AN INVESTIGATION OF STEREOTYPY IN RELATION TO SLEEP DISTURBANCE IN CHILDREN ON THE AUTISM SPECTRUM: EFFECTIVENESS OF FUNCTIONAL BEHAVIOUR ASSESSMENT-BASED INTERVENTIONS

Thesis submitted in fulfilment of

the requirements for the degree of Doctor of Philosophy in Psychology

by Jolene Hunter

University of Canterbury

2022

Acknowledgements

For Theodore, who came along in the midst of this journey and made it all the more worthwhile

To my supervisors Laurie, Karyn and Neville, I am extremely grateful for your invaluable expertise, guidance and support throughout many years working together. I have learned so much from each of you. I am truly fortunate to have had such a wonderful supervisory team.

A special thank you to my family and friends for always being encouraging of my aspirations, and for your belief in my ability to realise my goals. My deepest gratitude to my partner Mike for your unwavering and unconditional support that made all of this achievable, particularly after we became parents.

Thank you to the wider Autism and Sleep Team and my fellow PhD students for your encouragement and camaraderie over the years. Finally, yet importantly, my sincere appreciation to the participant families whose contributions made this research possible.

Abstract

Sleep is essential to our overall functioning and wellbeing. Children on the autism spectrum experience high rates of sleep problems that can be both chronic and severe, and which are associated with a wide range of detrimental effects for children and their families. Sleep problems can intensify the core symptoms of autism, including restricted and repetitive behaviours commonly known as *stereotypy*. The aetiology of sleep disturbance in autism is multifactorial, involving biological, psychological, social, medical, and environmental factors. Notwithstanding these determinants, sleep problems often have a behavioural basis. Functional behaviour assessment (FBA) is an evidence-based approach to identifying the environmental and behavioural factors underlying sleep problems for an individual, and to guiding the selection of behavioural sleep interventions (BSI).

Children on the autism spectrum may engage in stereotypy at night, which may contribute to sleep problems by delaying sleep onset or the re-initiation of sleep following night wakings (NWs). It is generally unclear, however, whether established BSI techniques can effectively treat sleep-related stereotypy, particularly when it is automatically maintained, or which types of strategies may be required to treat such behaviour. Further, little is known about the characteristics of sleep-related stereotypic behaviours, nor its role in relation to sleep, including the extent to which stereotypy contributes to or is a co-occurring feature of sleep disturbance. Finally, there is lack of research investigating whether improving sleep problems may result in collateral benefits for children and parents, including children's daytime stereotypy and wider symptoms of autism.

The primary objectives of the present research were three-fold: (1) to investigate the effectiveness of function-based BSI to treat stereotypy in the context of sleep problems in children on the autism spectrum, and to examine the maintenance of treatment effects and

acceptability to parents of the selected treatments; (2) to investigate whether and how stereotypy may contribute to sleep disturbance, and the characteristics (e.g., type, topography, function) of sleep-related stereotypy across individuals; and (3) to examine whether treating sleep problems produces collateral benefits for children and parents, including change in children's daytime stereotypy. Three empirical studies in this thesis used single-case experimental (multiple baseline or AB) design to evaluate the utility of function-based BSI to treat sleep problems including sleep onset delay (SOD) and NWs in 12 children (5 girls and 7 boys, aged 3-10 years) with formal diagnoses of autism spectrum disorder. In 10/12 children, sleep-related stereotypy occurred during SOD and/or NWs and appeared to interfere with the initiation and/or re-initiation of sleep.

A fourth, qualitative study thematically analysed the clinical assessment reports for 15 children on the autism spectrum (aged 3-15 years; including the children in Studies 1-3) to further examine the potential role of stereotypy within sleep problems, and characteristics of children's behaviour. A systematic review of the research literature was conducted to investigate the types of collateral effects that may occur following a BSI for children on the autism spectrum. Psychometric measures administered pre- and post-treatment in Studies 1-3 of this thesis were used to assess whether the BSI produced change in core symptoms of autism and internalising and externalising symptoms in participant children, as well as in parents' sleep quality, wellbeing, and relationship quality, with data examined using modified Brinley plots.

Collectively, results demonstrate the preliminary effectiveness, durability, feasibility, and social validity of a variety of function-based BSI, including preliminary evidence for strategies that may effectively reduce automatically maintained sleep-related stereotypy. Tentative support was found for procedures that manipulate the motivating operations for stereotypy and sleep, including sleep restriction. Findings demonstrate that a heterogeneous

range of behaviours including motor and vocal stereotypy and repetitive manipulation of objects can accompany sleep problems in children on the autism spectrum and may contribute to sleep disturbance by delaying the initiation of sleep, and/or re-initiation of sleep following NWs.

Results also showed that stereotypy may differentially affect sleep across and within children, which may have differing implications for behavioural treatment. Finally, an evaluation of the collateral effects of BSI in the research literature and in participant children and parents in Studies 1-3 indicate that BSI may produce a range of untargeted, beneficial effects including a reduction in children's daytime stereotypy, however, collateral effect results were mixed and inconsistent. Overall, findings emphasise the importance of utilising FBA to inform BSI for children on the autism spectrum, which may necessarily include strategies that specifically target automatically maintained behaviour. Future research is needed to better understand how sleep problems and core symptoms of autism interact across and within children, and address how these relationships are treated in the sleep context.

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Hunter, J. E., McLay, L. K., France, K. G., & Blampied, N. M. (forthcoming). The assessment and treatment of stereotypy in the sleep context. In L. K. McLay, K. G. France, & N. M. Blampied (Eds.), *Clinical handbook of behavioral sleep treatment for children on the autism spectrum.* Christchurch, NZ: Springer International

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L McLay, K France and N Blampied: Assisted with the development and conceptualisation of the chapter, edited the manuscript

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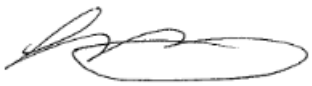
Name: *Laurie McLay* Signature:  Date: *09/02/2022*

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Chapter 6, Study 2, pp. 173-196.

A brief report based on this study has been published in the journal Sleep Medicine:
Hunter, J. E., McLay, L. K., France, K. G., & Blampied, N. M. (2021). Sleep and stereotypy in children with autism: Effectiveness of function-based behavioral treatment. *Sleep Medicine, 80,* 301-304. https://doi.org/10.1016/j.sleep.2021.01.062

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Developed the study, assessed participant eligibility through screening, carried out data collection, conducted the assessment, analysed FBA footage (e.g., video footage), scored and interpreted all psychometric measures, designed the intervention (under the clinical supervision of L McLay and K France), implemented the intervention, created resources (e.g., social stories), conducted all data analyses (e.g., IOA), wrote all sections of the article, created each table, edited the article including all formatting and referencing

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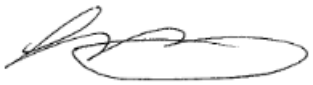
Name: *Laurie McLay* Signature:  Date: *09/02/2022*

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Chapter 8, Study 4, pp. 226-248 and 'James' case information within Chapter 7, Study 3, pp.197-225.

An article based on the thematic analysis in Chapter 8 and James' case study in Chapter 7 has been published in Advances in Neurodevelopmental Disorders:
Hunter, J. E., McLay, L. K., France, K. G., Swit, C. S., & Blampied, N. M. (2022). Parent perceptions of sleep-related stereotypy within sleep problems in children on the autism spectrum: Implications for behavioral treatment. *Advances in Neurodevelopmental Disorders,* https://doi.org/10.1007/s41252-022-00246-w

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sections of the article, edited the article including all formatting and referencing. James: assessed participant eligibility through screening, conducted the assessment, carried out data collection, analysed FBA footage (e.g., video footage), scored and interpreted all psychometric measures, designed the intervention (under the clinical supervision of L McLay and K France), implemented the intervention, created resources (e.g., social stories), conducted all data analyses (e.g., visual analysis, IOA), interpreted the results, wrote all sections of the article, created the table, edited the article including all formatting and referencing

L McLay: Assisted with the development and conceptualisation of the studies, provided clinical supervision, provided validation for the thematic analysis, edited the manuscript

K France: Assisted with the development and conceptualisation of the studies, provided clinical supervision, edited the manuscript

C Swit: Assisted with the development and conceptualisation of the qualitative study, provided validation for the thematic analysis, edited the manuscript

N Blampied: Assisted with the development and conceptualisation of the studies, assisted with the selection and interpretation of quantitative data analysis, constructed the figure (James), edited the manuscript

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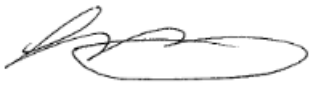
Name: *Laurie McLay* Signature:  Date: *09/02/2022*

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Chapter 9, pp. 249-279.

This systematic review has been published in Research in Autism Spectrum Disorders:
Hunter, J. E., McLay, L. K., France, K. G., & Blampied, N. M. (2020). Systematic review of the collateral effects of behavioral sleep interventions in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders, 79,* 101677. https://doi.org/10.1016/j.rasd.2020.101677

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L McLay, K France and N Blampied: Assisted with the development and conceptualisation of the article, edited the manuscript

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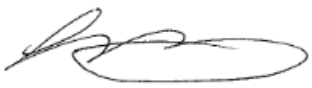
Name: *Laurie McLay* Signature:		Date: *09/02/2022*

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List of Abbreviations

Abolishing operation	AO
Applied behaviour analysis	ABA
American Psychiatric Association	APA
Attention-deficit/hyperactivity disorder	ADHD
Autism Spectrum Disorder	ASD
Behavioural sleep interventions	BSI
Centers for Disease Control and Prevention	CDC
Child Behaviour Checklist	CBCL
Children's Sleep Habits Questionnaire	CSHQ
Curtain Calls	CCs
Depression Anxiety Stress Scales – 21	DASS-21
Developmental disability/disabilities	DD
Diagnostic and Statistical Manual of Mental Disorders	DSM
Differential reinforcement of other behaviour	DRO
Discriminative stimulus	S^D
Early morning wakes	EMW
Establishing operation	EO
Functional behaviour assessment	FBA
Gilliam Autism Rating Scale - Third Edition	GARS-3
Inhibitory stimulus control procedures	ISCPs
Insistence on sameness	IS
Intellectual disability	ID
Interobserver agreement	IOA

Long-term follow-up	LTFU
Motivating Operations	MOs
New Zealand	NZ
Night wakings	NWs
Noncontingent reinforcement	NCR
Non-rapid eye movement	NREM
Non-verbal intellectual quotient	NVIQ
Percentage below the median	PBM
Periodic limb movement disorder	PLMD
Periodic limb movements in sleep	PLMS
Pittsburgh Sleep Quality Index	PSQI
Positive practice overcorrection	PPOC
Rapid eye movement	REM
Rare genetic neurodevelopmental disorder	RGND
Relationship Quality Index	RQI
Repetitive Behavior Scale - Revised	RBS-R
Repetitive manipulation of objects	RMO
Repetitive sensory motor	RSM
Response interruption and redirection	RIRD
Restless leg syndrome	RLS
Rhythmic movement (disorder)	RM(D)
Short-term follow-up	STFU
Sleep Assessment and Treatment Tool	SATT
Sleep onset delay	SOD
Sleep onset latency	SOL

Television	TV
Total sleep time	TST
Treatment Acceptability Rating Form – Revised	TARF-R
Typically developing	TD
Videosomnography	VSG
Vineland Adaptive Behavior Scales, Second Edition	VABS-II

Chapter 1

Introduction

Thesis Overview

This thesis began as an investigation into the use of functional behaviour assessment (FBA) to inform intervention for sleep disturbance in children on the autism spectrum but ended as an investigation of stereotypy in relation to sleep disturbance in children on the autism spectrum. This research went through four distinct phases. First, the pilot study (Study 1, Chapter 4) primarily examined whether function-based behavioural interventions can effectively reduce sleep problems in six children on the autism spectrum. The results of this research revealed that stereotypy (explained below in this chapter) accompanied sleep problems in 4/6 children. This raised questions regarding how stereotypy can be treated using behavioural sleep intervention (BSI) techniques, including whether and how treatments for daytime stereotypy might be applied in the sleep context. The second phase of research investigated these specific lines of inquiry in Chapters 5 and 6 (Study 2). Study 2 examines the impact of function-based behavioural interventions on both sleep-related stereotypy and sleep problems in three children on the autism spectrum.

The outcomes of the first and second phases of research highlighted pertinent, unanswered questions regarding the nature of sleep-related stereotypy in children on the autism spectrum, including its potential function in relation to sleep disturbance (e.g., whether stereotypy contributes to, or occurs because of, sleep disturbance, or both) and the types of behaviour that occur across individuals. In the third phase of research, these specific lines of inquiry were investigated (Chapters 7 and 8; studies 3 and 4). Study 3 aimed to elucidate the potential impact of stereotypy on sleep in three children on the autism spectrum by targeting stereotypy in isolation of other sleep variables, using sequential implementation

of function-based treatment strategies. Study 4 is a qualitative study that used thematic analysis to explore parent perceptions of stereotypy, including types of behaviour, its potential impact on sleep, and how parents manage night-time stereotypy, using clinical assessment reports obtained from 21 parents of children on the autism spectrum.

In the final phase of research, the bidirectional nature of the sleep-stereotypy relationship is considered more broadly by investigating whether BSI produce untargeted (collateral) improvement in autism symptom severity, including (daytime) stereotypy. Chapter 9 presents a systematic review of extant research examining the collateral effects of BSI in children on the autism spectrum, and Chapter 10 evaluates the collateral effects of BSI among participant children and parents within this thesis. Overall, embedded within the research process of this thesis was a flexible approach, based on responsivity to research implementation challenges and evolving research questions.

This chapter provides an overview of autism spectrum disorder (ASD) including diagnosis, aetiology, prevalence, and co-occurring challenges, and of sleep disturbance, including prevalence, course and types of sleep problems, aetiology, and consequences of sleep disturbance. Bidirectional influences between autism and sleep problems are then considered. Finally, this chapter discusses a behavioural model of sleep disturbance.

Autism Spectrum Disorder

ASD is an early onset neurodevelopmental disorder, characterised by persistent impairments in social communication and interaction, and the presence of restricted and repetitive patterns of behaviour, across multiple contexts (American Psychiatric Association [APA], 2013). Core symptoms typically manifest as difficulties with social-emotional reciprocity (e.g., difficulty with back-and-forth conversation, reduced sharing of interests or emotions) and nonverbal communication (e.g., lack of eye contact, facial expressions), and

with initiating and maintaining relationships (e.g., difficulty adapting behaviour in accordance with social cues and contexts; APA, 2013). Restricted and repetitive patterns of behaviour, commonly known as *stereotypy*, include repetitive motor movements (e.g., body-rocking, hand-flapping), non-contextual vocalisations (e.g., idiosyncratic phrases, non-word sounds) and repetitive manipulation of objects (RMO; e.g., mouthing or spinning objects), as well as rigid adherence to routines, insistence on sameness, and restricted interests of abnormal focus or intensity (APA, 2013).

