016/j.jaac.2012.02.018

Schneider, N., & Goldstein, H. (2010). Using social stories and visual schedules to improve socially appropriate behaviors in children with autism. *Journal of Positive Behavior Interventions*, *12*(3), 149-160. https://doi.org/10.1177/1098300709334198

Schreck, K. A., & Mulick, J. A. (2000). Parental report of sleep problems in children with autism. *Journal of Autism and Developmental Disorders*, *30*(2), 127-135. https://doi.org/10.1023/A:1005407622050

Schreck, K. A., Mulick, J. A., & Smith, A. F. (2004). Sleep problems as possible predictors of intensified symptoms of autism. *Research in Developmental Disabilities*, *25*(1), 57-66. https://doi.org/10.1016/j.ridd.2003.04.007

Shetty, J., Newton, A. T., & Reid, G. J. (2022). Parenting practices, bedtime routines, and consistency: Associations with pediatric sleep problems. Journal of Pediatric Psychology, 47(1), 49-58. https://doi.org/10.1093/jpepsy/jsab072

Shipman, D. L., Sheldrick, R. C., & Perrin, E. C. (2011). Quality of life in adolescents with autism spectrum disorders: Reliability and validity of self-reports. *Journal of Developmental & Behavioral Pediatrics, 32*(2), 85-89. https://doi.org/10.1097/DBP.0b013e318203e558

Sikora, D. M., Johnson, K., Clemons, T., & Katz, T. (2012). The relationship between sleep problems and daytime behavior in children of different ages with autism spectrum disorders. *Pediatrics*, *130*(Supplement 2), S83-S90. https://doi.org/10.1542/peds.2012-0900F

Singh, K., & Zimmerman, A. W. (2015). Sleep in autism spectrum disorder and attention deficit hyperactivity disorder. *Seminars in Pediatric Neurology, 22*(2), 113-125. https://doi.org/10.1016/j.spen.2015.03.006

Souders, M. C., Mason, T. B., Valladares, O., Bucan, M., Levy, S. E., Mandell, D. S., Weaver, T. E., & Pinto-Martin, J. (2009). Sleep behaviors and sleep quality in children with autism spectrum disorders. *Sleep*, *32*(12), 1566-1578. https://doi.org/10.1093/sleep/32.12.1566

Sparrow, S. S., Cicchetti, D. V., and Saulnier, C. A. (2016). Vineland Adaptive Behavior Scales, Third Edition (Vineland-3). Pearson.

Spielman, A. J., Caruso, L. S., & Glovinsky, P. B. (1987). A behavioral perspective on insomnia treatment. *Psychiatric Clinics of North America*, *10*(4), 541-553. https://doi.org/10.1016/S0193-953X(18)30532-X

Staples, A. D., Bates, J. E., & Petersen, I. T. (2015). Ix. Bedtime routines in early childhood: Prevalence, consistency, and associations with nighttime sleep. *Monographs of the Society for Research in Child Development*, *80*(1), 141-159. https://doi.org/10.1111/mono.12149

Stein, M. A. (1999). Unravelling sleep problems in treated and untreated children with ADHD. *Journal of Child and Adolescent Psychopharmacology*, *9*(3), 157-168. https://doi.org/10.1089/cap.1999.9.157.

Stepanski, E. J., & Wyatt, J. K. (2003). Use of sleep hygiene in the treatment of insomnia. *Sleep Medicine Reviews, 7*(3), 215-225. https://doi.org/10.1053/smrv.2001.0246

Storfer-Isser, A., Lebourgeois, M. K., Harsh, J., Tompsett, C. J., & Redline, S. (2013). Psychometric properties of the Adolescent Sleep Hygiene Scale. *Journal of Sleep Research, 22*(6), 707-716. https://doi.org/10.1111/jsr.1205

Sung, V., Hiscock, H., Sciberras, E., & Efron, D. (2008). Sleep problems in children with attention-deficit/hyperactivity disorder: Prevalence and the effect on the child and family. *Archives of Pediatrics and Adolescent Medicine*, *162*(4), 336-342. https://doi.org/10.1001/archpedi.162.4.336

Sustere, E., & Tarpey, E. (2019). Least restrictive practice: Its role in patient independence and recovery. *The Journal of Forensic Psychiatry & Psychology*, *30*(4), 614-629. https://doi.org/10.1080/14789949.2019.1566489

Tilford, J. M., Payakachat, N., Kuhlthau, K. A., Pyne, J. M., Kovacs, E., Bellando, J., Williams, D. K., Brouwer, W. B. F., & Frye, R. E. (2015). Treatment for sleep problems in children with autism and caregiver spillover effects. *Journal of Autism and Developmental Disorders*, *45*(11), 3613-3623. https://doi.org/10.1007/s10803-015-2507-5

Uljarević, M., Hedley, D., Rose-Foley, K., Magiati, I., Cai, R. Y., Dissanayake, C., Richdale, A., & Trollor, J. (2019). Anxiety and depression from adolescence to old age in autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *50*(9), 3155-3165. https://doi.org/10.1007/s10803-019-04084-z

van Deurs, J. R., France, K. G., McLay, L. K., & Blampied, N. M. (2021). Cognitive-behavioral treatment of sleep disturbance in children and adolescents with autism: Eight case studies using functional behavior assessment. *Research in Autism Spectrum Disorders, 86*, 101823. https://doi.org/10.1016/j.rasd.2021.101823

van Deurs, J. R., McLay, L. K., France, K. G., & Blampied, N. M. (2021). Sequential implementation of functional behavior assessment-informed treatment components for sleep disturbance in autism: A case study. Behavioral Sleep Medicine, 19(3), 333-351. https://doi.org/10.1080/15402002.2020.1758701

van Deurs, J. R., McLay, L. K., France, K. G., Blampied, N. M., Lang, R. B., & Hunter, J. E. (2019). Behavioral sleep intervention for adolescents with autism spectrum disorder: A pilot study. *Advances in Neurodevelopmental Disorders*, *3*(4), 397-410. https://doi.org/10.1007/s41252-019-00123-z

Varni, J. W., Seid, M., & Rode, C. A. (1999). The PedsQL™: Measurement model for the pediatric quality of life inventory. *Medical Care 37*(2), 126-139.

Vriend, J. L., Corkum, P. V., Moon, E. C., & Smith, I. M. (2011). Behavioral interventions for sleep problems in children with autism spectrum disorders: Current findings and future directions. *Journal of Pediatric Psychology*, *36*(9), 1017-1029. https://doi.org/10.1093/jpepsy/jsr044

Wasserstein, R. L., & Lazar, N. A. (2016). The ASA statement on p-values: Context, process, and purpose. *The American Statistician, 70*(2), 129-133. https://doi.org/10.1080/00031305.2016.1154108

Weiskop, S., Matthews, J., & Richdale, A. (2001). Treatment of sleep problems in a 5-year-old boy with autism using behavioural principles. *Autism*, *5*(2), 209-221. https://doi.org/10.1177/1362361301005002009

Weiskop, S., Richdale, A., & Matthews, J. (2005). Behavioural treatment to reduce sleep problems in children with autism or fragile X syndrome. *Developmental Medicine and Child Neurology*, *47*(2), 94-104. https://doi.org/10.1017/S0012162205000186

Weitlauf, A. S., Vehorn, A. C., Taylor, J. L., & Warren, Z. E. (2014). Relationship satisfaction, parenting stress, and depression in mothers of children with autism. *Autism, 18*(2), 194-198. https://doi.org/10.1177/1362361312458039

Wiggs, L., & Stores, G. (2004). Sleep patterns and sleep disorders in children with autistic spectrum disorders: Insights using parent report and actigraphy. *Developmental Medicine and Child Neurology*, *46*(6), 372-380. https://doi.org/10.1017/S0012162204000611

Will, E. A., Schworer, E. K., & Esbensen, A. J. (2021). The role of distinct executive functions on adaptive behavior in children and adolescents with Down syndrome. *Child Neuropsychology, 27(*8), 1054-1072. https://doi.org/10.1080/09297049.2021.1917531

Williams, P. G., Sears, L. L., & Allard, A. (2004). Sleep problems in children with autism. *Journal of Sleep Research*, *13*(3), 265-268. https://doi.org/10.1111/j.1365-2869.2004.00405.x

Williamson, A. A., Mindell, J. A., Hiscock, H., & Quach, J. (2020). Longitudinal sleep problem trajectories are associated with multiple impairments in child well-being. *Journal of Child Psychology and Psychiatry, 61*(10), 1092-1103. https://doi.org/10.1111/jcpp.13303

Wirojanan, J., Jacquemont, S., Diaz, R., Bacalman, S., Anders, T. F., Hagerman, R. J., & Goodlin-Jones, B. L. (2009). The efficacy of melatonin for sleep problems in children with autism, fragile X syndrome, or autism and fragile X syndrome. *Journal of Clinical Sleep Medicine*, *5*(2), 145-150. https://doi.org/10.5664/jcsm.27443

Wright, B., Sims, D., Smart, S., Alwazeer, A., Alderson-Day, B., Allgar, V., Whitton, C., Tomlinson, H., Bennett, S., Jardine, J., McCaffrey, N., Leyland, C., Jakeman, C., & Miles, J. (2011). Melatonin versus placebo in children with autism spectrum conditions and severe sleep problems not amenable to behaviour management strategies: A randomised controlled crossover trial. *Journal of Autism and Developmental Disorders*, *41*(2), 175-184. https://doi.org/10.1007/s10803-010-1036-5

Wu, S., Wu, F., Ding, Y., Hou, J., Bi, J., & Zhang, Z. (2017). Advanced parental age and autism risk in children: a systematic review and meta-analysis. *Acta Psychiatrica Scandinavica*, *135*(1), 29-41. https://doi.org/10.1111/acps.12666

Yasuhara, A. (2010). Correlation between EEG abnormalities and symptoms of autism spectrum disorder (ASD). *Brain and Development*, *32*(10), 791-798. https://doi.org/10.1016/j.braindev.2010.08.010

Yoon, S. H., Choi, J., Lee, W. J., & Do, J. T. (2020). Genetic and epigenetic etiology underlying autism spectrum disorder. *Journal of Clinical Medicine*, *9*(4), 966. https://doi.org/10.3390/jcm9040966

Zerbo, O., Qian, Y., Yoshida, C., Grether, J. K., Van de Water, J., & Croen, L. A. (2015). Maternal infection during pregnancy and autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *45*(12), 4015-4025. https://doi.org/10.1007/s10803-013-2016-3

Appendix A: Systematic Review

This systematic review has been reproduced with permission from Springer Nature.

Advances in Neurodevelopmental Disorders
https://doi.org/10.1007/s41252-021-00224-8

REVIEW

Systematic Review of the Effect of Modification of Antecedents in the Treatment of Sleep Problems Among Children on the Autism Spectrum

Kate A. M. Ford[1] · Laurie K. McLay[1] · Karyn G. France[1] · Neville M. Blampied[2] · Rosina M. Gibbs[1]

Accepted: 14 October 2021

Abstract

Objectives Sleep problems are widely reported among children and adolescents on the autism spectrum. There is emerging evidence of the effectiveness and social validity of parent-implemented behavioral sleep interventions for children and adolescents on the autism spectrum. However, most research is focused on multi-component interventions that include the use of extinction, and questions remain about the effectiveness of modification to antecedent variables (including sleep/wake rescheduling) alone. This systematic review aims to summarize and appraise the literature examining the effectiveness of antecedent-based modifications for treating sleep problems among children on the autism spectrum.