In addition to these core characteristics, people on the autism spectrum commonly present with atypical responses (i.e., hyper- or hypo-reactivity) to, or abnormal interest in, sensory aspects of their environment (e.g., indifference to pain, unusual fascination with reflected light; APA, 2013). Although these core difficulties typify autism, there exists huge heterogeneity in the manifestation of symptoms across and within individuals. Further, the degree of impairment can vary substantially across adaptive, cognitive and language functioning (Lombardo et al., 2019; Van Wijngaarden-Cremers et al., 2014). For example, autism can occur with or without intellectual disability (ID), and language ability can vary from fluent speech to no spoken language (APA, 2013).

Diagnosis of Autism Spectrum Disorder

Diagnosis of autism is based entirely on an individual's behaviour (APA, 2013). Since autism was first defined as *autistic disturbances of affective contact* (Kanner, 1943), and given clinical recognition in the Diagnostic and Statistical Manual of Mental Disorders (DSM)-III (APA, 1980), the diagnostic criteria have changed, in line with different iterations of diagnostic systems. The most recent changes to the diagnostic criteria occurred in 2013 with the introduction of the DSM-V. This included the addition of atypical sensory responses, the exclusion of language impairment from core symptomatology, and the collapse of three core domains into two (i.e., social and language impairments were collapsed into a single

measure of difficulties with social communication; APA, 2013; Lombardo et al., 2019). Further, four previously independent pervasive developmental disorders (i.e., autistic disorder, Asperger syndrome, childhood disintegrative disorder and pervasive developmental disorder-not otherwise specified) were subsumed under the single label of *autism* (APA, 2013). This decision came from a lack of reliable and replicable evidence to support distinct diagnostic conditions and reflected a major shift in the conceptualisation of autism, from a categorical to a dimensional approach, where autism is understood as a spectrum of disorders (Lombardo et al., 2019; Volkmar et al., 2014).

The broadened definition of autism has had implications for who is given clinical diagnosis of autism, and therefore who has access to funding and treatment (K. Williams et al., 2014; Lombardo et al., 2019). It has also enhanced the development of targeted and effective interventions that improve functional outcomes and quality of life for children on the autism spectrum (Lombardo et al., 2019).

Aetiology of Autism Spectrum Disorder

The aetiology of autism is not well understood and is a focus of much debate (Bölte et al., 2019; Campisi et al., 2018; Rutter, 2005). Evidence indicates that autism has multiple genetic and environmental causes, as well as complex gene-environment (i.e., epigenetic) interactions (Bölte et al., 2019; Currenti, 2010; Masi et al., 2017); however, an accurate understanding of the causal mechanisms underpinning autism is yet to be achieved (Bölte et al., 2019; Rutter, 2005). Genetic studies have identified hundreds of genes, as well as specific genetic deletions, duplications, and protein-altering mutations, inherited and *de novo,* associated with autism (Bölte et al., 2019; Currenti, 2010; Masi et al., 2017; Rutter, 2005). Family and twin studies also suggest there is a strong hereditary component (Campisi et al., 2018; Rutter, 2005; Sokol & Lahiri, 2011; Volkmar et al., 2014). Genetic pathways implicated in autism vary substantially, from a single protein mutation to cumulative risk

formed from thousands of low-risk alleles (Masi et al., 2017); this means there are multiple gene variants which may converge on an autism phenotype (Sokol & Lahiri, 2011). The complexity and substantial variation in autism-related genes means the concept of heterogeneity in autism applies at both phenotypic and genotypic levels (Bölte et al., 2019; Campisi et al., 2018; Lombardo et al., 2019; Masi et al., 2017; Rutter, 2005).

Notwithstanding genetic contributions, it is generally agreed that the environment also plays a role in the aetiology of autism (Bölte et al., 2019; Campisi et al., 2018; Rutter, 2005), though relatively less is known about non-genetic, and epigenetic, influences (Campisi et al., 2018; Rutter 2005). Extant research suggests that causative effects begin in utero and negatively affect the architecture, maturation and functioning of neuronal systems (Bölte et al., 2019; Currenti, 2010). Prenatal influences under investigation include nutrition (e.g., maternal depletion of essential nutrients such as iron), exposure to toxins (e.g., to heavy metals like mercury), substance use (e.g., smoking and alcohol), medication (e.g., selective serotonin reuptake inhibitors), maternal infection, and extreme psychosocial (e.g., prenatal maternal stress) factors (Bölte et al., 2019; Campisi et al., 2018; Jiang et al., 2016). Research suggests that postnatal influences are less likely than prenatal influences to contribute to autism (Rutter, 2005). Postnatal influences under investigation include immune and neurological disorders (e.g., herpes encephalitis) and immunisations; however, allegations that autism is caused by immunisation such as the measles-mumps-rubella (MMR) vaccination, have been comprehensively refuted (Bölte et al., 2019; Currenti, 2010; Rutter, 2005). Given its continuing mysteries, the aetiology of autism is an area of intense and on-going research focus (Bölte et al., 2019; Rutter, 2005).

Prevalence of Autism

The reported prevalence of autism has increased significantly in the past two decades. This is thought to be the result of the broadened definition of autism (Bölte et al., 2019;

Matson & Kozlowski, 2011), increased public and professional awareness of autism, an increase in favourable policies leading to improved recognition and support for the needs of individuals on the autism spectrum (e.g., inclusive education), and improved accessibility of services (Coughlan et al., 2020; Matson & Kozlowski, 2011; Saracino et al., 2010). As of yet, no research has investigated the exact prevalence of autism in Aotearoa New Zealand (NZ); however, current estimates based on prevalence rates in the United Kingdom suggest 1 in 100 people in NZ are on the spectrum, equating to approximately 50,000 people (Ministries of Health and Education, 2016). In the United States, 1 in 44 children are identified as being on the autism spectrum, according to the Centers for Disease Control and Prevention (CDC) Autism and Developmental Disabilities Monitoring Network (Maenner et al., 2021).

Autism is diagnosed in boys significantly more frequently than girls, with a male-to-female ratio of close to 3:1 (Loomes et al., 2017). An explanation posited for this difference is a diagnostic gender bias, where girls may be less likely to receive a diagnosis of autism owing to diagnostic criteria that aligns with a male autism phenotype. Further, girls may have greater internalising and 'camouflaged' symptoms compared to greater externalising (e.g., hyperactivity, repetitive behaviour) symptoms in boys, meaning symptoms in girls are less frequently brought to clinical attention (Bölte et al., 2019; Lai et al., 2015; Loomes et al., 2017; Masi et al., 2017). It is important for future research to better understand these gender differences, including enhancing diagnostic processes for girls (Loomes et al., 2017).

Co-occurring Challenges in Children on the Autism Spectrum

Children on the autism spectrum experience numerous co-occurring challenges ranging from medical to internalising and externalising problems (Gillberg, 2011; Mannion & Leader, 2013; Masi et al., 2017; Mazurek et al., 2019; Ming et al., 2008). Examples of co-occurring medical conditions include immune system abnormalities, gastrointestinal disorders, mitochondrial dysfunction, allergies, asthma, and seizure disorders (Aldinger et al.,

2015; Mannion & Leader, 2013; Masi et al., 2017; Ming et al., 2008). Common co-occurring internalising and externalising problems include anxiety, depression, attention-deficit/hyperactivity disorder (ADHD), bipolar and mood disorders, eating disorders, substance use disorders, schizophrenia spectrum and other psychotic disorders, obsessive-compulsive and related disorders, aggression, disruption, self-injury and extreme emotional outbursts (Hanley et al., 2014; Hossain et al., 2020; Lindor et al., 2019; Mannion & Leader, 2013; Masi et al., 2017; Matson & Nebel-Schwalm, 2007; Ming et al., 2008). The nature and severity of co-occurring challenges varies substantially across individuals, ranging from few and/or mild-moderate symptoms, to clinically significant concerns that may result in additional diagnoses (Lindor et al., 2019).

It is estimated that 70% of people on the autism spectrum have at least one co-occurring condition, and that close to 40% have two or more (Hossain et al., 2020). Anxiety is the most common co-occurring condition among individuals on the autism spectrum (D. Adams et al., 2019). These co-occurring challenges have a number of negative effects on children's daily functioning and quality of life and add further burden and complexity to the difficulties faced by those affected by autism (Hossain et al., 2020). One of the most detrimental and frequently reported conditions in children on the autism spectrum is sleep disturbance (Elrod & Hood, 2015; Elrod et al., 2016; McLay, France, Blampied, van Deurs, et al., 2021; Ming et al., 2008; Mazurek et al., 2019; P. Williams et al., 2004).

Sleep Disturbance in Children on the Autism Spectrum

Prevalence and Course

Although prevalence estimates vary greatly, sleep disturbance is thought to affect between 40% to 80% of children on the autism spectrum (Ballester et al., 2020; Couturier et al., 2005; Krakowiak et al., 2008; Malow, Marzec, et al., 2006; P. Williams et al., 2004;

Richdale & Schreck, 2009; S. Cohen, Conduit, et al., 2014; Souders et al., 2009). These rates are considerably higher than the 10% to 40% of typically developing (TD) children (Bangerter et al., 2020; Couturier et al., 2005; Fricke-Oerkermann et al., 2007; Krakowiak et al., 2008; Meltzer & Mindell, 2008; Richdale & Schreck, 2009; Souders et al., 2009; Uren et al., 2019) as well as rates reported among children with other types of developmental disabilities (Reynolds & Malow, 2011; Wiggs & Stores, 1996).

Sleep in children on the autism spectrum begins to differ from TD children early in life, with evidence suggesting that sleep duration may be reduced in children on the autism spectrum from as young as 30 months of age (Humphreys et al., 2014). Research suggests that in TD children, sleep problems reduce with time, and for most children do not persist past school-age (Couturier et al., 2005; Hodge et al., 2014; Uren et al., 2019). In comparison, sleep problems in children on the autism spectrum do not tend to remit without treatment and persist into adolescence (Baker et al., 2013; Deserno et al., 2019; Goldman et al., 2012; Hodge et al., 2014; Humphreys et al., 2014; Johnson et al., 2018; Richdale & Schreck, 2009; Roussis et al., 2021; Sivertsen et al., 2012). Research also suggests that for some individuals the types of and/or severity of sleep problems may change over time (Goldman et al., 2011; Mazurek et al., 2019). There are a number of reasons why sleep problems may be more common and persistent in children on the autism spectrum, including biological, psychological, behavioural, social and environmental factors; these are described later in this chapter.

Types of Sleep Problems

Insomnia. The most frequently reported types of sleep problems experienced by children on the autism spectrum are difficulties with sleep onset and maintenance (i.e., insomnia; Cortesi et al., 2010; Loring et al., 2016; Reynolds & Malow, 2011; Richdale & Schreck, 2009; Rigney et al., 2018; Singh & Zimmerman, 2015; Souders et al., 2017). In

particular, children on the autism spectrum experience sleep onset delay (SOD; i.e., prolonged sleep onset latency [SOL]: the duration of time taken to fall asleep once in bed) and frequent and/or prolonged night-wakings (NWs), resulting in reduced total sleep time (TST; i.e., less than an age-appropriate total amount of sleep per night, as per National Sleep Foundation guidelines; Baker & Richdale, 2017; Hirshkowitz et al., 2015; Ohayon et al., 2017; Cortesi et al., 2010; Mazurek et al., 2019; Reynolds & Malow, 2011; Rigney et al., 2018; Roussis et al., 2021; Souders et al., 2017; Vriend et al., 2011; Wiggs & Stores, 2004).

Other sleep and associated problems include early morning wakes (EMW), daytime sleepiness, bedtime resistance, unwanted co-sleeping, and sleep-interfering behaviours such as 'curtain calls' (CCs; i.e., bids for parental attention), screaming, roaming the house, or playing with toys or objects (Cortesi et al., 2010; Cotton & Richdale, 2006; Kirkpatrick, Louw, et al., 2019; Richdale, 2013; Richdale & Schreck, 2009; Roussis et al., 2021; Schreck et al., 2004; Wiggs & Stores, 2004). In many cases, these sleep problems co-exist (Cotton & Richdale, 2006; Liu et al., 2006; Roussis et al., 2021; Spruyt & Curfs, 2015). Research shows that compared to TD children, children on the autism spectrum tend to wake more frequently, for longer durations, and engage in more severe sleep-interfering behaviours (Richdale & Schreck, 2009).

Parasomnias. Parasomnias are a separate class of sleep disturbance, which affect arousal or partial arousal during sleep or the wake-sleep/sleep-wake transition, owing to activation of the central nervous system (Durand, 2002; Schreck & Mulick, 2000). Parasomnias include physiological events and behaviours such as night terrors, nightmares, nocturnal enuresis, sleep walking, sleep talking and bruxism (Durand, 2002; Krakowiak et al., 2008; Schreck & Mulick, 2000; Singh & Zimmerman, 2015). Parasomnias occur most commonly in very young children (Goldman et al., 2012). Although less frequently reported than sleep onset and maintenance problems, research suggests that parasomnias also occur at

high rates in children on the autism spectrum, compared to TD children (Díaz-Román et al., 2018; Goldman et al., 2012; Hodge et al., 2014; Liu et al., 2006; Richdale & Schreck, 2009; Singh & Zimmerman, 2015; Y. Wang et al., 2021). The assessment and treatment of parasomnias in children on the autism spectrum are not a focus of this thesis. As such, parasomnias are not examined further, with the exception of several sleep-related movement disorders (e.g., rhythmic movement disorder) that are discussed in relation to stereotypy in subsequent chapters of this thesis.

Research suggests that sleep problems in children on the autism spectrum may be able to be differentiated by severity into three distinct groups: (a) no sleep problems (i.e., 'good' sleepers); (b) mild-moderate sleep problems, comparable to those in TD children, including bedtime resistance, SOD > 1 hour, and/or NWs of short duration; (c) severe sleep problems, including SOD (i.e., ≥ several hours), NWs, and/or EMW (Hirshkowitz et al., 2015; Patzold et al., 1998; Roussis et al., 2021; Sikora et al., 2012; Souders et al., 2009; Wiggs & Stores, 2004). Studies show that approximately 20-30% of children on the autism spectrum have an absence of sleep difficulties, 50% may have mild-moderate sleep disturbance, and 20-30% experience moderate-severe sleep disturbance (Sikora et al., 2012; Roussis et al., 2021). The consequences of sleep problems for children on the autism spectrum are discussed below.