Methods Eligible studies included one or more participants aged 2 to 18 years with autism and treated sleep problems using modification to antecedent variables as standalone treatments. Data were identified from PsycINFO, Cochrane Library, Scopus, and MEDLINE databases and extracted according to participant characteristics and sleep problems, study design, characteristics of the intervention, dependent variables and sleep measures, treatment outcomes, and social validity. Study quality was evaluated using a standardized rubric and was classified as "strong," "adequate," or "weak."

Results Overall, 12 studies met inclusion criteria. All studies reported improvements in sleep problems for most or all participants following antecedent-based modifications. Improvements were maintained at follow-up for all participants for whom this data was collected. Only 3 studies met criteria reflecting strong methodological rigor.

Conclusions Modification to antecedent variables can reduce or eliminate sleep problems; however, there is a need for more rigorous evaluation and systematic extension of these findings. Minimally sufficient modification of antecedent variables may be implemented as a less restrictive alternative to consequence-based interventions.

Systematic review registration: PROSPERO CRD42021240539.

Keywords Autism spectrum disorder · Sleep problems · Behavioral sleep interventions · Antecedent-based modifications · Review

Children on the autism spectrum demonstrate core challenges in social interaction and communication and restricted, repetitive behaviors, activities, or interests (American Psychiatric Association, 2013). Many children and adolescents (henceforth referred to as children) on the autism spectrum also experience sleep disturbance. According to prevalence estimates, around 50 to 80% of children on the autism spectrum experience some type of sleep problem (Hodge et al., 2014; Kotagal & Broomall, 2012; Schreck & Schreck, 2020). This compares to 20 to 37% of typically developing children (Lozoff et al., 1985; Owens et al., 2000), around 20 to 50% of children with attention-deficit/hyperactivity disorder (ADHD; Stein, 1999; Sung et al., 2008), and 13 to 86% of children with other types of developmental disabilities (Didden & Sigafoos, 2001). Dyssomnias, a group of sleep problems defined by difficulty initiating or maintaining sleep, are the most frequently reported type of sleep problem among children on the autism spectrum (Cortesi

✉ Kate A. M. Ford
kate.ford@pg.canterbury.ac.nz

[1] School of Health Sciences (Te Kura Mātai Hauora), University of Canterbury (Te Whare Wānanga O Waitaha), Private Bag 4800, Christchurch 8140, New Zealand

[2] School of Psychology Speech & Hearing (Te Kura Mahi ā-Hirikapo), University of Canterbury (Te Whare Wānanga O Waitaha), Private Bag 4800, Christchurch 8140, New Zealand

Published online: 15 November 2021

et al., 2010; Goldman et al., 2012), often resulting in chronic sleep deprivation.

Sleep problems have a significant impact on the health and well-being of children on the autism spectrum, their parents, and family (Carnett et al., 2021; Meltzer & Thomas, 2020). For example, inadequate sleep has been associated with the severity of autism symptoms (Hoffman et al., 2008; Schreck et al., 2004), cognitive functioning (Hollway et al., 2013), and emotional (Malow et al., 2006), behavioral (Goldman et al., 2011), and medical issues (Liu et al., 2006; Williams et al., 2004). For other family members, childhood sleep problems have been shown to negatively affect marital relationships, parenting practices, family functioning and well-being, and parental sleep, mental health, and levels of stress (Carnett et al., 2021; Davis & Carter, 2008; Foody et al., 2015; Hodge et al., 2013; Meltzer, 2010; Meltzer & Mindell, 2007).

The etiology of sleep problems in children on the autism spectrum is multi-factorial (Richdale & Schreck, 2009), resulting from a complex interaction between biological (e.g., problems with the timing and production of melatonin), medical (e.g., sleep-related breathing disorders, seizures), psychological (e.g., sleep-interfering anxiety), social, and/or behavioral factors (Heussler & Malow, 2020; Johnson & Malow, 2008; Reynolds & Katz, 2020; Richdale & Schreck, 2009). In children on the autism spectrum in particular, there are salient biobehavioral factors that may contribute to sleep problems. The two-process model of sleep regulation (Borbély et al., 2016) proposes that sleep and wakefulness are regulated by circadian and homeostatic processes, which are both behavioral and physiological (Borbély et al., 2016; Glickman, 2010). Circadian rhythms produced by our internal biological clock must be reset and synchronized with environmental time cues daily (Borbély, 1982; Borbély et al., 2016; Glickman, 2010). Melatonin, the neurohormone integral to the regulation of the sleep/wake cycle, has been found in low levels in individuals on the autism spectrum (Bourgeron, 2007). Low levels of melatonin and abnormal melatonin regulation, synthesis, and synaptic transmission may lead to circadian rhythm disruptions and thus, sleep problems in children on the autism spectrum (Bourgeron, 2007; Melke et al., 2008). This biological predisposition for sleep problems may be exacerbated by behavioral factors, many of which are associated with the core symptoms of autism, placing these children at increased risk of sleep problems (Deliens & Peigneux, 2019).

The behavioral model of sleep disturbance postulates that falling asleep represents the endpoint of an operant behavior chain, which starts with the bedtime routine and ends with behavioral quietude (i.e., lying still and silently in bed, with minimal cognitive, behavioral, and emotional arousal) necessary for sleep onset (the natural reinforcer) (Blampied, 2013; Blampied & France, 1993; France & Blampied, 1999). There are two important ways in which antecedent variables (i.e., variables operating prior to the occurrence of a behavior that influence the behavior) affect this behavior chain. The first of these involves the role of discriminative stimuli: the operant chain of behaviors is under stimulus control wherein each step in the chain (e.g., brushing teeth, putting on pajamas, reading a bedtime story) may supply the discriminative stimulus for the following step (Blampied, 2013). By contrast, an absence of consistent discriminative stimuli as well as the occurrence of behaviors that are incompatible with sleep (behaviors that are part of other behavior chains) may inhibit sleep-related cues from being discriminated and result in abandonment of the behavior chain that facilitates sleep onset. Moreover, when particular stimuli are consistently paired with sleep, children develop a reliance on these cues for initiating and maintaining sleep. In the case of discriminative stimuli that are present throughout the night (e.g., a dimly lit bedroom or a cuddly toy), this can facilitate the onset and maintenance of sleep. If, however, these discriminative stimuli are not consistently present (e.g., the bedtime routine is highly variable) or the conditions present during initial sleep onset are not present during night wakings (e.g., parental presence at initial sleep onset but not at later wakings), this can interfere with sleep maintenance (Blampied & France, 1993; Weiskop et al., 2005).

The second set of critical antecedent variables is concerned with motivation to sleep, where this motivational state is referred to as sleep pressure (Borbély, 1982; Borbély et al., 2016). Sufficient sleep pressure is necessary for rapid sleep onset and initial maintenance. Sleep pressure can be understood as a motivational variable that can increase the reinforcement value of sleep and thereby increase both the salience of the discriminative stimuli that signal the availability of the reinforcer (sleep) and the potency of sleep as a reinforcer. The conditions that impact an individual's motivation to sleep, and subsequently, the value of the reinforcer, are called *motivating operations* (MOs; Cooper et al., 2020; Michael, 1982). In the case of sleep, feeling deprived of sleep, experiencing a delayed bedtime, the amount of time awake, and exhaustion from daytime behaviors may increase the reinforcement value of sleep itself, and therefore, and in turn, an individual's motivation to sleep (Borbély & Achermann, 1992; Michael, 1982).

Reinforcement is also necessary to strengthen and maintain sleep-compatible behaviors that occur as a part of the behavior chain. Consequences, defined as "a stimulus change that follows a behavior of interest," may increase or decrease the future likelihood of a behavior (Cooper et al., 2020, pg. 789). If sleep-conducive behaviors (e.g., lying quietly in bed) are reinforced, these behaviors are more likely to occur again. By contrast, reinforcement of sleep-interfering behavior (e.g., calling out from the bedroom) is likely to result in continuation of this behavior. This makes it imperative to identify and control the specific antecedent and consequence

variables that are instrumental in maintaining sleep problems (Jin et al., 2013).

In accordance with the known etiology, sleep problems in children on the autism spectrum are often treated using pharmacological and behavioral interventions. For example, there is strong evidence to support the use of exogenous melatonin for children on the autism spectrum (Malow et al., 2012; Wright et al., 2011). Melatonin regulates the circadian rhythm by entraining an individual's biological clock to a 24-hour cycle (Richdale & Schreck, 2009). While effective, a number of concerns have been raised about the safety and efficacy of long-term melatonin use. Further, many families encounter issues with accessing or funding it, and while it effectively treats issues with sleep onset, problems with night wakings are not always resolved. Behavioral interventions have, therefore, been recommended for use, in conjunction with melatonin (Cortesi et al., 2012).

The effectiveness of behavioral interventions has been widely investigated though scarcely, in combination with melatonin. A recent meta-synthesis of published systematic reviews examining sleep interventions for children on the autism spectrum provides evidence of the effectiveness of behavioral sleep interventions and those that are based on parent psychoeducation (Cuomo et al., 2017). These findings have been replicated in a number of systematic reviews, reports, and experimental studies (e.g., Carnett et al., 2020; Gringras et al., 2017; McLay et al., 2021; Rigney et al., 2018; Rossignol & Frye, 2011). Behavioral sleep interventions can include modification to both antecedent and consequence variables. Antecedent-based modifications include modification to sleep hygiene practices (i.e., modifiable parent and child habits that support good quality sleep), such as establishing a consistent bedtime routine and modification to the sleep environment (Bathory & Tomopoulos, 2017; Blampied, 2013; Mindell et al., 2009). Research suggests that a structured, consistent, and calming bedtime routine (e.g., having a bath, putting on pajamas, brushing teeth, bedtime stories) and a sleep-conducive environment (e.g., a cool, quiet, dark room) are critical in ensuring good quality sleep (Bathory & Tomopoulos, 2017; Blampied, 2013; Mindell et al., 2009). Antecedent-based modifications also include those that target circadian rhythms and physiological sleep pressure via modification to sleep/wake schedules (e.g., implementation of consistent sleep/wake schedules or sleep restriction).

Consequence-based interventions are those that involve modification to contingencies of reinforcement for sleep-competing or sleep-conducive behavior (Blampied, 2013). Approaches that include modification of consequences that have empirical support in the treatment of sleep problems in children on the autism spectrum include planned ignoring (i.e., extinction), modified extinction procedures (e.g., graduated extinction), and use of reinforcement for sleep-conducive behavior (e.g., use of rewards) (McLay et al., 2019; Weiskop et al., 2001). Typically, these approaches are delivered alongside other interventions, including modification to antecedent variables.

While extinction-based procedures are empirically supported, some parents express reservations about the side effects of these procedures, including extinction bursts and associated parent and child distress (Gradisar et al., 2016; Owens et al., 1999). Extinction bursts are a transient increase in the frequency, duration, or intensity of a problem behavior following the removal of reinforcement (Lerman & Iwata, 1995; Owens et al., 1999). While there is no evidence of negative long-term consequences of extinction bursts on parent–child attachment or the development of emotional or behavioral problems (Gradisar et al., 2016; Price et al., 2012), these beliefs persist among parents. The occurrence of an extinction burst can also make it difficult for parents to adhere to interventions, compromising treatment fidelity and in some cases, resulting in further deterioration in children's behavior. For example, parents may have difficulty tolerating an escalation in behavior, resulting in them abandoning the intervention plan. As a result, these escalations in behavior become reinforced, resulting in an increase in the likelihood that these behaviors will be repeated.