Aetiology of Sleep Disturbance in Autism

Sleep problems can occur for any child; however, children on the autism spectrum appear to be particularly vulnerable to sleep disturbance (Cortesi et al., 2010). Sleep problems are found to be independent of age, intellectual ability, gender and ethnicity, but are found to increase along with autism severity which suggests that sleep problems may be a part of wider autism symptomatology, and perhaps a function of atypical development (Cortesi et al., 2010; Deliens et al., 2015; Hodge et al., 2014; Johnson et al., 2018; Lindor et al., 2019; Mayes & Calhoun, 2009; Mazzone et al., 2018; Richdale & Schreck, 2009; Singh &

Zimmerman, 2015). The aetiology of sleep disturbance in children on the autism spectrum is unknown, however, it is widely agreed that the causes are multifactorial, involving biological, psychological, behavioural, social and environmental factors, either alone or in combination (Cortesi et al., 2010; Díaz-Román et al., 2018; Hollway et al., 2013; Lindor et al., 2019; Mazzone et al., 2018; Richdale & Schreck, 2009; Reynolds & Malow, 2011; Schreck, 2021; Singh & Zimmerman, 2015; Souders et al., 2017).

Biological Factors

Sleep depends on the central nervous system for its between- and within-cycle regulation (Jan et al., 2008). Children on the autism spectrum may experience sleep difficulties because of central nervous system abnormalities, such as atypical secretion of melatonin and disrupted sleep architecture (Maxwell-Horn & Malow, 2017; Mazzone et al., 2018; P. Williams et al., 2004; Richdale, 1999; Singh & Zimmerman, 2015).

Sleep architecture refers to how sleep patterns are structured in the brain. Sleep is a state during which much neurophysiological activity occurs, but physical activity and responsivity to the environment is heavily reduced (Ballester et al., 2020; Bathory & Tomopoulos, 2017; Jan et al., 2008; Reynolds et al., 2012; Wiggs, 2007). Four sleep states characterise sleep-wake cycles in humans: non-rapid eye movement (NREM) sleep stages 1-3, and rapid eye movement (REM) sleep. From wakefulness, humans transition into sleep through NREM 1 (light sleep), and progress through NREM 2 (initiation of true sleep) and NREM 3 (deep sleep). NREM 3 is the most restorative stage of sleep and is closely linked with sleep quality (Ballester et al., 2020; Bathory & Tomopoulos, 2017). REM sleep involves rapid eye movements, occurs more prominently in the latter half of the night, and is when dreaming mostly occurs (Ballester et al., 2020; Bathory & Tomopoulos, 2017). Separate brain states, involving distinct physiological and neurological features, distinguish each sleep state;

these sleep states cycle in rhythmic fashion throughout the night, typically repeating every 90-120 min (Ballester et al., 2020; Bathory & Tomopulos, 2017).

Two major processes interact to regulate our sleep-wake cycles within a 24-hour period: homeostatic and circadian rhythm processes. Homeostatic processes involve sleep-inducing substances called somnogens (e.g., adenosine) in the central nervous system, which accumulate during periods of wakefulness (i.e., sleep 'debt'), and increase physiological pressure for sleep (Ballester et al., 2020; Bathory & Tomopoulos, 2017). This process is governed by a biological drive for equilibrium between sleep and wakefulness; somnogen levels increase during wakefulness which compels us to sleep, and dissipate while we sleep, driving us to wake (Bathory & Tomopoulos, 2017).

Circadian processes are regulated through hormonal patterns (e.g., melatonin and cortisol) and clock gene information, as well as exogenous environmental cues ('zeitgebers') including light, temperature, and social signals such as daily routines (Baker & Richdale, 2017; Ballester et al., 2020; Bathory & Tomopoulos, 2017). Melatonin is an endogenous hormone secreted in the pineal gland in the brain, which helps to regulate sleep-wake cycles, in accordance with natural circadian rhythms of day and night (Bourgeron, 2007; Jan et al., 2008; Malow et al., 2012). Specifically, endogenous melatonin levels are typically depressed over the course of the day but increase around bedtime and remain elevated throughout the night, aiding in the establishment and maintenance of sleep (Cortesi et al., 2010; Glickman, 2010; Tordjman et al., 2005; 2012). Sleep problems are common in childhood, especially while a child is still young, as their sleep-wake cycles are immature (Richdale, 1999). The child's environment, inclusive of typical social activity, feeding routine, noise, temperature, and light and dark cycles, helps to establish and maintain the circadian sleep-wake pattern, and the REM-NREM rhythm, as a child matures (Bathory & Tomopoulos, 2017; Richdale, 1999; Spruyt & Curfs, 2015).

Studies have found that sleep architecture differs in children on the autism spectrum when compared to TD children, and children with other developmental disabilities (DD; Cortesi et al., 2010; Miano et al., 2007; Richdale & Schreck, 2009; Thirumalai et al., 2002). Specifically, research suggests that individuals on the autism spectrum exhibit immature organisation and undifferentiated stages of sleep, including increased NREM 1, decreased NREM 3 and decreased REM sleep stages, slower cyclic alternating patterns between sleep phases, and a lack of muscle atonia that is normal during REM sleep (Baglioni et al., 2016; Bourgeron, 2007; Buckley et al., 2020; Elia et al., 2000; Limoges et al., 2005; Mazzone et al., 2018; Miano et al., 2007; Richdale & Shreck, 2009; Thirumalai et al., 2002).

Further, children on the autism spectrum may have dysregulated melatonin levels, including irregular melatonin production (e.g., delayed onset of endogenous melatonin), and reduced melatonin secretion, resulting in depressed levels of melatonin at night (Díaz-Román et al., 2018; Lalanne et al., 2021; Melke et al., 2008; Souders et al., 2009; Tordjman et al., 2005; 2012). Subsequent circadian rhythm disturbances can result in sleep problems, including SOD and frequent NWs (Baker & Richdale, 2017; Cortesi et al., 2012; Liu et al., 2006; Glickman, 2010; Melke et al., 2008; Souders et al., 2009; Tordjman et al., 2005; 2012), and may increase perceived behavioural problems at bedtime (e.g., bedtime resistance if the child is not physiologically 'ready' for sleep; Baker & Richdale, 2017; K. Turner & Johnson, 2013). There is increasing evidence of biological alterations in endocrine systems and core circadian clock genes in autism, suggesting a genetic contribution to circadian rhythm disturbance (Baker & Richdale, 2017; Díaz-Román et al., 2018; Lalanne et al., 2021; Mazzone et al., 2018).

Co-occurring Conditions and Medication

Sleep problems are associated with and influenced by conditions that co-occur with autism**,** including medical conditions that disrupt sleep continuity such as epilepsy,

gastrointestinal problems (e.g., reflux), allergies and asthma, pain and obstructive sleep apnoea; and psychological and behavioural problems including ADHD, anxiety, depression, aggression and emotional dysregulation (Coury, 2010; Deliens et al., 2015; Hollway et al., 2013; Liu et al., 2006; Malow & McGrew, 2008; Mazzone et al., 2018; Nadeau et al., 2015; Reynolds & Malow, 2011; Singh & Zimmerman, 2015; Schreck, 2021; Spruyt & Curfs, 2015; Trickett et al., 2018). Anxiety, depression, and ADHD are each associated with sleep disturbance, and all commonly co-occur with autism (Cortesi et al., 2010; H. Adams, Matson & Jung, 2014; Martin et al., 2019; Mayes & Calhoun, 2009; Mazzone et al., 2018; Nadeau et al., 2015; Papadopoulos et al., 2019; Souders et al., 2017; Uren et al., 2019). These difficulties may contribute to sleep problems if there is cognitive (e.g., rumination, intrusive or racing thoughts) and/or physiological (e.g., rapid heart rate, nausea, sweating) pre-sleep arousal that interferes with sleep onset (A. Harvey, 2002; Deliens et al., 2015; Didden et al., 2014; Gregory & Sadeh, 2012; Kotagal & Broomall, 2012; Mazurek & Petroski, 2015; Mazzone et al., 2018; Mindell & Owens, 2015; Souders et al., 2017). Hyperarousal has been identified as an underlying mechanism of both sleep disturbance and anxiety in TD children (Gregory & Sadeh, 2012; Mazurek & Petroski, 2015; Richdale et al., 2014), and may contribute to sleep disturbance in children on the autism spectrum, although further research is needed (Mazurek & Petroski, 2015; Mazzone et al., 2018; Richdale et al., 2014; Richdale & Baglin, 2015; Roussis et al., 2021).

Alongside co-occurring conditions, around 50% of children on the autism spectrum take some form of medication (Hollway & Aman, 2011; Liu et al., 2006). Medication may include stimulants (e.g., methylphenidate), psychotropic medication (e.g. risperidone), and antiepileptic medication for seizures; all of which may have side effects (e.g., pre-sleep arousal, daytime sleepiness) that disrupt sleep, or exacerbate sleep issues (H. Adams, Matson,

Cervantes, et al., 2014; Jan et al., 2008; Krakowiak et al., 2008; Reynolds & Malow, 2011; Singh & Zimmerman, 2015; Mayes & Calhoun, 2009; Mazzone et al., 2018).

Characteristics of Autism

Core characteristics of autism, including difficulties with social communication, restricted and repetitive behaviours, and sensory reactivity, are all implicated in sleep disturbance (Loring et al., 2016; Mazzone et al., 2018; Reynolds & Malow, 2011; Waddington et al., 2020) leading to the suggestion that sleep disturbance may be an integral feature of autism (Martin et al., 2019; Mazzone et al., 2018). Impairments in social and verbal communication may mean children on the autism spectrum, particularly those who are non-verbal and/or who have an ID, struggle to understand sleep-related cues and instructions (Kotagal & Broomall, 2012; Reynolds & Malow, 2011). Furthermore, children on the autism spectrum may be less aware of and less motivated by social (e.g., family expectations) and environmental (e.g., increased darkness) cues by which to modify sleep-related behaviour (e.g., remaining in bed until morning; Kotagal & Broomall, 2012; Loring et al., 2016; Malow et al., 2016; Mazzone et al., 2018; S. Cohen, Conduit, et al., 2014). Indeed, greater social withdrawal has been associated with more severely disturbed sleep (Park et al., 2012; Roussis et al., 2021).

Restricted and repetitive behaviours, cognitive inflexibility and difficulty with emotional self-regulation may also contribute to sleep disturbance (Mazzone et al., 2018; Reynolds & Malow, 2011; Waddington et al., 2020). For example, children may have difficulty transitioning from preferred activities to sleep or show emotional distress in response to alterations to the bedtime routine (Kotogal & Broomall, 2012; Mazzone et al., 2018). Children may also perseverate on a thought, activity, or behaviour, such as engagement in obsessive or repetitive behaviours, or time-consuming rituals, all of which may delay bedtime or result in the child being in a stimulated state non-conducive to sleep

(Baker & Richdale, 2017; Mazzone et al., 2018; Reynolds & Malow, 2011). Further, persistent engagement in repetitive behaviour may contribute to sleep problems by delaying the onset of sleep (Hundley et al., 2016). The role of stereotypy in relation to sleep disturbance, which is a particular focus of this thesis, is discussed in detail in Chapter 5 and other subsequent chapters.

Sensory problems may also contribute to sleep difficulties (Maxwell-Horn & Malow, 2017; Mazurek & Petroski, 2015; Mazzone et al., 2018; Reynolds et al., 2012; Souders et al., 2017). Children on the autism spectrum can experience arousal dysregulation in the form of 'hyper' or 'hypo' arousal to internal and external stimuli (Reynolds et al., 2012; Souders et al., 2017). Children who experience greater sensitivity may overreact (i.e., hyper-responsivity) to sensory stimuli, whilst other children may seek out sensory stimulation as a means to achieve a greater or optimal level of sensory arousal (i.e., hypo-responsivity; Reynolds et al., 2012). Evidence suggests that sensory processing problems, particularly hyper-responsivity, may be at least partially associated with high rates of sleep problems in children on the autism spectrum (Mazurek & Petroksi, 2015; Souders et al., 2017).

Children who are hypersensitive to sensory stimuli may have difficulty disengaging from sensory aspects of their environment, resulting in difficulty with sleep onset and maintenance (Mazurek & Petroski, 2015; Reynolds et al., 2012; Souders et al., 2017). For example, light, noise, smell, sights, and tactile sensations may negatively impact on a child's ability to sleep if there is environmental distraction, stimulation, or unpleasantness (Liu et al., 2006; Mazurek & Petroski, 2015; P. Williams et al., 2006; Tomchek & Dunn, 2007). Children on the autism spectrum may also engage in sleep-interfering behaviours that produce a high level of sensory arousal, such as jumping on the bed (Reynolds et al., 2012). Research shows that greater autism symptom severity increases risk for and may predict

greater severity of sleep problems (Goldman, Surdyka, et al., 2009; H. Adams, Matson, Cervantes et al., 2014; Hollway et al., 2013; Schreck et al., 2004; Waddington et al., 2020).

Family Factors

Children's sleep problems do not occur in isolation and must be understood within the wider family context (Bronfenbrenner, 1979; Levin & Scher, 2016; Meltzer & Montgomery-Downs, 2011; Varma et al., 2021; Waddington et al., 2020). Compared to child variables, less is known regarding the types of, and ways in which, family characteristics affect children's sleep, yet it is an important aspect of understanding the factors that contribute to children's sleep problems (Hastings, 2002; Varma et al., 2021; Waddington et al., 2020). Research shows that family functioning may moderate the relationship between child variables and sleep problems in some cases (Boles et al., 2017; Meltzer & Montgomery-Downs, 2011). For example, a higher frequency of emotional and behavioural problems in children have been associated with greater bedtime resistant behaviours, which reduce TST (Boles et al., 2017). Family functioning, in particular family chaos (described as a disorganised, unpredictable, time-pressured, and noisy family environment; Matheny et al., 1995), is found to moderate the relationship between children's emotional/behavioural problems and bedtime resistance (Boles et al., 2017). It is plausible that families with such environments may be less likely to establish and enforce sleep-related structure (e.g., a bedtime routine, a consistent bedtime), and more likely to respond in ways that accidentally reinforce, rather than reduce, child sleep-interfering behaviours (Boles et al., 2017). For example, characteristics of the family environment such as the presence of a television in the child's bedroom and an absence of limits on screen time can contribute to children's sleep difficulties (Bagley et al., 2015; Boles et al., 2017).

Parents' sleep-related cognitions also affect their parenting, and thereby how well children sleep (Levin & Scher, 2016; Meltzer & Montgomery-Downs, 2011). For example,

maternal cognitions involving doubt regarding parenting self-efficacy have been associated with greater sleep disturbance (e.g., NWs) in children on and off the autism spectrum (Levin & Scher, 2016; Tikotzky & Shaashua, 2012). Mothers who have higher self-doubt may also have difficulty limiting parent-child bedtime interactions, resulting in greater parental involvement (e.g., lying with a child) throughout the night (Levin & Scher, 2016; Tikotzky & Shaashua, 2012). Conversely, parents who do not perceive their child's sleep to be problematic, even when sleep problems are clinically identified as such, are found to have higher perceived control and lower stress levels in relation to their child's sleep (Polimeni et al., 2007).

Unlike parents of TD children, parents of children on the autism spectrum may hold beliefs that sleep problems are too difficult or unable to be changed in the presence of the child's diagnosis (Beresford et al., 2016; Kirkpatrick, Louw, et al., 2019; Levin & Scher, 2016; McLay et al., 2020). Parents may also unintentionally reinforce sleep-interfering behaviours owing to uncertainty or sympathy related to the child's diagnostic condition, for example, providing food owing to uncertainty about whether their child is hungry, and allowing access to technological devices because they believe it calms their child down (Beresford et al., 2016; Kirkpatrick, Luow, et al., 2019). On the other hand, Levin and Scher (2016) found that mothers of children on the autism spectrum did not report more problematic sleep-related cognitions or greater difficulty limiting bedtime interactions than mothers of same-age TD children, which suggests these factors may not be specifically related to children's diagnostic condition.

It is important to note that parents' sleep-related cognitions and family value systems are also influenced by broader processes such as socio-economic status and culture (Bronfenbrenner, 1979; Hastings, 2002; Wiggs, 2007). For example, socioeconomic characteristics including lower paternal education and lower family income have been

associated with sleep disturbance in children on the autism spectrum; however, further research is needed to understand the specific mechanisms by which such factors affect children's sleep (Waddington et al., 2020). It is important that future research seeks to gain comprehensive understanding of the range of variables that affect children's sleep, outside of child factors alone (Deserno et al., 2019; Hastings, 2002; Levin & Scher, 2016; Waddington et al., 2020).

Consequences of Sleep Disturbance

The importance of sleep to all aspects of human functioning cannot be understated. Sleep is essential for life, and plays a vital role in health and development, promoting behavioural, emotional, and social regulation, learning and memory consolidation, brain maturation, physical growth, and energy restoration (Abel et al., 2018; Astill et al., 2012; Jan et al., 2008; K. Turner & Johnson, 2013; Richdale, 2013; Roussis et al., 2021; Spruyt & Curfs, 2015). Conversely, sleep deprivation is associated with a myriad of negative consequences across the life span, owing not only to the absence of sleep, but also to the effects of extended wakefulness (Krause et al., 2017). Sleep loss as modest as 30 min in duration can impair daytime neuropsychological functioning (Richdale & Wiggs, 2005).

In children on the autism spectrum for whom sleep problems are often severe and chronic, the consequences of poor sleep are considerable, and include physical, psychological, behavioural, and emotional sequelae (Jan et al., 2008; M. Taylor et al., 2012; Miano et al., 2007; Richdale & Wiggs, 2005; S. Cohen, Conduit, et al., 2014; Schreck et al., 2004; Spruyt & Curfs, 2015; Tudor et al., 2012; Wiggs & Stores, 1996). Whilst a predominance of studies have utilised parent-reported measures of sleep (e.g., the Child Sleep Habits Questionnaire [CSHQ]; J. Owens et al., 2000), available research suggests the effects of poor sleep are evidenced by instrumental sleep measures (e.g., actigraphy, polysomnography) as well as parent-report (Abel et al., 2018; Bangerter et al., 2020; Elrod &

Hood, 2015; Goldman, Surdyka, et al., 2009; Lindor et al., 2019; S. Cohen, Conduit, et al., 2014; Veatch et al., 2017).

Sleep disturbance is found to exacerbate internalising and externalising problems in children on the autism spectrum, including increased symptoms of anxiety, depression, negative affect and mood disturbance, irritability, inattention, impulsivity, hyperactivity, oppositional behaviour, aggression and self-injury (Allik et al., 2006; Goldman et al., 2011; H. Adams, Matson & Jang, 2014; Mayes & Calhoun, 2009; Mazurek & Sohl, 2016; Nadeau et al., 2015; Park et al., 2012; Patzold et al, 1998; Richdale & Baglin, 2015; Roussis et al., 2021; S. Cohen, et al., 2018; Schreck, 2021; Sikora et al., 2012; Soke et al., 2017; Y. Wang et al., 2021). Longitudinal research suggests sleep problems may predict later anxiety (May et al., 2015), and recently Schreck (2021) demonstrated that both sleep quality and sleep quantity is predictive of internalising (e.g., anxiety, depression) and externalising (e.g., aggression) challenges.

Further, sleep disturbance can impede cognitive and adaptive functioning, and has been associated with lower intellectual functioning, impaired daily living skills (e.g., difficulty completing self-care tasks such as toileting), academic performance (Deliens et al., 2015; M. Taylor et al., 2012; Polimeni et al., 2007; Richdale & Wiggs, 2005; Sikora et al., 2012; Tyagi et al., 2019), and poorer health-related quality of life (Delahaye et al., 2014). Examples of the consequences of sleep disturbance for physical health include associations with obesity, epilepsy/seizures, gastrointestinal symptoms, feeding problems (e.g., decreased appetite), respiratory issues, visual and motor impairment and increased accidental injuries (Accardo & Malow, 2015; Jan et al., 2008; Mannion & Leader, 2013; P. Williams et al., 2004; Tyagi et al., 2019; Yang et al., 2018; Zuckerman et al., 2014).

Sleep disturbance is also associated with autism symptom severity (Gabriels et al., 2005; Goldman et al., 2011; H. Adams, Matson, Cervantes, et al., 2014; Hundley et al., 2016;

S. Cohen, Conduit, et al., 2014; Schreck et al., 2004; Tudor et al., 2012). Sleep disturbance is associated with diminished prosocial behaviours, including social withdrawal and decreased reciprocal social communication and interaction (H. Adams, Matson, Cervantes, et al., 2014; Johnson et al., 2018; Richdale & Schreck, 2009; Schreck et al., 2004; Park et al., 2012; Phung & Goldberg, 2017; Roussis et al., 2021; Tudor et al., 2012; Veatch et al., 2017), as well as heightened sensory abnormalities (e.g., hyper-responsivity; Mazurek & Petroski, 2015; Tyagi et al., 2019; Tzischinsky et al., 2018). Numerous studies have demonstrated that higher rates of restricted and repetitive behaviours are found in children on the autism spectrum who are poor sleepers (Gabriels et al., 2005; Goldman et al., 2011; Hundley et al., 2016; Hoffman et al., 2005; Mayes & Calhoun, 2009; Schreck et al., 2004; Tudor et al., 2012). As noted, the impact of sleep disturbance on stereotypy is discussed in further detail in Chapter 5.

The severity of sleep disturbance may differentially affect autism symptom severity and the degree of internalising and externalising problems that children experience (Lindor et al., 2019; Sikora et al., 2012). For example, Schreck et al. (2004) examined the relationship between parent's reports of sleep problem severity and autism symptoms, using the Behavioral Evaluation of Disorders of Sleep (Schreck & Mulick, 2000), and the Gilliam Autism Rating Scale (GARS; Gilliam, 1995). Sleep problem severity was correlated with autism symptom severity; in particular, short sleep duration was found to predict stereotyped behaviour, social skill deficits and overall autism severity. Greater internalising and externalising symptoms are consistently found in children with sleep problems ('poor' sleepers) compared to those without ('good' sleepers; Goldman, Surdyka, et al., 2009; Goldman et al., 2011; H. Adams, Matson & Jang, 2014; Lindor et al., 2019; Malow, Marzec, et al., 2006; Park et al., 2012), and further, are more severe in those with more severe sleep disturbance (H. Adams, Matson & Jang, 2014; Roussis et al., 2021; Sikora et al., 2012).

Recently, Lindor and colleagues (2019) examined whether severity of sleep problems (none, mild, or moderate-severe) moderated the relationship between autism symptom severity and internalising and externalising symptoms as captured by the Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2001). Sleep, autism symptom severity, and internalising and externalising problems were assessed in 40 boys (aged 5-12 years) on the autism spectrum, using the CSHQ, the Social Responsiveness Scale – Second Edition (Constantino & Gruber, 2012) and CBCL, respectively. Results showed that the relationship between autism symptom severity and internalising/externalising problems differed according to sleep problem severity. Specifically, for children with mild or no sleep disturbance, autism symptom severity was associated with internalising/externalising problems wherein milder severity of autism symptoms were associated with milder severity of problems, and more severe autism symptoms with clinically significant problems. In comparison, for those with moderate-severe sleep disturbance, internalising/externalising problems were clinically elevated regardless of autism symptom severity (i.e., mild, moderate-severe; Lindor et al., 2019). These results are important, because they suggest that children on the autism spectrum with moderate-severe sleep disturbance may experience clinically significant internalising and externalising symptomatology, regardless of autism symptom severity (Lindor et al., 2019).

Further research is needed to better understand the complex relationship between autism and associated challenges (Lindor et al., 2019). For example, H. Adams, Matson and Jang (2014) found that externalising problems may be more affected by the degree of sleep problem severity than internalising challenges. Overall, a robust body of evidence suggests that challenges in development and broader functioning associated with autism are exacerbated by sleep disturbance, resulting in further impairment for these children (Johnson et al., 2018; Richdale & Schreck, 2009; Sikora et al., 2012; Mazzone et al., 2018).

Wider Consequences of Sleep Disturbance

Sleep problems do not solely affect the child but have a significant negative impact on family members (Bathory & Tomopoulos, 2017; Jan et al., 2008; Chu & Richdale, 2009; Cotton & Richdale, 2006; Gallagher et al., 2010; Kirkpatrick, Louw, et al., 2019; Kodak & Piazza, 2008; Martin et al., 2019; Meltzer & Montgomery-Downs, 2011; Quine et al., 1991). Of particular note, sleep problems are found to affect the personal sleep quality of other family members (Courtier et al., 2005; Gallagher et al., 2010; Johnson et al., 2018; Lopez-Wagner et al., 2008; Meltzer, 2008; Robinson & Richdale, 2004; Wiggs & Stores, 2001). For example, child sleep quality is found to significantly predict maternal (Hoffman et al., 2008; Meltzer, 2008; Meltzer & Mindell, 2008) and paternal (Meltzer, 2008; 2011) sleep quality. Sleep deprivation can be marked for parents and caregivers (henceforth referred to as 'parents') caring for a child on the autism spectrum and is linked to a variety of deleterious consequences (Krakowiak et al., 2008; Meltzer & Montgomery-Downs, 2011). For example, child sleep problems have been associated with parental stress and depression (Martin et al., 2019; Meltzer, 2011). Specifically, maternal sleep disturbance is significantly associated with poor maternal psychological wellbeing, including depression, anxiety, stress, and fatigue (Chu & Richdale, 2009; Doo & Wing, 2006; Gallagher et al., 2010; Hoffman et al., 2008; Johnson et al., 2018; Levin & Scher, 2016; Martin et al., 2019; Meltzer & Mindell, 2008; Meltzer, 2011; Wiggs & Stores, 2001).

Further, sleep problems can increase parental emotional distress (e.g., a sense of helplessness, frustration and being overwhelmed) and irritability, negatively affect family functioning, decrease relationship satisfaction, make parenting practices more taxing, and affect employment and financial opportunities (e.g., through reduced capacity to work) and can increase risk for child maltreatment (Durand & Mindell, 1990; Kirkpatrick, Louw, et al., 2019; Kodak & Piazza, 2008; Reynolds & Malow, 2011; Stores, 1996; Richdale et al., 2000).

Thus, sleep disturbance adds substantial burden and complexity to the challenges faced by parents of children on the autism spectrum (Hoffman et al., 2008; Johnson et al., 2018; Krakowiak et al., 2008; Patzold et al., 1998).

Given the widespread direct and indirect adverse consequences of sleep disturbance on health and quality of life for children and families, the need to routinely assess for and treat sleep problems in children on the autism spectrum is clear (Levin & Scher, 2016; Reynolds et al., 2012; Reynolds & Malow, 2011; Roussis et al., 2021). Sleep is a core mechanism for overall adaptive functioning (S. Cohen, Conduit, et al., 2014), and therefore represents a pivotal target for treatment, given that improved sleep may create far reaching and tangible benefits for children and their families (Delahaye et al., 2014; Lindor et al., 2019; Richdale & Wiggs, 2005; Roussis et al., 2021; Tudor et al., 2012; Vriend et al., 2011). Alleviation of sleep disturbance could ease autism symptomatology, resulting in beneficial effects on a child's neurodevelopmental growth, quality of life, and daily functioning (Hoffman et al., 2005; S. Cohen, Conduit, et al., 2014; Schreck et al., 2004; Sivertsen et al., 2012; Spruyt & Curfs, 2015). Better sleep for children is also likely to mean better sleep for adults and siblings, which could reduce parental stress, anxiety, and depression, and improve overall sense of family wellbeing (Chu & Richdale, 2009; Johnson et al., 2018; Tudor et al., 2012; Meltzer, 2008). Focussing research efforts on better understanding the factors that contribute to the development and maintenance of sleep problems in children on the autism spectrum can provide critical targets for early intervention, to minimise the impact of long-enduring problems (Baker et al., 2013; Deserno et al., 2019; Goldman et al., 2012; Hoffman et al., 2005; Mazurek et al., 2019; Singh & Zimmerman, 2015; Sivertsen et al, 2012).

Bidirectional Influences

Critically, many of the factors that contribute to sleep disturbance are also worsened by sleep disturbance; thus, the relationship between autism and sleep disturbance is

bidirectional and complex, and contributing factors are likely to overlap (Díaz-Román et al., 2018; H. Adams, Matson, Cervantes, et al., 2014; Krakowiak et al., 2008; Lindor et al., 2019; Maxwell-Horn & Malow, 2017; Martin et al., 2019; Mazzone et al., 2018; Reynolds & Malow, 2011; Roussis et al., 2021). Richdale and Schreck (2009) developed a biopsychosocial model of sleep disturbance, which hypothesised that biological, psychological, behavioural, and social factors reciprocally interact to contribute to the development and maintenance of sleep problems. This model was foundational in highlighting the complex and bidirectional influences underlying sleep problems in autism.