Within recent research, behavioral sleep interventions have been selected based upon the outcomes of functional behavioral assessment (FBA). FBA is an evidence-based approach to assessment that is used to identify the setting events, antecedents, and consequences precipitating and maintaining problem behaviors and the potential function of that behavior (Blampied, 2013; Cooper et al., 2020). This information is then used to develop an individualized treatment plan that directly targets these variables (Brown & Piazza, 1999; Jin et al., 2013). Within the sleep context, function-based interventions have typically consisted of multiple components, including antecedent- and consequence-based modifications, implemented concurrently or sequentially so as to address the various functions identified in the FBA. While there is sound rationale for this approach, it can be difficult to isolate individual treatment effects and in turn, to determine which elements of intervention result in behavior change. Moreover, these multi-component interventions are often time and resource intensive, as well as complex for families to implement.

Owing to the challenges associated with implementing sleep interventions (e.g., they are multi-faceted, implemented in the night when parents and children are tired and sleep-deprived, and implemented outside of the hours when clinical services are typically provided resulting in a lack of real-time support), it is important to identify sleep interventions that utilize the least restrictive approach, ones that are simple for parents to understand and implement, that minimize parent and child distress, and that

are effective, safe, and efficient. In psychology, these are conceptualized as *minimally sufficient* and *least restrictive* interventions (Johnston & Sherman, 1993; Sanders et al., 2014).

This research, along with the clinical experience of the current authors, has raised an important question about whether sleep problems in children on the autism spectrum can be effectively treated without the use of reinforcement, extinction, and modified extinction procedures, i.e., without manipulation of consequences. This systematic review partially addresses this question by identifying, synthesizing, and evaluating research that examines the effectiveness of modification to antecedent variables including modification to sleep/wake schedules alone, in the treatment of sleep problems among children on the autism spectrum. This review provides a systematic summary of study characteristics according to participants (including number, gender, age, medication, diagnosis, comorbidities), reported sleep problems, study design, characteristics of the intervention, dependent variables and sleep measures, treatment outcomes, social validity, and study quality.

Method

This systematic review was registered with PROSPERO in March 2021 and conducted in accordance with the PRISMA guidelines (Liberati et al., 2009; Moher et al., 2009).

Search Procedures

A search was undertaken in the PsycINFO, Cochrane Library, Scopus, and MEDLINE electronic databases. The final search was conducted in March 2021 by the corresponding author. The search terms entered were autis*; ASD; Asperger*; child*; preschool*; adolesc*; teen*; sleep; circadian rhythm*; circadian manipulation*; modifications to sleep/wake schedules; sleep schedules; sleep association*; stimulus substitution*; visual support*; antecedent modification*; intervention*; sleep hygiene; sleep restriction; faded bedtime. All identified articles were imported into an EndNote library and duplicates were removed. The corresponding author initially assessed the articles for eligibility by screening the titles and abstracts. Following initial screening, the full text of articles that appeared to meet inclusion criteria were read in full. An additional article was sourced through an ancestral search of the reference lists of included articles. The search procedures and process of selecting the final number of included studies is shown in Fig. 1.

Inclusion and Exclusion Criteria

Articles were eligible for inclusion if they (a) examined the effects of modification to antecedent variables, including modification to sleep/wake schedules as a treatment for sleep problems; (b) were published in a peer-reviewed journal; (c) were written in English; and (d) included at least one participant on the autism spectrum aged 2 to 18 years. There were no limitations placed on the study design or year of publication. Antecedent-based modifications were defined as interventions that are designed to increase physiological sleep pressure (e.g., faded bedtime), target the sleep environment (e.g., sleep hygiene), or events that precipitate the onset of sleep problems (e.g., stimulus control) (Blampied, 2013; McLay et al., 2017). Articles were excluded from the present study if (a) they examined the effects of consequence-based interventions (e.g., extinction, modified extinction, use of reinforcement) alongside antecedent-based modifications and (b) the full text was not available or in press (i.e., Cochrane Library database clinical trial). However, articles were eligible for inclusion if they included consequence-based modifications that could be clearly differentiated from antecedent-based modifications.

A total of 1248 articles were identified via database searching. One additional article was identified through the ancestral search. Overall, a total of 840 articles remained after duplicates were removed. Titles and abstracts were screened, and 779 articles were excluded. The full text of 61 articles were read and 49 were excluded because the article included consequence-based interventions in conjunction with antecedent-based modifications, did not report the effectiveness of antecedent-based modifications, the full text was not available, there were no participants on the autism spectrum, or it was a registered clinical trial that was not published. A second reviewer (fifth author) also read the full-texts of 61 articles and discussed eligibility with the corresponding author. The remaining 12 studies were included in this review.

Data Extraction

Each article that fulfilled the inclusion criteria was summarized in a table according to (a) authors; (b) participants including number, gender, age, medication, diagnosis, and comorbidities; (c) reported sleep problems; (d) study design; (e) characteristics of the intervention; (f) dependent variables and sleep measures; (g) treatment outcomes; (h) social validity; and (i) study quality. The corresponding author reviewed the included studies and extracted the data. Following this process, the second reviewer (fifth author) checked the summary table for accuracy. Any discrepancies

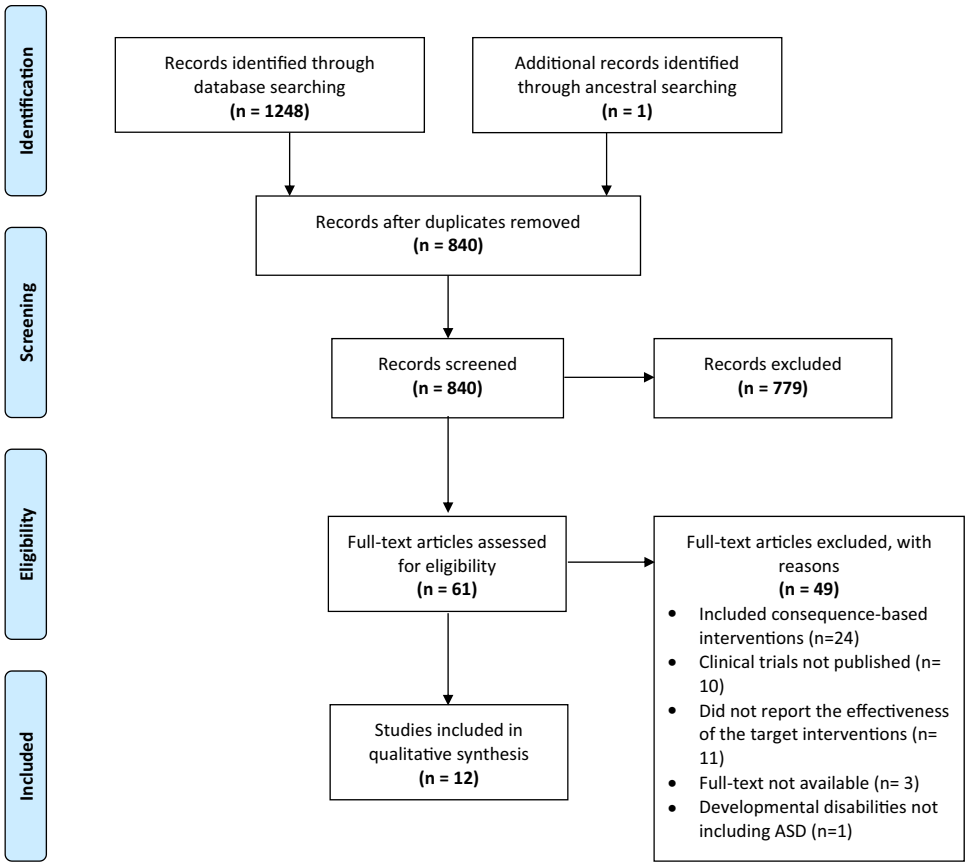


Fig. 1 PRISMA flowchart of search procedures and included studies (Moher et al., 2009)

between the first and fifth author were discussed until a consensus had been met.

Evaluation of Study Quality

To evaluate study quality, the authors used two pre-existing rubrics developed by Reichow et al. (2008), designed to identify evidence-based practices for children on the autism spectrum. One rubric was developed to evaluate single-subject research and the other for group research (Reichow et al., 2008). Within each rubric, study quality ("strong", "adequate", "weak") is determined based on primary quality indicators and secondary quality indicators (Reichow et al., 2008, p. 1313). Primary quality indicators were rated as either "high", "acceptable", or "unacceptable" (Reichow et al., 2008, p. 1312), and secondary quality indicators were rated on a dichotomous scale (i.e., "yes" or "no") (Reichow et al., 2010). For the single-subject research rubric, primary quality indicators pertained to (a) characteristics of the participant, (b) independent variable(s), (c) dependent variable(s), (d) baseline characteristics, (e) visual analysis graphs, and (f) experimental control, and secondary indicators were (a) interobserver agreement, (b) kappa, (c) treatment and/procedural fidelity, (d) blinding, (e) maintenance or generalization, and (f) social validity (Reichow et al., 2008). For the group research design rubric, primary indicators included items a–c above, in addition to criteria relating to the (d) comparison group, (e) relationship between data analysis and research questions, and (f) statistical testing. Secondary indicators included criteria identical to many of the indicators for single-subject research (a, c–f), in addition to (g) randomization, (h) attrition, and (i) effect sizes (Reichow et al., 2008). In addition to these rubrics (Reichow et al., 2008), researchers used a supplementary resource to aid in the evaluation (Reichow et al., 2010). The supplementary resource (Reichow et al., 2010) contains the criteria used to award each primary quality indicator as high, acceptable, or unacceptable. The evaluation was completed by the corresponding author in April 2021 and repeated by the second author in July 2021.

Results

A total of 12 studies were eligible for inclusion in this review. Table 1 summarizes included studies in terms of participant characteristics, reported sleep problems, study design, intervention characteristics, dependent variables, sleep measures, treatment outcomes, social validity, and study quality.

Table 1 Summary of articles examining modification to antecedent variables for treating sleep problems among children on the autism spectrum

Authors	Participants (*n*, gender, age, medication, diagnosis, comorbidities) and sleep problems	Study design; follow-up	Intervention characteristics	Sleep measures; dependent variables	Treatment outcomes	Social validity	Study quality
Christodulu and Durand (2004) [a]	*n* = 4, 2 M, 2F. 2–5 years. 2 = ASD [b] (autism, PDD + sensory integration disorder + hypotonia); 1 = Charge; 1 = immune deficiency (IgA) *Sleep problems:* freq. and duration of bedtime disturbances and NWs	Multiple baseline across participants design *FU:* 1-month post-treatment	Sleep restriction, positive bedtime routines (simultaneous)	*ASPS:* to assess type and severity of sleep problems *Actigraphy:* sleep/wake cycle data (*n* = 1); time asleep, time awake, timing and duration of NWs *Daily sleep charts and bedtime behavior logs:* child's sleep schedule, freq. and duration of bedtime disturbances, NWs and naps, behavior during bedtime disturbances and NWs, timing of NWs	*Daily sleep charts and bedtime behavior logs:* improvements across variables except reduced TST (*n* = 3) and no change in TST (*n* = 1 [b]) *FU:* reduced freq. and duration of bedtime disturbances and NWs were maintained	*SV-PSSQ:* scores for all children increased post-treatment, further increases at 1-month FU	Adequate
Delemere and Dounavi (2018)	*n* = 6, 4 M, 2F. 2–6 years. 6 = ASD *Sleep problems:* SOD, EMWs, NWs, sleep duration	Multiple baseline across participants design *FU:* not assessed	Bedtime fading (*n* = 3), positive routines (with visual schedules) (*n* = 3)	*SATT:* to assess the relationship between environmental variables and sleep problems *CSHQ:* to assess the child's sleep problems *Sleep diary:* bedtime, time asleep, time awake, freq. and duration of NWs, TST, freq. and duration of naps, bedtime resistance, co-sleeping, confidence in implementing procedure	*Sleep diary:* improvements across variables except no change in TST for one child with positive routines, no changes in freq. and duration NWs (low at baseline)	*SV -TAI:* high acceptability; bedtime fading (*M* = 47.7/50), positive routines (*M* = 48.3/50) *SV-TEI-SF:* high social validity for both interventions (*M* = 41/50)	Strong
DeLeon et al. (2004) [a]	*n* = 1, 1 M. 4 years. 1 = autism, developmental delay *Sleep problems:* NWs, nighttime SIB	AB single-case design *FU:* not assessed	Faded bedtime [b], scheduled awakenings (SIB)	*Trained observers*: Sleep state (awake/asleep) and SIB was recorded using a counter and a data sheet	*Trained observers:* reductions in freq. of NWs (and SIB)	*SV:* data not reported	Weak

Table 1 (continued)

Authors	Participants (*n*, gender, age, medication, diagnosis, comorbidities) and sleep problems	Study design; follow-up	Intervention characteristics	Sleep measures; dependent variables	Treatment outcomes	Social validity	Study quality
Durand (2002)	*n* = 3, 2 M, 1F. 3–7 years. 3 = autism *Sleep problems*: sleep terrors	Multiple baseline across participants design *FU:* 12 months' post-treatment	Scheduled awakenings	*Sleep charts and behavior logs*: onset and duration of sleep terrors, bedtime, SOL, wake time, time and duration of daytime naps, bedtime behavior and sleep terror behavior	*Sleep charts:* reduction in freq. of sleep terrors children experienced per week across participants *FU:* gains maintained at 12 months	*SV*: high treatment acceptability and effectiveness, at 12 months follow-up	Strong
Durand and Christodulu (2004) [a]	*n* = 2, 2F. 4 years. 1 = autism [b], 1 = developmental delay *Sleep problems*: bedtime disturbances, NWs	Multiple baseline across participants design	Sleep restriction	*ASPS*: to assess type and severity of sleep problems *Sleep charts and behavior logs:* child's sleep schedule, bedtime, time awake, freq. and duration of bedtime disturbances, bedtime disturbances behavior, freq. and duration of NWs, NWs behavior, freq. and duration of naps	*Sleep charts*: reductions in freq. and duration of NWs and bedtime disturbances for both participants	*SV-PSSQ:* improved scores post-treatment	Weak
Luiselli et al. (2020)	*n* = 1, 1F. 18 years. 1 = ASD, global developmental delay *Sleep problems*: SOD	AB single-case design *FU:* 1-month post-treatment	Faded bedtime	*Observation:* Care providers observed the child using a computer-assisted system to monitor and record sleep and recorded SOL and TST	*Observation:* improvement in SOL and TST *FU*: gains were maintained and improved at 1-month follow-up	*SV:* data not reported	Weak
Luiselli et al. (2021)	*n* = 1, 1 M. 14 years 1 = ASD, seizure disorder *Sleep problems*: SOD, short sleep duration	AB single-case design *FU:* 2 months' post-treatment	Faded bedtime	*Observation:* Care providers observed and recorded sleep information (awake/asleep) using a computer-assisted system to monitor and record sleep	*Observation:* improvement in SOL and TST *FU*: gains were maintained and improved at 2 months follow-up	*SV:* data not reported	Weak

Table 1 (continued)

Authors	Participants (*n*, gender, age, medication, diagnosis, comorbidities) and sleep problems	Study design; follow-up	Intervention characteristics	Sleep measures; dependent variables	Treatment outcomes	Social validity	Study quality
Malow et al. (2016) [a]	*n* = 10, 8 M, 2F. 3–9 years. 10 = ASD. 2 = melatonin, 1 = Singulair [b], 1 = Zyrtec + Flonase + albuterol, 1 = Vyvanse + risperidone *Sleep problems*: SOD, co-sleeping, bedtime resistance, NWs	Pre-/post-test design *FU:* not assessed	Sleep education program (child 3 [b] and child 6 [b]): consistent pre-bed routine with visual supports [b] (e.g., visual schedule), delayed bedtime [b], restricted access to screens before bed [b], relaxing activities before bed [b], restricted caffeine [b]	*CSHQ:* to assess the child's sleep problems *FISH:* to measure sleep habits *PAS:* parent's understanding and implementation of sleep education *Actigraphy:* SOD, WASO *Sleep diaries:* daytime habits (e.g., caffeine, exposure to light, exercise), evening activities, bedtime routine, sleep resistance, time awake, time asleep	*CSHQ/parent report:* improvements for both children. Child 3; reduction in NWs, no change in SOL. Child 6; improvements in SOD, NWs, bedtime resistance, co-sleeping *Actigraphy:* reduction in SOL for both children	*SV:* data not reported	Weak
Piazza et al. (1997) [a]	*n* = 14. 4–14 years. *FBRC group*: 2 = autism [b], 1 = PDD [b] + seizure disorder + cerebral palsy, 1 = Down syndrome, 1 = Prader-Willi + sleep apnea, 1 = mixed motor encephalopathy, 1 = seizure disorder *Bedtime schedule group*: 1 = autism [b], 1 = autism [b] + seizure disorder + cerebral palsy, 1 = cerebral palsy + seizure disorder, 1 = Down syndrome, 3 = various seizure disorders *Sleep problems*: NWs, EMWs, SOD, sleep duration	Between-groups design *FU:* not assessed	FBRC, bedtime scheduling [b]	*Momentary time sampling procedure:* Each child was recorded as awake or asleep every 30 minutes by observers. Observers also recorded time asleep, time awake, timing and duration of NWs or EMWs	*Momentary time sampling procedure:* improvement in sleep, reduction in hours of disturbed sleep in the children with ASD, FBRC = greater improvements in SOL, NWs, EMWs	*SV:* data not reported	Weak

Table 1 (continued)

Authors	Participants (*n*, gender, age, medication, diagnosis, comorbidities) and sleep problems	Study design; follow-up	Intervention characteristics	Sleep measures; dependent variables	Treatment outcomes	Social validity	Study quality
Piazza et al. (1998)	*n* = 1, 1F. 8 years. 1 = autism, intellectual disability, congenital omphalocele, food refusal *Sleep problems*: irregular sleep onset times, NWs, EMWs, sleep duration	AB single-case design *FU:* 4 months post-treatment	Chronotherapy	*Momentary time sampling:* Trained observers recorded the child as awake or asleep, and in or not in bed every 30 minutes. Observers also recorded time asleep, time awake, timing and duration of NWs or EMWs	*Momentary time sampling:* improvements in child's sleep schedule (time asleep/awake), goal bedtime implemented in 11 days *FU:* gains were maintained at 4 months follow-up	*SV:* data not reported	Weak
van Deurs et al. (2019) [a]	*n* = 3, 3 M. 9–14 years. 1 = ASD [b], 1 = ASD, 1 = Asperger's syndrome. 1 = melatonin + risperidone + fluoxetine *Sleep problems*: NWs, EMWs, SOD, CCs	AB or ABCDE single-case design *FU:* 18 to 24 months post-treatment	Multi-component intervention ("Peter"): Bedtime fading [b], social story [b] and relaxation strategies [b]	*Sleep diaries:* freq. and duration of naps, SOL, freq. of CCs, freq. and duration of NWs, time of EMWs *VSG:* nighttime and sleep behavior, SOL, freq. of CCs, freq. and duration of NWs, time of EMWs *CSHQ:* to assess changes in sleep problems pre- and post-treatment	*Sleep diaries:* improvements across variables for all children *CSHQ*: improvements across variables for all children *FU*: gains were maintained at 18- or 24-month post-treatment	*SV-post-treatment interviews:* interventions were acceptable to all children and parents *SV-TARF-R:* highly acceptable ratings by parents	Strong

Table 1 (continued)

Authors	Participants (*n*, gender, age, medication, diagnosis, comorbidities) and sleep problems	Study design; follow-up	Intervention characteristics	Sleep measures; dependent variables	Treatment outcomes	Social validity	Study quality
van Deurs et al. (2020)	*n* = 1, 1F. 9 years. 1 = ASD, selective mutism *Sleep problems:* CCs, SOD, NWs, sleep duration, SE	AB single-case design *FU:* 10 weeks post-treatment	*Multi-component intervention:* white noise [b], relaxation instruction [b], stimulus control [b], reinforcement, unmodified extinction	*SATT:* to assess factors underlying the child's sleep problems *CSHQ:* to assess changes in sleep problems pre- and post-treatment *SSR:* child-report measure to assess changes in sleep pre- and post-treatment *Sleep diaries:* freq. and duration of naps, SOL, freq. of CCs, freq. and duration of NWs, time of EMWs *VSG:* nighttime and sleep behavior, freq. and duration of naps, SOL, freq. of CCs, freq. and duration of NWs, time of EMWs	*Sleep diaries, CSHQ, SSR:* improvements across variables by phase 3 (white noise + relaxation + stimulus control) *FU:* gains maintained at 10 weeks follow-up	*SV-YPTE:* rated moderately acceptable by child *SV-TARF-R:* highly effective, reasonable, and low-cost but moderately time-consuming and disruptive	Weak

ASD autism spectrum disorder, *ASPS* Albany Sleep Problems Scale, *CCs* curtain calls, *CSHQ* Children's Sleep Habits Questionnaire, *EMWs* early morning wakings, *F* female, *FBRC* faded bedtime with response cost, *FISH* Family Inventory of Sleep Habits, *freq.* frequency, *FU* follow-up, *M* male, *n* number, *NWs* night wakings, *PAS* Parent Absorption Scale, *PDD* pervasive developmental disorder, *PSSQ* Parental Sleep Satisfaction Questionnaire, *SATT* Sleep Assessment Treatment Tool, *SE* sleep efficiency, *SIB* self-injurious behavior, *SOD* sleep onset delay, *SOL* sleep onset latency, *SSR* Sleep Self-Report, *SV* social validity, *TAI* Therapy Attitude Inventory, *TARF-R* Treatment Acceptability Rating Form Revised, *TEI-SF* Treatment Evaluation Inventory-Short Form, *TST* total sleep time, *VSG* videosomnography, *WASO* wake after sleep onset, *YPTE* Young Person Treatment Evaluation

[a] indicates a study with a subset of ineligible participants or treatments

[b] the participants or intervention components that meet inclusion criteria

Participant Characteristics and Sleep Problems

The 12 studies included a total of 47 participants; however, only 35 participants had a formal diagnosis of ASD (range of 1 to 10 participants per study, $M = 2.9$). Further, data from 13 participants on the autism spectrum was not eligible for inclusion as consequence-based modifications were implemented alongside antecedent-based modifications. The mean age of participants on the autism spectrum was six years (range of 2 to 18 years). There were 20 males (57%), 10 females (29%), and 5 (14%) participants for whom gender was not reported. Six studies reported that participants had a co-occurring disability including developmental delay, seizure disorder, cerebral palsy, intellectual disability, or selective mutism. Four participants took medication, including melatonin, Singulair, Vyvanse, risperidone, and fluoxetine. All participants experienced at least one sleep problem, including bedtime disturbances, night wakings, sleep onset delay, early morning wakings, short sleep duration, sleep terrors, unwanted co-sleeping, irregular sleep onset times, and curtain calls.