Subsequently, Hollway and Aman (2011) also theorised a bidirectional framework for conceptualising sleep problems in children on the autism spectrum. Within this framework, the core characteristics of autism, moderated by intellectual functioning, serve as *vulnerability factors*, which interact with *environmental stressors* (e.g., changes to routine) to trigger maladaptive *coping strategies* in the form of internalising (e.g., anxiety) and externalising (e.g., aggression) problems. Internalising and externalising problems lead to hyperarousal and/or sleep problems, which reciprocally exacerbates internalising and externalising problems. Co-occurring medical conditions (e.g., epilepsy), including medication use, independently increase risk for hyperarousal and/or sleep disturbance (Hollway & Aman, 2011). In return, medical conditions (e.g., epilepsy) may also be worsened by sleep disturbance (Accardo & Malow, 2015; Mazzone et al., 2018). Conversely, sleep disturbance and greater internalising and externalising problems intensify autism symptom severity (H. Adams, Matson, Cervantes, et al., 2014; Hollway & Aman, 2011; Schreck et al., 2004; Meltzer & Mindell, 2008).

The frameworks provided by Richdale and Schreck (2009) and Hollway and Aman (2011) encapsulate the bidirectional influence of sleep-interfering variables and sleep disturbance in autism, including how additive effects may occur in children with multiple risk

factors (e.g., greater autism symptom severity, co-occurring medical conditions; Hollway & Aman, 2011). It is important that research seeks to better understand the autism and sleep relationship, as the directionality of this relationship carries important implications for treatment; for example, treating sleep problems may indirectly reduce autism symptom severity and co-occurring problems (H. Adams, Matson, Cervantes, et al., 2014; Hollway & Aman, 2011; Hundley et al., 2016; Richdale & Schreck, 2009).

Evidence suggests there is also a cyclic and interactive relationship between parent/family characteristics and child sleep disturbance (Johnson et al., 2018; Levin & Scher, 2016; Martin et al., 2019; Meltzer & Mindell, 2008; Richdale & Schreck, 2009; Varma et al., 2021; Waddington et al., 2020). For example, research in TD infants shows that infant sleep problems contribute to maternal depressive symptoms (with this relationship moderated by maternal sleep quality), and that maternal depressive symptoms may also contribute to infant sleep problems (Meltzer & Montgomery-Downs, 2011). Greater autism symptom severity and child problem behaviour can also change parents' beliefs about their parenting self-efficacy (e.g., reduced belief in self-efficacy), and contribute to poor parental wellbeing, which in return affects how well children sleep (Deserno et al., 2019; Hastings, 2002; Martin et al., 2019; Polimeni et al., 2007).

Although the mechanisms by which child sleep problems affect parent/family factors and vice versa are unclear, there are a number of ways by which these factors may interact (Martin et al., 2019; Richdale & Schreck, 2009). As previously discussed, child sleep problems can negatively affect parents' sleep quality, functioning and wellbeing, and make parenting practices more taxing (Chu & Richdale, 2009; Hoffman et al., 2008; Johnson et al., 2018; Kodak & Piazza, 2008; Martin et al., 2019; Meltzer, 2008; Meltzer & Montgomery-Downs, 2011). In return, diminished parental and family functioning can disrupt children's sleep patterns (Meltzer & Montgomery-Downs, 2011; Varma et al., 2021). Parents who are

fatigued from their own sleep being disrupted, and/or stressed, anxious or depressed may in turn, be more likely to respond to children's behaviour in ways that are non-conducive to quality sleep (i.e., that reinforce the child's behaviour problems, e.g., allowing extended screen time, reactively lying down with a child to settle or resettle them to sleep), and strain parent-child interactions, which unsettles the child's sleep (Hastings, 2002; Johnson et al., 2018; Levin & Scher, 2016; Martin et al., 2019; Richdale & Shreck, 2009; Waddington et al., 2020). It also is possible that child sleep and daytime problem behaviours are independently affected by parenting practices (e.g., difficulty with limit-setting during the day and at night), thus children who have greater daytime behavioural problems may also have greater behavioural problems that interfere with sleep processes (Polimeni et al., 2007). Thus, the processes that shape and are shaped by child sleep problems are dynamic and multifaceted (Martin et al., 2019; Richdale & Schreck, 2009).

Future research is needed to better understand the directionality of effects between autism and sleep disturbance, including whether relationships are bidirectional, the extent to which they are causal or correlational, and/or if they are mediated or moderated by other factors (Deserno et al., 2019; Hastings, 2002; Levin & Scher, 2016; Martin et al., 2019; Mazzone et al., 2018; Polimeni et al., 2007; Richdale & Schreck, 2009; Tikotzky & Shaashua, 2012; Uren et al., 2019). Further, the mechanisms by which sleep disturbance affects autism symptom severity and co-occurring problems are not well understood (Mazurek & Sohl, 2016; Mazzone et al., 2018; S. Cohen et al., 2018; Sannar et al., 2018). A large majority of research examining the sleep and autism relationship has been cross-sectional and correlational in design, thereby limiting our understanding of causality and directionality (H. Adams, Matson, Cervantes, et al., 2014; Hoffman et al., 2008; Hollway et al., 2103; Hollway & Aman, 2011; Hundley et al., 2016; Martin et al., 2019; Mazurek et al., 2019; S. Cohen, Conduit, et al., 2014; Singh & Zimmerman, 2015; Wiggs & Stores, 1999).

Furthermore, sleep problems in autism are heterogeneous, but have largely been examined as a single construct (Martin et al., 2019). Research suggests that certain types of sleep problems may be associated with distinct problems behaviours; for example, stereotypic behaviours have been specifically associated with SOD (Tudor et al., 2012), short sleep duration (Sannar et al., 2018; Schreck et al., 2004; Tudor et al., 2012; Veatch et al., 2017), sleep fragmentation (Goldman, Surdyka, et al., 2009), sleep-disordered breathing (Hoffman et al., 2005) and with nightmares, night terrors, confusional arousals, and screaming during the night (Schreck et al., 2004). Specific sleep problems may also differentially affect parent stress and mental health (Martin et al., 2019). Future research is needed to better understand the complex interplay between sleep problems and common and associated characteristics of autism (Martin et al., 2019; S. Cohen et al., 2018). Notwithstanding the multiple determinants of sleep disturbance, the onset and maintenance of sleep problems is often behavioural in origin (Beresford et al., 2016; Blampied, 2013a; Blampied & France, 1993; Didden et al., 2002; Richdale & Schreck, 2009; Richdale & Wiggs, 2005).

A Behavioural Model of Sleep Disturbance

A behavioural model of sleep can account for difficulties with sleep onset and sleep maintenance (Blampied & France, 1993) by paying particular attention to environmental and social-interactional variables. Operant behaviour theory stipulates that behaviour operates in an environmental context that provides both antecedents (i.e., events that precede behaviour) and consequences (i.e., events that directly follow behaviour; Blampied, 2013a; Skinner 1969). Antecedents, behaviour, and its consequences create a three-term contingency (‘A-B-C’). Antecedents signal the consequences likely to occur when a behaviour occurs, and consequences affect the likelihood that the behaviour will reoccur (i.e., the strength of the behaviour) via the processes of reinforcement and punishment. In turn, consequences affect the degree to which the antecedent stimulus controls the behaviour (i.e., the strength of

stimulus control; Skinner, 1969). Consequences take the form of either reinforcers that increase the probability of a response reoccurring, or as punishers that decrease the probability of a response reoccurring (Hanley et al., 2003; Skinner, 1969). Both reinforcement and punishment can be either positive, meaning behaviour is affected by a consequence appearing in the environment and being experienced by the individual, or negative, meaning behaviour is affected by a consequence being removed from the experience of the individual or being reduced in its experienced intensity (Blampied, 2013a; Skinner, 1969). Consequences must follow behaviour in close temporal proximity to have a behavioural effect.

Sleep is a primary reinforcer because it is a cyclic bio-behavioural state which human beings are fundamentally motivated to enter (Blampied & France, 1993). The state of being asleep, therefore, reinforces the behaviour of falling asleep (i.e., falling asleep is an operant). It is this behaviour of 'falling asleep', and the conditions and pre-requisites associated with falling asleep, that is the focus of attention in behavioural accounts of sleep (Blampied & Bootzin, 2013; Blampied & France, 1993; Jin et al., 2013). The behaviour of falling asleep can be thought of as the final step in an operant behaviour chain that begins with bed-preparation behaviours and ends with behavioural quietude (lying quietly in bed, with low levels of behavioural, cognitive, and emotional arousal), which immediately precedes falling asleep (Blampied & France, 1993). Behavioural quietude must be maintained for a sufficient period for the wake-sleep transition to occur (Blampied, 2013a; Blampied & France, 1993).

The operant chain of behaviours by which an individual ultimately enters into sleep is under stimulus control. Distinct behaviours (e.g., brushing teeth, putting on pyjamas) are linked together by stimuli that serve both as antecedents for the next behaviour in the chain and as reinforcing consequences for the previous behaviour, with the terminal link in the chain being behavioural quietude as the consummatory response (i.e., the response that

consumes the reinforcer of sleep; Blampied, 2013a). Stimuli that are reliably present when reinforcing consequences occur acquire discriminative properties (and are termed discriminative stimuli [S^D]) that signal that the reinforcer is available. For example, a dark and quiet bedroom with comfortable bedding and temperature are common S^D that set the occasion for sleep (Blampied, 2013a; Cooper et al., 2020; Jin et al., 2013). The behavioural response (e.g., 'falling asleep') is not compelled to occur in the presence of S^D, it is only more probable because of past learned experiences (e.g., sleep typically occurs in a darkened room). S^D can include discrete events and stimuli; for example, a specific bed-related instruction from a parent (e.g., "put on your pyjamas") or seeing pyjamas laid on the bed are stimuli that make the child's behaviour of 'putting on pyjamas' more probable, if doing so has historically been met with a reinforcing consequence, such as a story and a cuddle.

Sleep items, referring to any item(s) consistently taken into the bed (e.g., soft toy, pacifier), can also become S^D for sleep (Burnham et al., 2002). Research shows that infants across cultures tend to incorporate sleep items into independent sleep practices as they learn to self-settle, and that age tends to affect the type of sleep item used, as well as the strength of the attachment to a particular item (Burnham et al., 2002). For example, sleep items can include a child's own body parts, such as the behaviour of sucking a thumb observed in infants and young children (Burnham et al., 2002). Appropriate sleep items are those that promote sleep-conducive behaviour (e.g., lying quietly in bed). Routine and regularity in the pre-sleep environment and bedtime activities is highly important to firmly embed appropriate S^D within the behaviour chain leading up to sleep, which in turn affects how well children sleep (i.e., strengthens stimulus control for sleep and enhances circadian control of sleep onset; Blampied, 2013a; Blampied & France, 1993; Mindell et al., 2009).

The modifiable parent and child practices that promote healthy sleep (i.e., regular sleep of age-appropriate sufficient duration, and of good quality) can be collectively called

'sleep hygiene' practices. This encompasses several domains including the bedtime routine, bed and wake times, and the sleep environment (Hauri, 2011; Jan et al., 2008; Mindell et al., 2009). Research shows that a structured, predictable bedtime routine that includes quiet, calming pre-sleep activities (e.g., a bath, putting on pyjamas, brushing teeth, reading stories), and a consistent sleep environment (e.g., a cool, quiet, dark, sleeping environment), are associated with healthy sleep outcomes (Bathory & Tomopoulos, 2017; Blampied, 2013a; Jan et al., 2008; Mindell et al., 2009). Where and how children fall asleep also affects how well children sleep (Bathory & Tomopoulos, 2017; Mindell et al., 2009). It is important that children go to bed in their own bed drowsy (i.e., physiologically ready to go to sleep) but awake, so that the child is aware of the location where they are falling asleep, and so that the child's own bed becomes established as an S^D for sleep (Bathory & Tomopoulos et al., 2017; Mindell et al., 2009).

Whilst parents may necessarily be actively involved in the initial stages of a child's bedtime behaviour chain (e.g., instructing and helping a child to put on their pyjamas and brush teeth), it is important that parents reduce their involvement in the final stages of the chain (i.e., once the child is bid goodnight), so as not to interfere with behavioural quietude (e.g., through on-going attention) and thereby allow the child to settle to sleep independently. Children's ability to self-settle (i.e., attain behavioural quietude on their own) is an important developmental life skill that allows children to initiate sleep without parental assistance (Bathory & Tomopoulos, 2017; Blampied & France, 1993; Burnham et al., 2002; Mindell et al., 2009). Importantly, children who are able to self-settle are more likely to resettle quietly and independently during brief arousals in between sleep cycles throughout the night, without requiring parental assistance (Bathory & Tomopoulos, 2017; Blampied & France, 1993; Burnham et al., 2002).

Conversely, difficulties with sleep onset and maintenance can occur when there is disruption to or a lack of stimulus control for sleep (Blampied & France, 1993). Difficulty with independent re-initiation of sleep can occur when S^D are initially present at bedtime but are then unavailable in the sleep environment during the night (Blampied & France, 1993; Weiskop et al., 2005). For example, if a parent is regularly present when the child falls asleep (e.g., by lying with the child), then the presence of the parent becomes an S^D for sleep, and sleep cannot occur when the parent is absent. This includes during the night if the child wakes, where the parent's presence is again needed for the child to re-initiate sleep (Blampied, 2013a; Blampied & France, 1993; Henderson et al., 2010; Mazzone et al., 2018; Weiskop et al., 2005). The presence of a parent at bedtime is associated with increased NWs and reduced TST in children of all ages (Mindell et al., 2009). Parent rituals such as singing to, rocking, or patting a child to sleep can also become established as S^D for sleep, however, once established, can become difficult for parents to change or cease, without disturbing sleep onset and re-initiation (Blampied, 2013a; Blampied & France, 1993; Jan et al., 2008; Polimeni et al., 2007).

Changes within the physical sleep environment such as variations in light, temperature, or noise level, can also disrupt stimulus control for sleep (Bathory & Tomopoulos, 2017; Blampied, 2013a; Jin et al., 2013). Likewise, a lack of or a dysfunctional bedtime routine can make the onset of sleep more difficult, as may be the case in family environments that are characterised by higher family chaos, or in the absence of enforced bedtime limits (Boles et al., 2017; Díaz-Román et al., 2018; Matheny et al., 1995; Mazzone et al., 2018; Wachs, 2010). Finally, stimuli leading to alternative sources of reinforcement that are present in a child's sleep environment may control opportunities for behaviour that disrupts a child's bedtime behaviour chain (Blampied, 2013a; Blampied & France, 1993; Jin et al., 2013). For example, access to electronic devices or stimulating toys are common and

salient S^D that control sleep-interfering behaviour (e.g., play) which compete with a child's ability to attain the behavioural quietude necessary for sleep to occur (Blampied, 2013a; J. Owens et al., 1999; Jan et al., 2008).