Study Design

All but two studies used single-case research designs, including an AB single-case design ($n = 6$) or a multiple baseline across participants design ($n = 4$). One study used a between-groups design to compare faded bedtime (with response cost) and bedtime scheduling procedures. The other remaining study used a pre-/post-test design to pilot a sleep education program for parents.

Intervention Characteristics

All studies included modification to antecedent variables including modification to sleep/wake schedules as standalone interventions. Treatment components included setting a consistent bedtime and wake time, faded bedtime, sleep restriction, scheduled awakenings, chronotherapy, sleep hygiene practices, visual supports, and stimulus control. The faded bedtime procedure was used in five studies and typically involved selecting an initial bedtime that was within 15 minutes of the child's usual sleep onset time and waking the child at the same time each morning (Piazza et al., 1997; Vriend et al., 2011). The bedtime was then shifted progressively earlier until the goal bedtime was reached, while maintaining a consistent wake time (Piazza et al., 1997; Vriend et al., 2011). Sleep restriction was used in two studies and involved restricting the time the child was allowed to spend in bed to 90% of the child's baseline total sleep time (Christodulu & Durand, 2004; Durand & Christodulu, 2004). As with faded bedtime, the child's bedtime was then shifted earlier until the goal bedtime was met. Scheduled awakenings were used in one study and involved parents pre-emptively waking the child before the time of their usual night wakings (Owens et al., 1999). The amount of time between scheduled awakenings was progressively increased with the goal of the child sleeping through the night undisturbed (Owens et al., 1999). Chronotherapy was used in one study and involved shifting the child's average bedtime and wake time two hours later for eight nights, and then one hour later until the goal sleep schedule was achieved (Piazza et al., 1998; Vriend et al., 2011). The child's normal daytime routine of eating breakfast, doing schoolwork, and having a lunch break was maintained despite the unconventional timing of the sleep/wake schedule.

Treatment components that addressed sleep hygiene included establishing a consistent bedtime routine/positive routine ($n = 4$ studies), restricted access to TV and/or electronic devices before bedtime ($n = 2$), restricted caffeine intake ($n = 1$), and relaxation activities ($n = 3$) such as reading, progressive muscle relaxation, and diaphragmatic breathing. Additional antecedent-based interventions included white noise ($n = 1$), visual supports ($n = 3$), and stimulus control strategies ($n = 1$). White noise (e.g., fire crackling, cat purring) was used to minimize the child's ability to hear external parental noises and provide a discriminative stimulus for sleep onset. Visual supports included the use of a Gro-Clock™ (a clock-like device that shows a face with stars or sun, depending on when waking time is set), pictorial schedule, and/or a social story. Stimulus control techniques included switching off the child's light at bedtime and having a consistent bedtime routine.

Most interventions ($n = 7$) were implemented by parents in the home, following either a one-off training providing verbal and/or written instructions ($n = 3$), instructions delivered with ongoing support and contact ($n = 3$), or a sleep education program manual ($n = 1$). Five studies used interventions that were implemented by staff in an inpatient unit ($n = 3$), a group home ($n = 1$), or in a residential care setting ($n = 1$). There was variable treatment duration across studies, ranging from 11 nights to 26 weeks ($M = 65.35$ nights).

Dependent Variables and Sleep Measures

The most common sleep measure was a daily sleep diary or chart ($n = 7$). Eighteen psychometric instruments were used across the 12 studies. Five psychometric instruments were used to measure children's sleep problems and sleep hygiene, namely, the Sleep Assessment and Treatment Tool ($n = 2$), the Children's Sleep Habits Questionnaire ($n = 4$), the Albany Sleep Problems Scale ($n = 2$), the Family Inventory of Sleep Habits ($n = 1$), and the Sleep Self-Report ($n = 1$). Five psychometric measures were used to assess children's communication abilities, adaptive behavior, and social, emotional, and behavioral difficulties, namely the

Child Behavior Checklist ($n = 1$), the Multidimensional Anxiety Scale for Children 2nd Edition ($n = 1$), the Questions About Behavior Function ($n = 1$), the Verbal Behavior Milestones Assessment and Placement Program ($n = 1$), and the Vineland Adaptive Behavior Scales Second or Third Edition ($n = 2$).

Five other psychometric measures were used to assess social validity. These were the Parental Sleep Satisfaction Questionnaire ($n = 2$), the Therapy Attitude Inventory ($n = 1$), the Treatment Evaluation Inventory-Short Form ($n = 1$), the Treatment Acceptability Rating Form-Revised ($n = 2$), and the Young Person Treatment Evaluation ($n = 1$). Other measures included treatment appropriateness, via the Sleep Intervention Screening Questionnaire ($n = 1$), socioeconomic status, (the Hollingshead Four-Factor Index of Socio-economic Status, $n = 1$), and parents' understanding and implementation of the sleep education strategies via the Parent Absorption Scale ($n = 1$). Additional measures included actigraphy ($n = 2$), behavior logs ($n = 3$), and video recordings ($n = 2$).

Dependent variables included sleep onset latency, frequency and/or duration of night wakings, total sleep duration, frequency, onset and/or duration of sleep terrors, frequency and/or duration of bedtime disturbances (e.g., tantrums, getting out of bed, and leaving their bedroom), frequency and/or duration of daytime naps, presence or absence of co-sleeping, frequency of curtain calls, timing of early morning wakings, as well as sleep onset time, wake time, behavior during bedtime disturbances and night wakings, and sleep efficiency.

Treatment Outcomes

Overall, modification to antecedent variables including modification to sleep/wake schedules reduced or eliminated sleep problems in children on the autism spectrum. For the most part, these findings were consistent across intervention type and dependent variables with some improvements reported in each of the studies. However, there were some participants that did not show improvement across all dependent variables. For example, total sleep time did not change for one child and reduced for three children following the implementation of sleep restriction and positive bedtime routines; however, sleep quality increased for the three children including two on the autism spectrum (Christodulu & Durand, 2004). Mixed findings were reported for two studies (Delemere & Dounavi, 2018; Malow et al., 2016), wherein a sleep education program for parents improved sleep onset latency, night wakings, co-sleeping, and/or bedtime resistance for six out of the eight children (Malow et al., 2016). Further, following the establishment of positive bedtime routines, sleep onset latency reduced for all children though sleep duration increased for only two of the three children (Delemere & Dounavi, 2018).

Seven studies evaluated the effectiveness of modification to sleep/wake scheduling alone on treatment outcome. Across five studies, the faded bedtime procedure resulted in reduced night wakings and sleep onset latency, and increased total sleep duration (Delemere & Dounavi, 2018; DeLeon et al., 2004; Luiselli et al., 2020; Luiselli et al., 2021; van Deurs et al., 2019). Two studies also found that sleep restriction (with positive bedtime routines) was effective for reducing night wakings and bedtime resistance for all participants (Christodulu & Durand, 2004; Durand & Christodulu, 2004).

Three studies implemented multi-component interventions consisting of both antecedent- and consequence-based modifications and found that antecedent-based modifications were effective in reducing a number of sleep problems in all of the children using those components in isolation (Malow et al., 2016; van Deurs et al., 2019; van Deurs et al., 2020). A number of individual studies examined the efficacy of chronotherapy, scheduled awakenings, and bedtime scheduling alone (Durand, 2002; Piazza et al., 1997; Piazza et al., 1998). To summarize: scheduled awakenings decreased the frequency of sleep terrors, chronotherapy improved the sleep/wake schedule, and bedtime scheduling reduced hours of disturbed sleep (Durand, 2002; Piazza et al., 1997; Piazza et al., 1998). Piazza et al.'s study was the only between-groups design, and these authors found that faded bedtime with response cost was more effective than bedtime scheduling alone in reducing sleep onset latency, night wakings, and early morning wakings.

Seven of the 12 studies collected data to assess the maintenance of treatment effects. Follow-up length varied between 4 weeks and 24 months, and findings indicated that gains were maintained in all studies for all participants.

Social Validity

The social validity and acceptability of antecedent-based modifications were assessed in six out of 12 studies. As previously mentioned, this was assessed using questionnaires and post-treatment interviews with parents and/or children. All parents and/or children reported high levels of satisfaction and acceptability with treatment, with the exception of one child who rated treatment as moderately acceptable. This participant's parents also reported that treatment was time-consuming and disruptive.

Study Quality

Three of the 12 studies met criteria to be classified as having strong study rigor; one study was classified as being of adequate quality; and eight studies including the between-groups design were evaluated as being weak. Five of the

eight studies were classified as weak, due to an unacceptable quality rating on the experimental control indicator. Furthermore, only seven of these 12 studies reported interobserver agreement (IOA), five reported treatment or procedural fidelity, and none reported kappa values.

Discussion

This systematic review aimed to identify, synthesize, and evaluate research examining the effectiveness of modification to antecedent variables for treating sleep problems in children on the autism spectrum. All 12 studies included in this review reported a reduction in sleep problems following modification of antecedents for most or all participants. Most studies reported consistency across interventions and sleep problem type (i.e., improvements were reported with faded bedtime, sleep restriction, positive bedtime routines, chronotherapy, scheduled awakenings, sleep hygiene, and/or bedtime scheduling), and treatment gains were maintained for all participants for whom follow-up data was collected. However, a small number of participants did not improve across all of the dependent variables. For example, not all children improved in terms of sleep duration in response to positive bedtime routines with or without sleep restriction. Overall, faded bedtime was the most frequently researched intervention and resulted in improvement across all participants. In general, interventions were implemented by parents in the home setting or by staff in an inpatient unit, a group home, or a residential care setting. The average duration of treatment was 65 nights; however, dosage ranged from 11 nights to 26 weeks. Parents were provided with either one-off training, ongoing support and contact, or a sleep education program manual. Interestingly, three studies used one-off parent training, suggesting that sleep interventions can be delivered efficiently.

There is currently a strong and growing body of literature that has demonstrated the effectiveness of behavioral sleep interventions for children on the autism spectrum. This systematic review examines the effectiveness of modification to antecedent variables, including modification to sleep/wake schedules, when implemented on their own, not combined jointly with modifications of consequences. The findings of this review highlight the effectiveness of less restrictive sleep interventions for treating a variety of sleep problems (e.g., problems with sleep onset delay, night wakings, short sleep duration, sleep terrors, bedtime disturbances, unwanted co-sleeping, curtain calls, early morning wakings, and irregular sleep onset and wake times). In particular, these findings suggest that sleep hygiene practices, faded bedtime, sleep restriction, positive bedtime routines, bedtime scheduling, scheduled awakenings, and chronotherapy may provide less restrictive alternatives to extinction-based procedures. Therefore, it is imperative that the design of a sleep intervention considers sleep hygiene practices, modification of discriminative stimuli for sleep onset, and increasing motivating operations for sleep. These findings also raise questions about the role of contingencies, such as social attention and access to preferred items, in sleep-interfering behavior, at the very least, suggesting that for many children treatment could commence with modification to antecedent variables alone, before proceeding to the implementation of extinction procedures or contingent reinforcement for sleep-conducive behavior. Further, additional external reinforcement or extinction may not be necessary if it is feasible to increase the reinforcement value of sleep by enhancing physiological sleep pressure and in turn increasing motivating operations for sleep. The evidence obtained in this review also suggests that antecedent-based modifications are viewed favorably by those responsible for implementing these procedures. As parents are required to implement sleep interventions overnight, it is important to identify interventions that are both socially valid, simple to implement, effective, and efficient. Identifying such interventions may increase treatment fidelity, a common issue in sleep research (van Deurs et al., 2021).

Limitations and Future Research

Notwithstanding the importance of these findings, this review has highlighted a number of limitations in the existing research. There are many challenges that have been identified in sleep research in children on the autism spectrum that were evident in this study. Measurement and data collection issues were noted such as the use of non-instrumental sleep measures (e.g., sleep diaries, questionnaires, versus actigraphy), limited treatment fidelity data, and an absence of individualized treatment planning (i.e., non-use of FBA). These challenges may be overcome by using multiple methods of data collection including instrumental observation (e.g., actigraphy, video footage, polysomnography), assessing social validity before commencing treatment, and conducting FBA during assessment using appropriate psychometric measures and observational data (Luiselli, 2021). Greater reporting of treatment or procedural fidelity is also needed.

Unfortunately, there is insufficient evidence to draw strong conclusions about the effectiveness of individual intervention procedures, due to there being very few replications of treatment effects within or across studies. For example, with the exception of faded bedtime, all other treatments were only used in individual studies or in addition to other interventions. This includes positive bedtime routines, scheduled awakenings, bedtime scheduling, and

chronotherapy. Sleep medicine practice parameters for typically developing infants and children report that parent education, positive bedtime routines, faded bedtime, and scheduled awakenings are effective in reducing common sleep problems (e.g., bedtime problems, night wakings) (Morgenthaler et al., 2006). However, the extent to which these findings can be generalized to children on the autism spectrum, given their unique characteristics and etiologies of sleep problems, is unclear.

The strength of our conclusions is also compromised by the quality of the existing research. As a result of the above limitations, most studies were classified as weak or adequate; only three studies included in this review were deemed to have strong research rigor, one study was classified as adequate, and eight studies were classified as weak. Common research limitations included insufficient experimental control, a small sample size, lack of reporting of effect sizes, limited reporting of maintenance or generalization of treatment gains, and limited interobserver agreement and reliability data. Thus, we conclude that antecedent-based modifications as intervention types are effective, but comparisons cannot be drawn at a more specific level, with confidence.

Based on the current corpus of research and associated limitations, future research might aim to improve methods of data collection, perhaps by using digital technologies to analyze all-night video recordings of sleep and bedtime behavior and improve reliability of data. Digital and internet-based technology also provides opportunities for real-time, or close to real-time, support for parents at bedtime and during the night and possibly for coaching during any initial parent education phase, with benefits for procedural fidelity and treatment acceptability (McLay et al., 2020). Future research could also assess social validity before starting the intervention to ensure that parents or care providers perceive the treatment plan as suitable and practical for the setting and efficient and acceptable (Luiselli, 2021). To address small sample sizes and insufficient experimental control, replication using multiple baseline across participant designs or RCTs is warranted to strengthen our conclusions and strengthen the evidence base. However, both single-case designs and RCTs should be analyzed in accordance with best practice research design and data analysis procedures, for example, measuring each dependent variable over time, conducting IOA, showing at least three demonstrations of the experimental effect and effect size reporting in single-case research (Kratochwill et al., 2010; Vannest & Ninci, 2015), and appropriate measurement and reporting of effect size and the avoidance of questionable research practices (including misinterpretation of *p*-values) in the case of group research (Cumming, 2014; Fritz et al., 2012; Kline, 2013; Lakens, 2013; Ranganthan et al., 2015; Wasserstein & Lazar, 2016; Wilkinson, 1999).

Overall, our findings indicate that modification to antecedent variables including modification to sleep/wake schedules may be both effective and acceptable for treating multiple sleep problems in children on the autism spectrum; however, there is a need for further robust, high-quality research in this area. This review adds to the current research base supporting the implementation of behavioral treatments for sleep problems in such children, including faded bedtime, and to our understanding of minimally sufficient interventions. These procedures may be implemented as an effective, less restrictive alternative to consequence-based interventions.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s41252-021-00224-8.

Author Contribution KF: conducted the search, assessed articles for eligibility, extracted the data, evaluated study quality, wrote the original draft, edited the paper. LM: conceptualization, reviewed and edited the paper, provided supervision. KF: conceptualization, reviewed and edited the paper, provided supervision. NB: conceptualization, reviewed and edited the paper, provided supervision. RG: assessed articles for eligibility, reviewed the extracted data, evaluated study quality.

Declarations

Competing Interests The authors declare no competing interests.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). https://doi.org/10.1176/appi.books.9780890425596

Bathory, E., & Tomopoulos, S. (2017). Sleep regulation, physiology and development, sleep duration and patterns, and sleep hygiene in infants, toddlers, and preschool-age children. *Current Problems in Pediatric and Adolescent Health Care, 47*(2), 29–42. https://doi.org/10.1016/j.cppeds.2016.12.001

Blampied, N. M. (2013). Functional behavioural analysis of sleep in infants and children. In A. R. Wolfson & H. E. Montgomery-Downs (Eds.), *The Oxford handbook of infant, child and adolescent sleep and behavior* (pp. 169–188). Oxford University Press.

Blampied, N. M., & France, K. G. (1993). A behavioral model of infant sleep disturbance. *Journal of Applied Behavior Analysis, 26*(4), 477–492. https://doi.org/10.1901/jaba.1993.26-477

Borbély, A. A. (1982). A two-process model of sleep regulation. *Human Neurobiology, 1*(3), 195–204.

Borbély, A. A., & Achermann, P. (1992). Concepts and models of sleep regulation: An overview. *Journal of Sleep Research, 1*(2), 63–79. https://doi.org/10.1111/j.1365-2869.1992.tb00013.x

Borbély, A. A., Daan, S., Wirz-Justice, A., & Deboer, T. (2016). The two-process model of sleep regulation: A reappraisal. *Journal of Sleep Research, 25*(2), 131–143. https://doi.org/10.1111/jsr.12371

Bourgeron, T. (2007). The possible interplay of synaptic and clock genes in autism spectrum disorders. *Cold Spring Harbor Symposia on Quantitative Biology, 72*, 645–654. https://doi.org/10.1101/sqb.2007.72.020

Brown, K. A., & Piazza, C. C. (1999). Commentary: Enhancing the effectiveness of sleep treatments: Developing a functional approach. *Journal of Pediatric Psychology, 24*(6), 487–489. https://doi.org/10.1093/jpepsy/24.6.487

Carnett, A., Hansen, S., McLay, L., Neely, L., & Lang, R. (2020). Quantitative-analysis of behavioral interventions to treat sleep problems in children with autism. *Developmental Neurorehabilitation, 23*(5), 271–284. https://doi.org/10.1080/17518423.2019.1646340

Carnett, A., McLay, L., Hansen, S., France, K., & Blampied, N. (2021). Sleep problems in children and adolescents with autism: Type, severity and impact. *Journal of Developmental and Physical Disabilities, 1–15*. https://doi.org/10.1007/s10882-020-09783-5

Christodulu, K. V., & Durand, V. M. (2004). Reducing bedtime disturbance and night waking using positive bedtime routines and sleep restriction. *Focus on Autism and Other Developmental Disabilities, 19*(3), 130–139. https://doi.org/10.1177/10883576040190030101

Cooper, J. O., Heron, T. E., & Heward, W. L. (2020). *Applied behavior analysis* (3rd ed.). Pearson.

Cortesi, F., Giannotti, F., Ivanenko, A., & Johnson, K. (2010). Sleep in children with autistic spectrum disorder. *Sleep Medicine, 11*(7), 659–664. https://doi.org/10.1016/j.sleep.2010.01.010

Cortesi, F., Giannotti, F., Sebastiani, T., Panunzi, S., & Valente, D. (2012). Controlled-release melatonin, singly and combined with cognitive behavioural therapy, for persistent insomnia in children with autism spectrum disorders: A randomized placebo-controlled trial. *Journal of Sleep Research, 21*(6), 700–709. https://doi.org/10.1111/j.1365-2869.2012.01021.x

Cumming, G. (2014). The new statistics: Why and how. *Psychological Science, 25*(1), 7–29. https://doi.org/10.1177/0956797613504966

Cuomo, B. M., Vaz, S., Lee, E. A. L., Thompson, C., Rogerson, J. M., & Falkmer, T. (2017). Effectiveness of sleep-based interventions for children with autism spectrum disorder: A meta-synthesis. *Pharmacotherapy, 37*(5), 555–578. https://doi.org/10.1002/phar.1920

Davis, N. O., & Carter, A. S. (2008). Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism and Developmental Disorders, 38*(7), 1278–1291. https://doi.org/10.1007/s10803-007-0512-z

Delemere, E., & Dounavi, K. (2018). Parent-implemented bedtime fading and positive routines for children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 48*(4), 1002–1019. https://doi.org/10.1007/s10803-017-3398-4

DeLeon, I. G., Fisher, W. W., & Marhefka, J.-M. (2004). Decreasing self-injurious behavior associated with awakening in a child with autism and developmental delays. *Behavioral Interventions, 19*(2), 111–119. https://doi.org/10.1002/bin.154

Deliens, G., & Peigneux, P. (2019). Sleep-behaviour relationship in children with autism spectrum disorder: Methodological pitfalls and insights from cognition and sensory processing. *Developmental Medicine and Child Neurology, 61*(12), 1368–1376. https://doi.org/10.1111/dmcn.14235

Didden, R., & Sigafoos, J. (2001). A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. *Research in Developmental Disabilities, 22*(4), 255–272. https://doi.org/10.1016/s0891-4222(01)00071-3

Durand, V. M. (2002). Treating sleep terrors in children with autism. *Journal of Positive Behavior Interventions, 4*(2), 66–72. https://doi.org/10.1177/109830070200400201

Durand, V. M., & Christodulu, K. V. (2004). Description of a sleep-restriction program to reduce bedtime disturbances and night waking. *Journal of Positive Behavior Interventions, 6*(2), 83–91. https://doi.org/10.1177/10983007040060020301

Foody, C., James, J. E., & Leader, G. (2015). Parenting stress, salivary biomarkers, and ambulatory blood pressure: A comparison between mothers and fathers of children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 45*(4), 1084–1095. https://doi.org/10.1007/s10803-014-2263-y

France, K., & Blampied, N. (1999). Infant sleep disturbance: Description of a problem behaviour process. *Sleep Medicine Reviews, 3*(4), 265–280. https://doi.org/10.1053/smrv.1999.0071

Fritz, C. O., Morris, P. E., & Richler, J. J. (2012). Effect size estimates: Current use, calculations, and interpretation. *Journal of Experimental Psychology: General, 141*(1), 2–18. https://doi.org/10.1037/a0024338

Glickman, G. (2010). Circadian rhythms and sleep in children with autism. *Neuroscience and Biobehavioral Review, 34*(5), 755–768. https://doi.org/10.1016/j.neubiorev.2009.11.017

Goldman, S. E., McGrew, S., Johnson, K. P., Richdale, A. L., Clemons, T., & Malow, B. A. (2011). Sleep is associated with problem behaviors in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders, 5*(3), 1223–1229. https://doi.org/10.1016/j.rasd.2011.01.010

Goldman, S. E., Richdale, A. L., Clemons, T., & Malow, B. A. (2012). Parental sleep concerns in autism spectrum disorders: Variations from childhood to adolescence. *Journal of Autism and Developmental Disorders, 42*(4), 531–538. https://doi.org/10.1007/s10803-011-1270-5

Gradisar, M., Jackson, K., Spurrier, N. J., Gibson, J., Whitham, J., Williams, A. S., Dolby, R., & Kennaway, D. J. (2016). Behavioral interventions for infant sleep problems: A randomized controlled trial. *Pediatrics, 137*(6), e20151486. https://doi.org/10.1542/peds.2015-1486

Gringras, P., Nir, T., Breddy, J., Frydman-Marom, A., & Findling, R. L. (2017). Efficacy and safety of pediatric prolonged-release melatonin for insomnia in children with autism spectrum disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(11), 948–957. https://doi.org/10.1016/j.jaac.2017.09.414

Heussler, H., & Malow, B. A. (2020). Sleep disturbances in autism spectrum disorder: Contributions of neurological conditions (and their medications). In A. L. Richdale & K. P. Johnson (Eds.), *Sleep difficulties and disorders in autism spectrum disorder* (pp. 61–71). Information Age Publishing.