A child is unlikely to fall asleep in the presence of external S^D alone; sufficient physiological sleep pressure is also required for sleep onset to occur (Ballester et al., 2020; Bathory & Tomopoulos, 2017). Sleep pressure may be understood as a motivational variable that alters the value of sleep as a reinforcer, and in turn affects the salience and strength of S^D that signal that the reinforcer is available. Motivational variables are antecedent conditions that establish the value of reinforcement for a particular response, and increase or decrease an individual's motivation to engage in that particular response and related responses. For example, not eating lunch is an antecedent condition that may highly motivate an individual to eat later (e.g., at dinner), and make the act of eating dinner highly reinforcing.

The conditions that cause motivations to change, and in turn which alter the value of the putative reinforcer, are called *motivating operations* (MOs; Cooper et al., 2020; Michael, 1982). Specifically, reinforcer value is influenced by the level of satiation or deprivation an individual experiences with regard to a particular event or experience (Vollmer & Iwata, 1991; Lang, Koegal, et al., 2010). For example, the act of not eating lunch creates a state of deprivation with regards to the value of reinforcement typically obtained by lunch-time food. This deprivation increases the value of the reinforcer, and, therefore, the individual's motivation to eat; in this instance, the deprivation is an *establishing operation* (EO; Michael, 1982; 1993). Conversely, eating a large lunch may lead to a state of satiation which reduces the value of the reinforcer, thereby, in the immediate future, reducing the individual's motivation to eat; this is an *abolishing operation* (AO; Lang, Koegal, et al., 2010; Michael, 1982; 1993; Rapp et al., 2017). Thus, MOs alter both the value of reinforcement and the

likelihood of behaviour (i.e., behaviour associated with that motivational state is more or less likely to occur).

With regards to sleep, the homeostatic processes that increase sleep pressure over the course of the day thus increasing our motivation to sleep function as an EO, and the dissipation of physiological sleep pressure following sleep, thus decreasing our motivation to sleep, function as an AO (Ballester et al., 2020; Bathory & Tomopoulos, 2017). Factors such as the quantity and quality of the previous night's sleep, time awake (temporal proximity to previous sleep) and exertion from daytime activities, have a motivational effect on the behaviour of falling asleep (Borbely & Achermann, 1992). It is important therefore, that parents restrict children's sleep so that it only occurs between set hours (i.e., a set bed and wake time, including any age-appropriate naps), since allowing children to sleep in and make up for lost sleep time can dissipate sleep pressure, thereby contributing to or exacerbating difficulties with sleep onset and maintenance (Vriend et al., 2011).

A later bedtime than usual can induce mild sleep deprivation and temporarily increase the value of sleep as well as the chain of behaviours leading to this reinforcer (Michael, 1982). Conversely, if a child is put to bed too early (i.e., before they are ready for sleep) then sleep onset may be less likely to occur. Motivational states can also compete with one another (i.e., *motivational state competition*); for example, a child may be motivated to sleep, but if also feeling hungry, may call out to a parent for food, thus interfering with their ability to fall asleep. Furthermore, if a child frequently lies in bed awake for extended periods of time, then the bed and bedroom environment may become S^D for responses of arousal, rather than sleep, thus making sleep onset more difficult (Blampied & Bootzin, 2013; Mindell & Owens, 2015). It is important therefore, that parents maintain a consistent and age-appropriate bed and wake time for children, to ensure there is sufficient motivation for the child to enter into and maintain sleep (Ballester et al., 2020; Bathory & Tomopoulos, 2017; Mindell et al., 2009).

Parents must also provide predictable responses to child behaviour, while they instruct and supervise the child during the sleep onset chain each night (Blampied, 2013a; Blampied & France, 1993). This includes minimising stimuli and reinforcement in the sleep environment that are associated with sleep-interfering behaviours. Reinforcement for sleep-interfering behaviour includes positive (e.g., parent attention) and negative (e.g., escape from the demand to go to sleep, escape from fear in the sleep setting) consequences. In children on the autism spectrum, sleep-interfering behaviour may also be reinforced via non-social sensory-based (i.e., automatic) consequences the behaviour itself produces, such as motor or vocal stereotypy (e.g., body-rocking, or laughing or talking to oneself in bed) or self-injurious behaviour (e.g., head-banging; Didden et al., 2002; Jin et al., 2013; Rapp & Lanovaz, 2016). Parent attention is a common reinforcing consequence of behaviour that can serve to shape coercive parent-child interactions, and disrupt sleep (Blampied, 2013a; Didden et al., 2002; Lawton et al., 1991). Child behaviour such as making demands of parents after being put to bed and seeking out parent contact (i.e., CCs) is socially mediated behaviour that is positively reinforced when a child receives parent attention (Didden et al., 2002; Jin et al., 2013). Similarly, a child who does not want to be in bed and protests may be positively and negatively reinforced for their behaviour by being removed from the bed (Didden et al., 2002; Jin et al., 2013).

Parent-child 'behaviour traps' occur when parent's responses to children's behaviour are also reinforced; for example, a parent may be negatively reinforced for removing a child from their bed through the temporary cessation of the child's protest behaviour. Thus, coercive parent-child interactions that maintain inappropriate sleep behaviour can be mutually reinforcing for parent and child (France & Blampied, 1999; G. Patterson, 1982; Polimeni et al., 2007). In other words, such interactions are likely to continue and to increase when both child and parent learn that engaging in such behaviours results in a preferred

outcome and/or avoids an aversive outcome (Blampied & France, 1993; Didden et al., 2002; France et al., 2003). Parents may be particularly likely to respond to children in ways that avoid or reduce perceived child distress (e.g., by providing attention/reassurance), which in turn may reduce the parent's own anxiety and distress (Blampied & France, 1993; France et al., 2003; Polimeni et al., 2007).

As discussed above, there are a number of parent/family and child interactive effects that may increase the likelihood of sleep-related behaviour traps occurring, and contribute to their maintenance, including parent and child characteristics, parenting practices and attributions (e.g., self-efficacy) and diminished parental functioning (e.g., owing to fatigue, stress, anxiety and/or depression; France & Blampied, 1999; Levin & Scher, 2016; Polimeni et al., 2007; Richdale & Wiggs, 2005). These interactions may occur during the night as well as during the sleep onset period (France et al., 2003; Polimeni et al., 2007). Disruption in the child's sleep onset chain is particularly likely to occur when stimuli and reinforcement associated with sleep-interfering behaviour are salient, desirable, and more immediately available than the delayed reinforcement of sleep (Blampied, 2013a; Blampied & Bootzin, 2013).

Overall, optimal sleep outcomes are achieved when the behaviour of 'falling asleep' is brought under clear and consistent stimulus control (Blampied & France, 1993). Regularity in the sleep environment and bedtime routine are necessary to establish appropriate (sleep-conducive) S^D within a child's sleep onset chain, whilst disruption to or a lack of routine can make the onset of sleep more difficult (Blampied, 2013a). Antecedent MOs that increase or decrease sleep pressure also affect sleep onset and maintenance. Inappropriate S^D for sleep includes locations other than the child's own bed (e.g., the parent bed), and that which prompt sleep-interfering behaviour (e.g., stimulating toys), or which the child cannot produce themselves (e.g., dependence on a parent's presence to fall asleep). Consequences for sleep-

interfering behaviours include positive, negative and/or automatic consequences. In essence, the behaviour of falling asleep is intimately affected by its dynamic and reciprocal relationship with antecedent factors and consequences.

Conclusion

In sum, sleep problems in children on the autism spectrum are ubiquitous, and commonly include difficulties with sleep onset and maintenance. The aetiology of sleep problems is likely to be the result of complex and interactive effects between a range of factors, including biological, psychological, social, behavioural, medical, and environmental variables. Sleep problems have a widespread detrimental impact on children and their families, including worsening autism symptom severity and co-occurring problems, parental stress, and mental health; thereby adding burden and complexity to those affected by autism. There is clear and pressing need for effective intervention to reduce the adverse consequences associated with sleep disturbance, to allow for improved functioning within whole families. Behavioural models of sleep disturbance reveal learned, and therefore modifiable, contributing factors, highlighting the crucial role of behaviour management techniques for reducing sleep problems in children on the autism spectrum.

Chapter 2

Functional Behaviour Assessment and Treatment of Sleep Problems in Children on the Autism Spectrum

This chapter focusses on treatment for sleep disturbance in children on the autism spectrum. Behavioural and pharmacological interventions are the two main types of treatment for sleep disturbance in children on the autism spectrum (Pattison et al., 2020). Behavioural treatments are the focus of this chapter and were used with all participant children reported in this thesis, for several reasons. First, when medical aetiologies have been excluded (Buckley et al., 2020; Pattison et al., 2020; Scantlebury et al., 2018), behavioral interventions are recommended as the first line of treatment for pediatric sleep problems (Buckley et al., 2020; Keogh et al., 2019; Pattison et al., 2020; Phillips et al., 2020; Richdale & Wiggs, 2005; Rigney et al., 2018; Singh & Zimmerman, 2015; Vriend et al., 2011). Pharmacological treatments (e.g., melatonin, antipsychotics, antidepressants, benzodiazepines) are typically recommended if behavioural interventions have been trialled and are ineffective in resolving a child's sleep problems, or when the adverse impact of sleep disturbance on the child and family is chronic (Keogh et al., 2019; Pattison et al., 2020; Phillips et al., 2020; Rigney et al., 2018).

Second, there is insufficient evidence regarding the efficacy, safety (particularly regarding long-term use), and side effects of pharmacological treatments for the management of sleep problems in pediatric populations (Buckley et al., 2020; Keogh et al., 2019; McLay, Schluter, et al., 2021; Rigney et al., 2018; Singh & Zimmerman, 2015). Research shows that in comparison to pharmacological treatments, behavioural interventions are preferred by parents and are viewed as being as effective as medication, may result in long-term gains, and are not associated with the adverse side effects of medication (Beebe, 2016; Keogh et al., 2019; Rigney et al., 2018). Third, as discussed in Chapter 1, behavioural variables clearly contribute to the development and maintenance of sleep problems, indicating the need for

behavioural management strategies (Blampied, 2013a; Blampied & France, 1993). These reasons apply generally to pediatric sleep disturbance, and specifically to children on the autism spectrum.

This chapter begins with brief consideration of melatonin as a frequently prescribed pharmacological treatment to manage sleep disturbance in children on the autism spectrum. This chapter then reviews the behavioural interventions available for treating sleep problems in children on the autism spectrum, and the evidence-base for these treatments[1]. Following this, consideration is given to 'non-traditional' (i.e., non-behavioural and non-pharmacological) therapies (with a specific focus on white noise), because research suggests that many parents are interested in and employ alternative therapies to treat sleep problems in children on the autism spectrum (Buckley et al., 2020; Cuomo et al., 2017; McLay et al., 2020). Finally, the importance of using FBA to inform BSI for children on the autism spectrum is discussed.

Pharmacological Treatment: Melatonin

It is important to note that despite best practice recommendations, pharmacological treatments are frequently prescribed instead of behavioural interventions for the management of sleep problems in children, particularly those with DD (J. Owens et al., 2010; Pelayo & Dubrik, 2008; Rigney et al., 2018; McLay, Schluter, et al., 2021; Singh & Zimmerman, 2015). In a recent survey of 244 parents of children on the autism spectrum in NZ, medication was reported by parents to be the most commonly used strategy to treat sleep problems and was perceived by parents to be highly effective (McLay et al., 2020). There are several reasons why medication may be used instead of behavioural interventions, including that parents may be unable to access behavioural intervention services (e.g., owing to cost,

[1] Behavioural interventions available for the treatment of stereotypy, and the potential application of such strategies to the treatment of sleep problems, are discussed in Chapter 5.

local unavailability, or travel requirements), and inadequate training for health professionals who advise on service care. Further, behavioural treatments may be viewed by medical practitioners as falling outside a medical scope of practice (Cuomo et al., 2017; McLay et al., 2020; Pelayo & Dubik, 2008; Rigney et al., 2018). Medication is also less effortful and complex than behavioural interventions for parents to implement. This may be important if parent coping is diminished (e.g., owing to stress, fatigue, depression) and/or if parents do not have the necessary support (e.g., a solo parent with other children), particularly when the child has a DD (Carnett et al., 2020; Meltzer, 2016).

Of the pharmacological treatments available, exogenous melatonin, a synthetic version of the endogenous hormone implicated in the regulation of sleep/wake cycles, is frequently prescribed for children on the autism spectrum (Buckley et al., 2020; Pattison et al., 2020). This includes in NZ, where almost 25% of children on the autism spectrum have been dispensed melatonin (McLay, Schluter, et al., 2021). Among children reported on in this thesis, 3/12 children were taking melatonin as a hypnotic medication at the time of their involvement in this study. Melatonin is typically prescribed in low doses beginning at 1 mg and is typically taken 30 min before going to bed (Buckley et al., 2020; Singh & Zimmerman, 2015). Melatonin can effectively reduce SOD and improve TST in children on the autism spectrum (Buckley et al., 2020; Cuomo et al., 2017; Gringas et al., 2017; McLay, Schluter, et al., 2021; Pattison et al., 2020). However, there is limited evidence that melatonin can effectively reduce other sleep problems, including bedtime resistance, NWs, and co-sleeping (Carnett et al., 2020; Cuomo et al., 2017; Singh & Zimmerman, 2015), highlighting the importance of behavioural techniques where such sleep problems exist.

Review of Behavioural Sleep Interventions

Behavioural interventions are based on operant learning principles (as discussed in Chapter 1; Blampied & France, 1993; Meltzer & Mindell, 2014), and address sleep problems

by: (a) modifying the environment and/or instructional context to create a more sleep-conducive environment and improve stimulus control for sleep; (b) manipulating motivational variables to ensure a child's physiological state at bedtime is sleep-conducive (e.g., by inducing a non-stimulated state, with sufficient sleep pressure); and/or (c) disrupting the contingency between sleep-interfering behaviour and its reinforcers (K. Turner & Johnson, 2013; McLay, France, Blampied, van Deurs, et al., 2021; Pattison et al., 2020; Rigney et al., 2018). Behavioural interventions may be particularly well suited to children who have cognitive and/or language difficulties as they use non-verbal methods to alter behaviour (K. Turner & Johnson, 2013; Kodak & Piazza, 2008; Richdale & Wiggs, 2005).

A variety of behavioural intervention strategies are available for the treatment of sleep problems in children on the autism spectrum (Scantlebury et al., 2018). These interventions are predominantly the same types of interventions used to treat sleep problems in TD children (K. Turner & Johnson, 2013; Rigney et al., 2018; Vriend et al., 2011; Wiggs & France, 2000). In comparison to research among TD children, however, substantially fewer studies have been conducted investigating the efficacy of BSI for children on the autism spectrum (K. Turner & Johnson, 2013; Rigney et al., 2018; Vriend et al., 2011).