Hodge, D., Carollo, T. M., Lewin, M., Hoffman, C. D., & Sweeney, D. P. (2014). Sleep patterns in children with and without autism spectrum disorders: Developmental comparisons. *Research in Developmental Disabilities, 35*(7), 1631–1638. https://doi.org/10.1016/j.ridd.2014.03.037

Hodge, D., Hoffman, C. D., Sweeney, D. P., & Riggs, M. L. (2013). Relationship between children's sleep and mental health in mothers of children with and without autism. *Journal of Autism and Developmental Disorders, 43*(4), 956–963. https://doi.org/10.1007/s10803-012-1639-0

Hoffman, C. D., Sweeney, D. P., Lopez-Wagner, M. C., Hodge, D., Nam, C. Y., & Botts, B. H. (2008). Children with autism: Sleep problems and mothers' stress. *Focus on Autism and Other Developmental Disabilities, 23*(3), 155–165. https://doi.org/10.1177/1088357608316271

Hollway, J. A., Aman, M. G., & Butter, E. (2013). Correlates and risk markers for sleep disturbance in participants of the autism treatment network. *Journal of Autism and Developmental Disorders, 43*(12), 2830–2843. https://doi.org/10.1007/s10803-013-1830-y

Jin, C. S., Hanley, G. P., & Beaulieu, L. (2013). An individualized and comprehensive approach to treating sleep problems in young children. *Journal of Applied Behavior Analysis, 46*(1), 161–180. https://doi.org/10.1002/jaba.16

Johnston, J. M., & Sherman, R. A. (1993). Applying the least restrictive alternative principle to treatment decisions: A legal and behavioral analysis. *The Behavior Analyst, 16*(1), 103–115. https://doi.org/10.1007/BF03392615

Johnson, K. P., & Malow, B. A. (2008). Sleep in children with autism spectrum disorders. *Current Treatment Options in Neurology, 10*(5), 350–359. https://doi.org/10.1007/s11940-008-0038-5

Kline, R.B. (2013). Beyond significance testing: Statistics reform in the behavioral sciences (2nd ed). American Psychological Association. https://doi.org/10.1037/14136-000

Kotagal, S., & Broomall, E. (2012). Sleep in children with autism spectrum disorder. *Pediatric Neurology, 47*(4), 242–251. https://doi.org/10.1016/j.pediatrneurol.2012.05.007

Kratochwill, T. R., Hitchcock, J., Horner, R. H., Levin, J. R., Odom, S. L., Rindskopf, D. M., & Shadish, W. R. (2010). Single-case designs technical documentation. *What Works Clearinghouse*. Retrieved from http://ies.ed.gov/ncee/wwc/pdf/wwc_scd.pdf

Lakens, D. (2013). Calculating and reporting effect sizes to facilitate cumulative science: A practical primer for t-tests and ANOVAs. *Frontiers in Psychology, 4*, 1–12. https://doi.org/10.3389/fpsyg.2013.00863

Lerman, D. C., & Iwata, B. A. (1995). Prevalence of the extinction burst and its attenuation during treatment. *Journal of Applied Behavior Analysis, 28*(1), 93–94. https://doi.org/10.1901/jaba.1995.28-93

Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P. A., Clarke, M., Devereaux, P. J., Kleijnen, J., & Moher, D. (2009). The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: Explanation and elaboration. *PLoS Medicine, 6*(7), e1000100. https://doi.org/10.1371/journal.pmed.1000100

Liu, X., Hubbard, J. A., Fabes, R. A., & Adam, J. B. (2006). Sleep disturbances and correlates of children with autism spectrum disorders. *Child Psychiatry and Human Development, 37*(2), 179–191. https://doi.org/10.1007/s10578-006-0028-3

Lozoff, B., Wolf, A. W., & Davis, N. S. (1985). Sleep problems seen in pediatric practice. *Pediatrics, 75*(3), 477–483.

Luiselli, J. K. (2021). Applied behavior analysis measurement, assessment, and treatment of sleep and sleep-related problems. *Journal of Applied Behavior Analysis, 54*(2), 654–667. https://doi.org/10.1002/jaba.774

Luiselli, J. K., Harper, J. M., Murphy, K. J., Leach, M., & Luke, K. (2021). Case report of faded bedtime intervention on delayed sleep-onset and sleep duration in an adolescent with autism spectrum disorder. *Child and Family Behavior Therapy, 43*(2), 114–122. https://doi.org/10.1080/07317107.2021.1894712

Luiselli, J. K., Harper, J. M., Shlesinger, A., Murphy, K. J., & Luke, K. (2020). Faded bedtime intervention for delayed sleep onset in an adolescent with autism spectrum disorder. *Clinical Case Studies, 19*(3), 180–188. https://doi.org/10.1177/1534650119897964

Malow, B., Adkins, K. W., McGrew, S. G., Wang, L., Goldman, S. E., Fawkes, D., & Burnette, C. (2012). Melatonin for sleep in children with autism: A controlled trial examining dose, tolerability, and outcomes. *Journal of Autism and Developmental Disorders, 42*(8), 1729–1737. https://doi.org/10.1007/s10803-011-1418-3

Malow, B. A., MacDonald, L. L., Fawkes, D. B., Alder, M. L., & Katz, T. (2016). Teaching children with autism spectrum disorder how to sleep better: A pilot educational program for parents. *Clinical Practice in Pediatric Psychology, 4*(2), 125–136. https://doi.org/10.1037/cpp0000138

Malow, B. A., Marzec, M. L., McGrew, S. G., Wang, L., Henderson, L. M., & Stone, W. L. (2006). Characterizing sleep in children with autism spectrum disorders: A multidimensional approach. *Sleep, 29*(12), 1563–1571. https://doi.org/10.1093/sleep/29.12.1563

McLay, L., France, K., Blampied, N., Danna, K., & Hunter, J. (2017). Using functional behavioral assessment to develop a multicomponent treatment for sleep problems in a 3-year-old boy with autism. *Clinical Case Studies, 16*(3), 254–270. https://doi.org/10.1177/1534650116688558

McLay, L., France, K., Blampied, N., van Deurs, J., Hunter, J., Knight, J., Hastie, B., Carnett, A., Woodford, E., Gibbs, R., & Lang, R. (2021). Function-based behavioral interventions for sleep problems in children and adolescents with autism: Summary of 41 clinical cases. *Journal of Autism and Developmental Disorders, 51,* 418–432. https://doi.org/10.1007/s10803-020-04548-7

McLay, L. K., France, K. G., Knight, J., Blampied, N. M., & Hastie, B. (2019). The effectiveness of function-based interventions to treat sleep problems, including unwanted co-sleeping, in children with autism. *Behavioral Interventions, 34*(1), 30–51. https://doi.org/10.1002/bin.1651

McLay, L., Sutherland, D., Machalicek, W., & Sigafoos, J. (2020). Systematic review of telehealth interventions for the treatment of sleep problems in children and adolescents. *Journal of Behavioral Education, 29*(2), 222–245. https://doi.org/10.1007/s10864-020-09364-8

Melke, J., Goubran Botros, H., Chaste, P., Betancur, C., Nygren, G., Anckarsater, H., Rastam, M., Stahlberg, O., Gillberg, I. C., Delorme, R., Chabane, N., Mouren-Simeoni, M.-C., Fauchereau, F., Durand, C. M., Chevalier, F., Drouot, X., Collet, C., Launay, J.-M., Leboyer, M., … Bourgeron, T. (2008). Abnormal melatonin synthesis in autism spectrum disorders. *Molecular Psychiatry, 13*(1), 90–98. https://doi.org/10.1038/sj.mp.4002016

Meltzer, L. J. (2010). Clinical management of behavioral insomnia of childhood: Treatment of bedtime problems and night wakings in young children. *Behavioral Sleep Medicine, 8*(3), 172–189. https://doi.org/10.1080/15402002.2010.487464

Meltzer, L. J., & Mindell, J. A. (2007). Relationship between child sleep disturbances and maternal sleep, mood, and parenting stress: A pilot study. *Journal of Family Psychology, 21*(1), 67–73. https://doi.org/10.1037/0893-3200.21.1.67

Meltzer, L. J., & Thomas, J. H. (2020). Sleep disturbance in children with autism spectrum disorder: Impact on family functioning and quality of life. In A. L. Richdale & K. P. Johnson (Eds.), *Sleep difficulties and disorders in autism spectrum disorder* (pp. 109–125). Information Age Publishing.

Michael, J. (1982). Distinguishing between discriminative and motivational functions of stimuli. *Journal of the Experimental Analysis of Behavior, 37*(1), 149–155. https://doi.org/10.1901/jeab.1982.37-149

Mindell, J. A., Telofski, L. S., Wiegand, B., & Kurtz, E. S. (2009). A nightly bedtime routine: Impact on sleep in young children and maternal mood. *Sleep, 32*(5), 599–606. https://doi.org/10.1093/sleep/32.5.599

Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *BMJ, 339*, b2535. https://doi.org/10.1136/bmj.b2535

Morgenthaler, T. I., Owens, J., Alessi, C., Boehlecke, B., Brown, T. M., Coleman, J., Friedman, L., Kapur, V. K., Lee-Chiong, T., Pancer, J., & Swick, T. J. (2006). Practice parameters for behavioural treatment of bedtime problems and night wakings in infants and young children. *Sleep, 29*(10), 1277–1281. https://doi.org/10.1093/sleep/29.10.1277

Owens, J. A., Spirito, A., McGuinn, M., & Nobile, C. (2000). Sleep habits and sleep disturbance in elementary school-aged children. *Journal of Developmental and Behavioral Pediatrics, 21*(1), 27–36. https://doi.org/10.1097/00004703-200002000-00005

Owens, L. J., France, K. G., & Wiggs, L. (1999). Behavioural and cognitive-behavioural interventions for sleep disorders in infants

and children: A review. *Sleep Medicine Reviews, 3*(4), 281–302. https://doi.org/10.1053/smrv.1999.0082

Piazza, C. C., Fisher, W. W., & Sherer, M. (1997). Treatment of multiple sleep problems in children with developmental disabilities: Faded bedtime with response cost versus bedtime scheduling. *Developmental Medicine and Child Neurology, 39*(6), 414–418. https://doi.org/10.1111/j.1469-8749.1997.tb07456.x

Piazza, C. C., Hagopian, L. P., Hughes, C. R., & Fisher, W. W. (1998). Using chronotherapy to treat severe sleep problems: A case study. *American Journal of Mental Retardation, 102*(4), 358–366. https://doi.org/10.1352/0895-8017(1998)102%3c0358:ucttss%3e2.0.co;2

Price, A. M., Wake, M., Ukoumunne, O. C., & Hiscock, H. (2012). Five-year follow-up of harms and benefits of behavioral infant sleep intervention: Randomized trial. *Pediatrics, 130*(4), 643–651. https://doi.org/10.1542/peds.2011-3467

Ranganthan, P., Pramesh, C. S., & Buysse, M. (2015). Common pitfalls in statistical analysis: Clinical vs statistical significance. *Perspectives in Clinical Research, 6*(3), 169–170. https://doi.org/10.4103/2229-3485.159943

Reichow, B., Doehring, P., Cicchetti, D. V., & Volkmar, F. R. (Eds.). (2010). *Evidence-based practices and treatments for children with autism*. Springer.

Reichow, B., Volkmar, F. R., & Cicchetti, D. V. (2008). Development of the evaluative method for evaluating and determining evidence-based practices in autism. *Journal of Autism and Developmental Disorders, 38*(7), 1311–1319. https://doi.org/10.1007/s10803-007-0517-7

Reynolds, A., & Katz, T. (2020). Medical issues affecting sleep in children with autism spectrum disorder. In A. L. Richdale & K. P. Johnson (Eds.), *Sleep difficulties and disorders in autism spectrum disorder* (pp. 51–59). Information Age Publishing.

Richdale, A. L., & Schreck, K. A. (2009). Sleep problems in autism spectrum disorders: Prevalence, nature, & possible biopsychosocial aetiologies. *Sleep Medicine Reviews, 13*(6), 403–411. https://doi.org/10.1016/j.smrv.2009.02.003

Rigney, G., Ali, N. S., Corkum, P. V., Brown, C. A., Constantin, E., Godbout, R., Hanlon-Dearman, A., Ipsiroglu, O., Reid, G. J., Shea, S., Smith, I. M., Van der Loos, H. F. M., & Weiss, S. K. (2018). A systematic review to explore the feasibility of a behavioural sleep intervention for insomnia in children with neurodevelopmental disorders: A transdiagnostic approach. *Sleep Medicine Reviews, 41*, 244–254. https://doi.org/10.1016/j.smrv.2018.03.008

Rossignol, D. A., & Frye, R. E. (2011). Melatonin in autism spectrum disorders: A systematic review and meta-analysis. *Developmental Medicine and Child Neurology, 53*(9), 783–792. https://doi.org/10.1111/j.1469-8749.2011.03980.x

Sanders, M. R., Kirby, J. N., Tellegen, C. L., & Day, J. J. (2014). The triple P-positive parenting program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clinical Psychology Review, 34*(4), 337–357. https://doi.org/10.1016/j.cpr.2014.04.003

Schreck, K. A., Mulick, J. A., & Smith, A. F. (2004). Sleep problems as possible predictors of intensified symptoms of autism. *Research in Developmental Disabilities, 25*(1), 57–66. https://doi.org/10.1016/j.ridd.2003.04.007

Schreck, K. A., & Schreck, R. E. (2020). The unique association of autism spectrum disorder with sleep problems. In A. L. Richdale & K. P. Johnson (Eds.), *Sleep difficulties and disorders in autism spectrum disorder* (pp. 3–28). Information Age Publishing.

Stein, M. A. (1999). Unravelling sleep problems in treated and untreated children with ADHD. *Journal of Child and Adolescent Psychopharmacology, 9*(3), 157–168. https://doi.org/10.1089/cap.1999.9.157

Sung, V., Hiscock, H., Sciberras, E., & Efron, D. (2008). Sleep problems in children with attention-deficit/hyperactivity disorder: Prevalence and the effect on the child and family. *Archives of Pediatrics and Adolescent Medicine, 162*(4), 336–342. https://doi.org/10.1001/archpedi.162.4.336

van Deurs, J. R., McLay, L. K., France, K. G., & Blampied, N. M. (2020). Sequential implementation of functional behavior assessment-informed treatment components for sleep disturbance in autism: A case study. *Behavioral Sleep Medicine, 19*(3), 333–351. https://doi.org/10.1080/15402002.2020.1758701

van Deurs, J. R., McLay, L. K., France, K. G., Blampied, N. M., Lang, R. B., & Hunter, J. E. (2019). Behavioral sleep intervention for adolescents with autism spectrum disorder: A pilot study. *Advances in Neurodevelopmental Disorders, 3*(4), 397–410. https://doi.org/10.1007/s41252-019-00123-z

van Deurs, J. R., France, K. G., McLay, L. K., & Blampied, N. M. (2021). Cognitive-behavioral treatment of sleep disturbance in children and adolescents with autism: Eight case studies using functional behavior assessment. Research in Autism Spectrum Disorders, 86, 101823. https://doi.org/10.1016/j.rasd.2021.101823

Vannest, K. J., & Ninci, J. (2015). Evaluating intervention effects in single-case research designs. *Journal of Counseling & Development, 93*(4), 403–411. https://doi.org/10.1002/jcad.12038

Vriend, J. L., Corkum, P. V., Moon, E. C., & Smith, I. M. (2011). Behavioral interventions for sleep problems in children with autism spectrum disorders: Current findings and future directions. *Journal of Pediatric Psychology, 36*(9), 1017–1029. https://doi.org/10.1093/jpepsy/jsr044

Wasserstein, R. L., & Lazar, N. A. (2016). The ASA statement on p-values: Context, process, and purpose. *The American Statistician, 70*(2), 129–133. https://doi.org/10.1080/00031305.2016.1154108

Weiskop, S., Matthews, J., & Richdale, A. (2001). Treatment of sleep problems in a 5-year-old boy with autism using behavioural principles. *Autism, 5*(2), 209–221. https://doi.org/10.1177/1362361301005002009

Weiskop, S., Richdale, A., & Matthews, J. (2005). Behavioural treatment to reduce sleep problems in children with autism or fragile X syndrome. *Developmental Medicine and Child Neurology, 47*(2), 94–104. https://doi.org/10.1017/S0012162205000186

Wilkinson, L. (1999). Statistical methods in psychology journals: Guidelines and explanations. *American Psychologist, 54*(8), 594–604. https://doi.org/10.1037/0003-066X.54.8.594

Williams, P. G., Sears, L. L., & Allard, A. (2004). Sleep problems in children with autism. *Journal of Sleep Research, 13*(3), 265–268. https://doi.org/10.1111/j.1365-2869.2004.00405.x

Wright, B., Sims, D., Smart, S., Alwazeer, A., Alderson-Day, B., Allgar, V., Whitton, C., Tomlinson, H., Bennett, S., Jardine, J., McCaffrey, N., Leyland, C., Jakeman, C., & Miles, J. (2011). Melatonin versus placebo in children with autism spectrum conditions and severe sleep problems not amenable to behaviour management strategies: A randomised controlled crossover trial. *Journal of Autism and Developmental Disorders, 41*(2), 175–184. https://doi.org/10.1007/s10803-010-1036-5

Appendix B: Parent Consent Form

An investigation into the effectiveness of treatments for slee children with autism

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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 C: Young Person Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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

Young Person Consent Form

My name is _____.

- ☐ XXX has told me about the work she is going to be doing with me and my parent/s.
- ☐ XXX told me she is going to be working with me and my parent/s to help me learn to sleep better.
- ☐ I understand that while XXX works with me she will be asking me and 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, or I can delete it myself.
- ☐ I understand that no-one outside the study and my family will know any information about me unless staff on the project are worried about my or anyone's safety. Information about my sleep will be published but no-one will know my name.
- ☐ I know if I want to stop at any time or if I do not want to be a part of this project anymore that is fine. I can tell XXX or my parents.
- ☐ I would like a summary of the results of this project.

Date: _____

Young Person's Signature: _____

Please return this form to XXX.

Appendix D: Children's Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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

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 E: Children's Assent Form

Children's Assent Form

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

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

OR

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

My name: _____

You can give this form back to XX now.

Appendix F: Audiovisual Recording Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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

AUDIOVISUAL RECORDING CONSENT FORM

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

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

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

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

Signed: _____

Date: _____

Appendix G: Video/actigraph Recording Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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

VIDEO/ ACTIGRAPH RECORDING CONSENT FORM

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

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

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

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

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

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

You may still eligible to participate in the research study, should you refuse to allow video recordings to be made

Only people involved in the project can view the recordings.

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

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

Signed:

Date:

Appendix H: Information Sheet for Parents

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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.

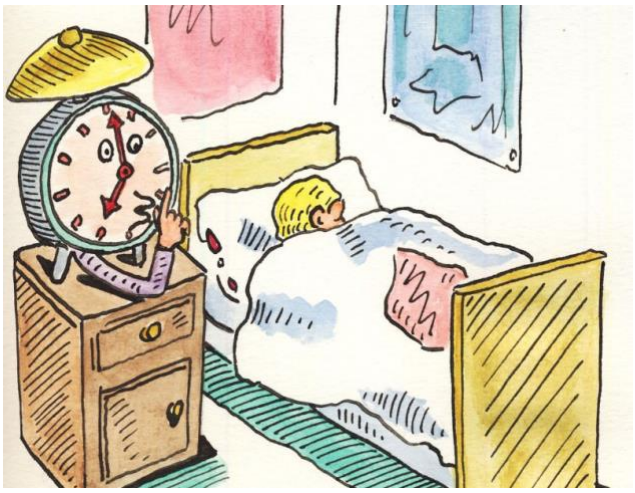
Should you require any additional information about the study or if you would like to access the study findings you are able to do so at any stage. The data which is produced from the research will be kept in a locked cabinet at the University of Canterbury for a minimum of ten years.

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: laurie.mclay@canterbury.ac.nz

Appendix I: Young Person Information Sheet



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

Young Person Information Sheet

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

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

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

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

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

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

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

Appendix J: Children's Information Sheet

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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.

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.

Appendix K: Parent-report Sleep Diary

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

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday

1st Night time awakening	Time & Duration of awakening	_____ mins	_____mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
		Monday	**Tuesday**	**Wednesday**	**Thursday**	**Friday**	**Saturday**	**Sunday**
2nd Night time awakening	Time & Duration of awakening	_____ mins	_____mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
3rd Night time awakening	Time & Duration of awakening	_____ mins	___mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
Time awake in the morning								

- Curtain calls: Any 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 L: Alternative Parent-report Sleep Diary

	AM																								PM																							
	00		01		02		03		04		05		06		07		08		09		10		11		12		13		14		15		16		17		18		19		20		21		22		23	
Date:																																																
Comments																																																
Date:																																																
Comments																																																
Date:																																																
Comments																																																
Date:																																																
Comments																																																
Date:																																																
Comments																																																
Date:																																																
Comments																																																

Date:																																																
Comments																																																

Record a code for the behaviours that were observed when your child was put to bed and while awake. Also record a code for your response to these behaviours.
Behaviour codes:
Your child *– Moving arms & legs (M) Smiling (S) Roll towards parents (R); Crying (C)*
Your Response – *Breastfed (B); Ignored (I); Lay with them (L)*

If behaviour isn't covered by one of these codes, then please write down the behaviour that was observe

Appendix M: Self-report Sleep Diary

Sleep Diary	Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
Date:							
What time did you go to bed last night?							
How long did it take you to fall asleep (in minutes)?							
Did you fall asleep: 1. Easily 2. After some time 3. With difficulty							
How many times did you wake in the night?							
What time did you wake up in the night?							
How long were you awake during the night?							
How long did you sleep in total?							
What disturbed your sleep (e.g. noise, lights, comfort, etc)?							
How did you feel this morning: Refreshed OK Tired							
What time did you wake up in the morning?							

Appendix N: Example Post-treatment Interview

Post Treatment Feedback Discussion

1.) Tell me about the intervention?

2.) How did you find the intervention process over all? (What were the strengths/positives, challenges).

3.) What is it you did, that you feel made a difference?

4.) Anything more – anything you think that didn't really make a difference?

5.) Comparing life before intervention to life now, what's changed for you and the family?

6.) Any suggestions for how the process could be improved?

7.) Any final words/comments