The following section provides an overview of research investigating the effectiveness of BSI for children on the autism spectrum. Given that sleep problems occur overnight in the home setting, a fundamental requirement of behavioural intervention strategies is that they are feasible for in-home implementation by parents (K. Turner & Johnson, 2013). As such, studies that focussed on parent-implemented sleep interventions were the focus of this review. The aims of this review were to systematically identify the types of behavioural intervention strategies that are available for the treatment of sleep problems in children on the autism spectrum, and the evidence-base for these strategies. The findings of this review, along with the findings from the evidence-based treatment literature

for stereotypy (Chapter 5), were used to inform the interventions that were selected for use with study participants reported in this thesis.

Methods

Inclusion Criteria

Studies were included if they: (a) included at least one participant with a formal diagnosis of ASD (including Asperger syndrome or pervasive developmental disorder; APA, 2013); (b) included participants aged $\leq$ 18 years (henceforth referred to as 'children'); (c) treated sleep problems using interventions that were based on the principles of applied behaviour analysis (ABA; e.g., stimulus control, graduated extinction); (d) reported sleep treatment outcomes (e.g., SOD, NWs or co-sleeping) as a dependent variable; (e) were published in a peer-reviewed journal; and (f) were published in English. Studies were excluded from the review if they did not directly treat sleep problems, or if the treatment was not based on the principles of ABA (e.g., medical or pharmacological approaches). Alternative strategies that have only been implemented in an inpatient facility (e.g., chronotherapy) were excluded.

Cognitive strategies (e.g., addressing an individual's sleep-related beliefs) have been more recently utilised alongside behavioural intervention components in some studies (e.g., McCrae et al., 2020; van Deurs et al., 2019). Cognitive strategies necessarily require that an individual possesses sufficient cognitive and language skills including metacognition (i.e., an ability to think about one's own thinking), causal reasoning, and receptive and expressive language ability. These skills may not have developed in children on the autism spectrum, particularly young children, and those with limited cognitive ability and/or limited/no-verbal ability (Lickel et al., 2012). This thesis includes young child participants, including children with limited or no verbal ability; therefore, cognitive strategies are not examined within this

thesis. Due to a lack of extant research, no restriction was placed on study design nor date of publication. Eligible articles included children with co-occurring diagnoses (e.g., ADHD) and children with and without ID.

Search Strategy and Study Selection

A systematic search of the following databases was undertaken: Education Resources Information Center (ERIC), Education Research Complete, PsycARTICLES and PsycINFO. The search combined diagnostic ("autism", "autism spectrum disorder", "autistic", "ASD", "Asperger", "pervasive developmental disorder", neurodevelopmental disorder") sleep ("sleep", "sleep problems", "sleep disturbance", "sleep difficulties", "insomnia") and treatment ("treatment", "intervention", "therapy", "management", "training") related terms. In addition to database searching, ancestral searching was conducted to identify relevant articles that were not otherwise identified.

Results

A wide range of antecedent- and consequence-based procedures have been used to effectively reduce sleep problems in children on the autism spectrum. Antecedent-based strategies identified during this search included sleep hygiene practices, including positive bedtime routines and parent education (e.g., regarding healthy sleep), modification to sleep/wake schedules, sleep restriction and the faded bedtime procedure, and the use of social stories. Consequence-based strategies included extinction procedures, and modified extinction procedures including graduated extinction, the minimal check procedure, the parental presence procedure, and the bedtime pass, as well as the use of positive reinforcement. In addition, one strategy, systematic fading of parental presence, involves modification of stimulus control and extinction; however, as the stimulus control modification operates concurrently with the contingency modification (i.e., extinction) it was

considered primarily a consequence-based procedure. The procedures and evidence-base for these intervention strategies are discussed below.

Antecedent-Based Interventions

Antecedent-based procedures involve modification to the environment or instructional context as experienced prior to the target behaviour(s), to improve stimulus control for sleep and reduce the likelihood of problem behaviour occurring subsequently. Antecedent interventions also include procedures that manipulate the MOs for sleep (i.e., to increase motivation for sleep, and/or decrease motivation for engagement in sleep-interfering behaviour).

Sleep Hygiene. As discussed in Chapter 1, difficulties with sleep onset and maintenance commonly have a behavioural basis, which can be affected by one's behaviour during the day as well as in the lead up to going to bed (Blampied & France, 1993; Mindell et al., 2009). Poor sleep habits, such as late or inconsistent bedtimes, the presence of entertainment devices in the bedroom, and caffeinated food or beverages at night, have a negative impact on sleep quality and can contribute to the presence of sleep problems (Jan et al., 2008; Malow et al., 2014). Research has consistently shown that good sleep hygiene, a term which describes "*modifiable parent and child practices that promote good sleep quality, allow sufficient sleep duration, and prevent daytime sleepiness"* (Mindell et al., 2009, p.771), is an important contributor to quality sleep across the lifespan (Jan et al., 2008; Mindell et al., 2009; Spruyt & Curfs, 2015; Tan et al., 2012).

Sleep hygiene involves specific recommendations across a number of domains, including maintaining a consistent sleep schedule (i.e., a set bed and wake time, including age-dependent naps where appropriate); maintaining a consistent, calming bedtime routine that avoids stimulating activities and electronic-device use before bed; minimising the intake

of caffeine, alcohol and large amounts of food before bed; and having an optimal sleep environment (e.g., dark room, pleasant sleeping temperature [thermal comfort], low noise level, comfortable bedding; Hauri, 2011; Jan et al., 2008; Mindell et al., 2009; Richdale & Schreck, 2019; Singh & Zimmerman, 2015; Vriend et al., 2011). Of these recommendations, the importance of a consistent bedtime routine has garnered the most empirical attention and support (Mindell et al., 2009; Richdale & Schreck, 2019). Clear and consistent routines are likely to help to entrain internal sleep rhythms to external zeitgebers, thereby bringing the behaviour of falling asleep under stimulus control of both exteroceptive (external) and interoceptive (internal) stimuli (Blampied & France, 1993; Jan et al., 2008; Richdale, 1999; Schreck, 2001; Singh & Zimmerman, 2015; Spruyt & Curfs, 2015).

Sleep hygiene practices should be assessed and addressed as a foundational step within any BSI (Jan et al., 2008). Healthy sleep hygiene practices provide a basis of appropriately timed and effective sleep without which other intervention strategies are more likely to fail (Jan et al., 2008; Tudor et al., 2012). Evidence suggests, however, that sleep hygiene alone is unlikely to effectively resolve sleep problems (Adkins et al., 2012; Jan et al., 2008). For example, Adkins et al. (2012) investigated the effectiveness of a pamphlet containing sleep hygiene information given to parents of children on the autism spectrum on a primary outcome measure of sleep onset latency. Parents of 36 children on the autism spectrum were randomised to receive the information, or to a control group who did not receive sleep hygiene information. Results showed no significant differences between groups, and Adkins et al. (2012) concluded that sleep information, while necessary for parents, is insufficient in isolation to alter sleep behaviour in children on the autism spectrum. In another study, Delemere and Dounavi (2018) investigated the effects of a consistent bedtime routine in isolation from other treatment strategies, on sleep outcomes in three children (aged 2-6

years) on the autism spectrum. Results showed that the bedtime routine alone did not significantly reduce sleep problems (Delemere & Dounavi, 2018).

Parent education regarding healthy sleep practices is an important aspect of sleep hygiene, as poor sleep practice may be the result of inadequate parental knowledge about sleep-promoting factors, or unrealistic expectations (Adkins et al., 2012; Malow et al., 2014; Weiss et al., 2006). For example, a parent who is unaware that physical stimulation can make sleep onset more difficult may engage in high-energy games with their child prior to bedtime, rather than calming activities that promote behavioural quietude (Malow et al., 2014; Weiss et al., 2006). Sleep hygiene including parent education has been widely incorporated into BSI for children on the autism spectrum, and its use in intervention is strongly endorsed by research (Cortesi et al., 2010; Knight & Johnson, 2014; Kodak & Piazza, 2008; Malow et al., 2014; Singh & Zimmerman, 2015; Vriend et al., 2011). Parent education about sleep hygiene is a particularly important consideration within treatment for children on the autism spectrum, given their extraordinarily high rates of sleep problems (Jan et al., 2008; Richdale & Schreck, 2019) and should be a routine component of intervention advice.

Sleep/Wake Scheduling. As previously discussed in Chapter 1, sufficient motivation by way of sleep pressure is necessary for a child to successfully initiate and maintain sleep. Sleep pressure functions as an MO in the form of an EO, as it increases the value of sleep as a reinforcer and an individual's motivation to engage in 'falling asleep' behaviour. In turn, this also affects the salience and strength of the S^D that signal the reinforcer of sleep is available. It is important that parents maintain a consistent and age-appropriate sleep/wake schedule for children as a necessary part of sleep hygiene practice, to ensure children are physiologically ready for sleep when they go to bed. In addition, parents must restrict children's sleep to occur between their set bed and wake time (including any age-appropriate naps) since allowing a child to sleep during the day (e.g., sleeping in, age-inappropriate naps) can

decrease pressure for sleep the following night (i.e., an AO), and contribute to sleep difficulties.

The age-appropriate timing and duration of children's sleep can be determined from evidence-based recommendations, such as in accordance with The National Sleep Foundation (Hirshkowitz et al., 2015; Ohayon et al., 2017). For example, The National Sleep Foundation recommend that school-age children (6-13 years) receive 9-11 hours of TST (Hirshkowitz et al., 2015; Ohayon et al., 2017). These recommendations apply generally to TD children, with an absence of evidence to suggest that the sleep needs of children on the autism spectrum are any different. Modifications to children's sleep/wake schedule may involve the restriction or elimination of daytime naps, and the establishment or adjustment of bed and wake times. For example, children may have a set bed and wake time that is not consistently maintained (e.g., being allowed to sleep in on weekends/holidays). Modifications to a child's sleep/wake schedule, and the restriction of sleep to occur between these times, can enhance sleep pressure and improve stimulus control for sleep, by reducing the amount of time spent in bed awake (Christodulu & Durand, 2004; Vriend et al., 2011).

Sleep Restriction: The Faded Bedtime Procedure. For children who have an age-appropriate and consistently maintained sleep schedule but spend extended periods of time in bed awake (e.g., having SOD, NWs), then further sleep restriction procedures may be needed to increase motivational sleep pressure and improve stimulus control for sleep. The faded bedtime procedure is a sleep restriction method derived from Stimulus Control Therapy (Bootzin, 1977) for adult insomnia. The faded bedtime procedure involves delaying the child's bedtime to be within 15 min of the child's typical sleep onset (K. Turner & Johnson, 2013; Kodak & Piazza, 2011; Piazza & Fisher, 1991a; 1991b; Piazza et al., 1997; Vriend et al., 2011). In addition, a set wake time is established, and daytime sleep is eliminated, in order to increase motivational sleep pressure (Piazza & Fisher, 1991a; 1991b). The child

should continue to receive an age-appropriate total duration of sleep during this procedure (e.g., as per National Sleep Foundation recommendations; Hirshkowitz et al., 2015; Luiselli et al., 2021; Ohayon et al., 2017).

Once the child is regularly and efficiently falling asleep at the delayed bedtime, then the bedtime can be systematically moved earlier in small increments (e.g., 15-30 min) until a more desirable bedtime is reached, while maintaining an age-appropriate TST (Kodak & Piazza, 2008; 2011; Richdale & Wiggs, 2005). The faded bedtime can also be implemented with a 'response cost', whereby if the child does not fall asleep within 15 min of going to bed, they are removed from bed to engage in a non-stimulating activity (e.g., reading in the lounge) for a short amount of time, before being returned to bed (Richdale & Wiggs, 2005; Vriend et al., 2011; see also Bootzin, 1977). This procedure can be repeated until sleep onset occurs within 15 min (Richdale & Wiggs, 2005; Vriend et al., 2011). The faded bedtime (with or without a response cost) is thought to induce mild sleep deprivation (i.e., increase homeostatic sleep pressure) and consequently (temporarily) increase the value of sleep, and the chain of behaviours leading to this reinforcer (Luiselli et al., 2021; Michael, 1982; 1993; Piazza & Fisher, 1991a; 1991b). It also improves stimulus control by strengthening the bedroom environment as S^D for sleep (i.e., via reduced time spent in bed awake; Bootzin, 1977; Piazza & Fisher, 1991a; 1991b).

Sleep restriction is a routine treatment for sleep problems in TD individuals with a well-established evidence base (Maurer et al., 2021). Sleep restriction is also an evidence-based treatment for sleep problems in individuals with ID (Didden et al., 2014; Piazza et al., 1997; Piazza & Fisher, 1991b). Compared to the literature for TD children, far fewer studies have examined the efficacy of the faded bedtime procedure to treat sleep problems in children on the autism spectrum (Luiselli et al., 2020). For children on the autism spectrum, the faded bedtime procedure has been used with (Moon et al., 2011; Sanberg et al., 2018) and without

(Delemere & Dounavi, 2018; Jin et al., 2013; McLay, France, Knight, et al., 2019) a response cost. For example, Moon et al. (2011) evaluated the effects of a faded bedtime procedure with response cost and positive reinforcement to treat SOD in three children on the autism spectrum aged 8-9 years. Parents were provided with a treatment handbook describing the treatment procedures and were contacted weekly by phone call with a therapist. For all three children, SOD reduced following treatment, and gains were found to be generally maintained at a 12-week follow-up. In addition, parent-reported satisfaction with treatment was high (Moon et al., 2011).

Delemere and Dounavi (2018) compared the efficacy of the faded bedtime procedure without a response cost to sleep hygiene (positive bedtime routines) to treat sleep problems including SOD, frequency and duration of NWs, and TST, in six children aged 2-7 years on the autism spectrum, using two multiple baseline designs. Both interventions demonstrated some efficacy at reducing sleep difficulties; for children who received the faded bedtime intervention, improvements were found in SOD and in TST. Social validity and treatment acceptability outcomes for both procedures were high (Delemere & Dounavi, 2018).

A case report by Luiselli et al. (2021) describes the effects of the faded bedtime procedure on SOD in a 14-year-old boy on the autism spectrum in a residential-care setting. The boy's bedtime was delayed by one hour from his average time to sleep onset, determined during a baseline phase. A visual schedule depicting leisure (e.g., a craft activity) and household tasks (e.g., folding laundry) based on adaptive-living learning objectives was provided to enable the boy to choose activities to do during the extended pre-bedtime period. In result of intervention, SOD was reduced and TST was increased, with a further improvement in results evident at a 2-month follow-up (Luiselli et al., 2021).

Other studies have utilised the faded bedtime procedure within multicomponent BSI for children on the autism spectrum (Jin et al., 2013; McLay, France, Knight, et al., 2019;

Sanberg et al., 2018; van Deurs et al., 2019). Results of these studies show that the interventions were largely effective at reducing sleep problems; however, the contribution of the faded bedtime procedure is difficult to determine owing to the multicomponent nature of the interventions (Luiselli et al., 2020). Overall, there is an emerging evidence-base for the faded bedtime procedure to treat sleep problems in children on the autism spectrum, and studies suggest that this procedure may be effective at treating multiple sleep problems simultaneously (e.g., SOD and NWs; Piazza et al., 1997; Piazza & Fisher, 1991a; 1991b; Vriend et al., 2011).

Similar to the faded bedtime procedure, two studies (Christodulu & Durand, 2004; Durand & Christodulu, 2004) investigated the efficacy of sleep restriction to reduce sleep problems including bedtime resistance and NWs in children; participants on the autism spectrum included two girls and a boy aged 2-4 years. Although the procedure included bedtime fading, the primary focus was on TST rather than the set bedtime and involved restricting children's sleep to 90% of their baseline TST (Christodulu & Durand, 2004; Durand & Christodulu, 2004). One study (Christodulu & Durand, 2004) employed sleep hygiene practices alongside sleep restriction. The results of each study showed a reduction in bedtime resistant behaviour and NWs. For one child, however, parasomnias (e.g., sleepwalking and night terrors) increased (Durand & Christodulu, 2004). Further methodologically stronger research needs to establish the efficacy of this procedure for the treatment of sleep problems in children on the autism spectrum (Vriend et al., 2011).

Social Stories. Social stories are stories that present a narrative in pictures and/or text, which teach a child an appropriate behavioural sequence in a specific situation (e.g., toilet training; Gray & Garand, 1993). Social stories have a positive emphasis on what the child should do, rather than what the child should not do, and unambiguously represent the sequence of behaviours from the child's perspective (Gray & Garand, 1993). Social stories

are consistent with the principles of observational learning (Bandura & Walters, 1963), and provide a means of introducing information that a child can refer to repeatedly and consume at their own pace (K. Turner & Johnson, 2013; P. Moore, 2004). They may be helpful in part because they make explicit the nonverbal messages of others, providing a more literal interpretation of situations that may otherwise be confusing for a child on the autism spectrum to understand (K. Turner & Johnson, 2013; P. Moore, 2004; Test et al., 2011). Traditionally social stories are implemented on paper, but variations have been used throughout the research literature, including the use of digitally mediated social stories (Hanrahan et al., 2020; Singh & Zimmerman, 2015; Test et al., 2011).

Social stories were originally developed to improve social skills in children on the autism spectrum, but as they are easily individualised, they have been used to teach children a range of skills in a variety of contexts, particularly within education (Hanrahan et al., 2020; Kurt & Kutlu, 2019; P. Moore, 2004; Swaggart et al., 1995). This includes teaching sleep-conducive behaviour (e.g., following a bedtime routine) to TD children and children on the autism spectrum (e.g., Burke et al., 2004; Malow et al., 2014; McLay, France, Knight, et al., 2019; P. Moore, 2004). For example, Burke et al. (2004) used a social story coupled with positive reinforcement to effectively decrease disruptive bedtime behaviour and NWs in four TD children aged of 2-7 years. Malow et al. (2014; described below) incorporated a social story as part of a wider BSI to enhance understanding and use of a bedtime pass for the children on the autism spectrum aged 2-10 years. Social stories have high social validity when used by parents in a family setting (P. Moore, 2004; Test et al., 2011). They may assist BSI strategies by supplying modelled examples of behaviour, increasing children's understanding of sleep-conducive behaviour (e.g., the steps in a bedtime routine, lying quietly in bed) and/or the use of other intervention strategies (Cuomo et al., 2017; Malow et al., 2014; Pattison et al., 2020; Reynolds & Malow, 2011; Singh & Zimmerman, 2015).

Consequence-Based Interventions

Consequence-based procedures disrupt the relationship between sleep-interfering behaviour and its reinforcer(s) or may be used to strengthen the performance of sleep-conducive behaviour (e.g., compliance with a bedtime routine).

Extinction Procedures. Extinction is both a process and a procedure. Extinction is a procedure that reduces the frequency or strength of a problem behaviour (the process), by permanently revoking reinforcement maintaining the problem behaviour (the procedure; Blampied & France, 1993; Lawton et al., 1991; Lerman & Iwata, 1995; Lerman et al., 1999). When a response no longer produces reinforcement then the occurrence of that behaviour is reduced, if not eliminated (Lerman & Iwata, 1995). In other words, the process of extinction is to weaken behaviour (i.e., make it less likely to occur) as a result of exposure to the procedure of extinction.

When used in the sleep context, an extinction procedure involves the withdrawal of reinforcement for a previously reinforced problem (sleep-interfering) behaviour. Thus, the procedure involves modification to the environment (i.e., revoking access to reinforcement) to disrupt the contingency between sleep-interfering behaviour and its reinforcers (McLay, France, Blampied, van Deurs, et al., 2021; Rigney et al., 2018; Wiggs & France, 2000). As the process of extinction takes effect, sleep-interfering behaviour becomes less likely to occur, or is eliminated.

Extinction procedures are frequently used to address difficulties with sleep onset and maintenance, specifically when sleep-interfering behaviours are being reinforced at these times (Schreck, 2001; Richdale, 2013; Wiggs & France, 2000). For example, a child who calls out to parents (i.e., CCs) after being bid goodnight may struggle to fall asleep because calling out is a behaviour incompatible with falling asleep and calling out results in

experiencing the reinforcing consequence of parent attention. If the parent purposefully withholds their attention (known as 'planned ignoring'), then call-out behaviour is placed on extinction, and the behaviour will decrease (Didden et al., 2002; Montgomery et al., 2004; Vriend et al., 2011; Weiskop et al., 2001; 2005) making falling asleep more likely.

Extinction procedures are well-established as an effective treatment for sleep problems, including SOD, and frequency and duration of NWs, in TD individuals, as well as children with DD including those on the autism spectrum (Carnett et al., 2020; France & Blampied, 2005; K. Turner & Johnson, 2013; L. Owens et al., 1999; McLay, France, Blampied, van Deurs, et al., 2021; Meltzer & Mindell, 2014; Richdale, 2013; Schreck, 2001; Thackeray & Richdale, 2002; Vriend et al., 2011; Weiskop et al., 2001; 2005). For children on the autism spectrum, extinction procedures are often implemented as part of a multicomponent sleep intervention. Such interventions often include antecedent strategies that enhance sleep onset and maintenance (e.g., sleep hygiene, sleep/wake scheduling) as well as teaching replacement behaviours (e.g., lying quietly in bed) that support sleep (Deliens et al., 2015; Freidman & Luiselli, 2008; K. Turner & Johnson, 2013; Meltzer & Mindell, 2014; Singh & Zimmerman, 2015; Vriend et al., 2011).

Weiskop et al. (2001) carried out a case study with a 5-year-old boy on the autism spectrum, with settling, co-sleeping and NW difficulties. Intervention consisted of parent-training workshops based on behavioural principles, where parents learnt positive bedtime routine practices, reinforcement and communication strategies, partner support, and how to implement an extinction procedure (Weiskop et al., 2001). Parents learnt to implement an extinction procedure by ignoring any requests or demands for attention, and to return the boy to his bedroom immediately with minimal interaction if he left his bed (Weiskop et al., 2001). Furthermore, as the boy shared a bedroom with a brother, the brother also learnt to ignore the boy's bids for attention and not to call out to parents himself (Weiskop et al., 2001). Parents

were able to successfully implement treatment, which resulted in the boy learning to self-settle and sleep independently, and results were maintained at a 3- and 12-month follow-up (Weiskop et al., 2001).

A later study by the authors (Wieskop et al., 2005) evaluated the efficacy of the same treatment programme with six children on the autism spectrum and seven with Fragile X Syndrome; again, problems of settling, NWs and co-sleeping were effectively reduced and maintained at follow-up. Weiskop et al. (2005) reported that parents rated treatment as socially acceptable, and that parent-set goals were largely achieved.

Montgomery et al. (2004) examined the effects of a brief behavioural treatment for 66 children (aged 2-8 years) with learning disabilities, 21 of whom had a diagnosis of autism, and sleep difficulties (settling problems and/or NWs). Treatment consisted of an extinction programme, where all parents were instructed to ignore any demands for attention, in combination with other behavioural approaches including sleep hygiene, graduated extinction (described below), consistent resettling strategies and a positive reward system. The study compared whether treatment was more effective when delivered via a booklet, or by face-to-face consultation with a researcher. Results showed that both modes of delivery were effective in changing the sleep habits of children in the study, and gains were maintained at a 6-month follow-up assessment (Montgomery et al., 2004).

In another study, Papadopoulos et al. (2019) used a multimodal treatment approach that combined extinction techniques (i.e., planned ignoring of unwanted behaviour) with other behavioural techniques including parental presence, bedtime fading and a bedtime pass procedure (described below) to treat sleep problems in 61 children aged 5-13 years with co-occurring diagnoses of autism and ADHD. Significant improvements in sleep outcomes were reported as a result of the brief multimodal intervention, with gains found to be maintained at a 3- and 6-month follow-up period (Papadopoulos et al., 2019).

One of the earliest accounts of an extinction procedure being used to treat sleep problems in a child on the autism spectrum is a study by Wolf, Risley and Mees (1963), who treated bedtime resistant (severe tantrum) behaviour and NWs in a 3.5-year-old boy, in both an inpatient ward and the home setting. The procedure involved inpatient attendants or parents closing the boy's bedroom door dependent on him displaying tantrum behaviour or leaving his bed. Door closure eliminated the social interaction that reinforced tantrum behaviour and decreased the value of the boy's signalling behaviour if he woke (Wolf et al., 1963). Results showed that the intervention was effective at reducing sleep problems, and outcomes were maintained at a 6-month follow-up (Wolf et al., 1963). An undesirable side effect of the procedure, however, was that the severity and duration of tantrum behaviour increased during the first five nights of treatment, before resolving on the sixth night (Wolf et al., 1963). This effect, known as an extinction burst, is a common side effect of extinction procedures whereby the undesirable behaviour can increase in intensity, duration, and/or frequency, before it improves (Lerman & Iwata, 1995; Lerman et al., 1999). An extinction burst is most likely to occur during initial treatment stages when the previously reinforced response no longer produces reinforcement (France et al., 1996; Lawton et al., 1991).

While an extinction burst is a predictable side effect of extinction procedures (Lerman & Iwata, 1995), its occurrence can make treatment difficult for parents to adhere to. It may be hard for parents to withhold attention from a child who is expressing intense upset, and to remain consistent in the management of difficult behaviour (France & Blampied, 2005; Kodak & Piazza, 2008; Lerman & Iwata, 1995; Singh & Zimmerman, 2015; Thackeray & Richdale, 2002; Vriend et al., 2011). Parents commonly experience stress and unease in response to a child's emotional and behavioural outbursts, which can increase the likelihood of parents abandoning the procedure in order to attend to their child (Didden et al., 2002; Kodak & Piazza, 2008; Lawton et al., 1991; Singh & Zimmerman, 2015; Weiskop et al.,

2005). Unfortunately, if a parent does attend to their child during an extinction burst, the child's behaviour is inadvertently reinforced at its intensified level, making it more likely that the problem behaviour will reoccur at a heightened level, and the behaviour becomes more resistant to change (Freeman, 2006; Kodak & Piazza, 2008; Lawton et al., 1991).

In addition, challenging behaviours (e.g., aggression, self-injury) are common in children on the autism spectrum, and care must be taken to ensure that any burst in behaviour does not pose a safety concern for the child or other household members (Hanley et al., 2014; Kodak & Piazza, 2008; Lerman et al., 1999; Singh & Zimmerman, 2015). Thus, while extinction procedures can be effective, and there is no evidence of long-term detrimental effects of such procedures, careful consideration on the part of both the parent and therapist is needed to mitigate the potential pragmatic challenges and undesirable effects (Didden et al., 2002; France & Blampied, 2005; France et al., 1991; K. Turner & Johnson, 2013; Kodak & Piazza, 2008; Price et al., 2012; Richdale, 2013).

As an alternative approach, many studies have examined the effects of modified extinction procedures, with the aim of retaining the effective properties of extinction but which enhance treatment acceptability and adherence (B. Moore et al., 2007; Blampied, 2013a; France & Hudson, 1993; K. Turner & Johnson, 2013; Lawton et al., 1991; Vriend et al., 2011). It is important to note, however, that extinction procedures may be acceptable to some parents and able to be implemented with fidelity. For example, Weiskop et al. (2005; described above) reported that parents in their study rated BSI including extinction procedures as socially acceptable and effective. This highlights the importance of determining and including parent's preferences within the design of BSI, which is discussed further below.

Modified Extinction Procedures. In contrast to unmodified extinction where reinforcement maintaining a problem behaviour is irrevocably revoked from the outset,

modified extinction procedures involve the progressive withdrawal of reinforcement over time (Ferber, 1985; Lawton et al., 1991; Singh & Zimmerman, 2015; Wiggs & France, 2000). Modified procedures used within sleep interventions include graduated extinction and minimal check procedures, the parental presence procedure, and the bedtime pass. Within each of these procedures, the rate or intensity of reinforcement is gradually rather than abruptly reduced, whilst the process of extinction remains the same (i.e., the effect of modified extinction is to weaken a target behaviour; Lawton et al., 1991; Singh & Zimmerman, 2015; Wiggs & France, 2000). Systematic fading of parental presence is discussed alongside the parental presence procedure, although it is important to note that the modified component of this procedure is on stimulus control (i.e., from parent-provided S^D to bed-related S^D), rather than extinction.

Graduated Extinction. Graduated extinction involves systematic changes in the baseline contingency of reinforcement (i.e., the rate, quantity, or immediacy of reinforcement via parent attention or access that a child has to reinforcing items or activities) so that ultimately no reinforcement is delivered. For example, in the case of parent attention maintaining sleep problems (e.g., SOD and/or NWs), graduated extinction involves parents reducing the frequency, duration and/or quality of their interactions after bidding the child goodnight, so that only a minimal amount of reassurance is provided (e.g., the parent briefly restores the child's sleep position without verbal interaction). The level of attention provided is systematically reduced over a course of nights, until the child learns to fall asleep without parental attendance (Lawton et al., 1991; Schreck, 2001; Wiggs & France, 2000). This may be achieved by parents waiting successively longer each interval before attending (i.e., the Ferber procedure; Ferber, 1985), or by parents reducing the duration of their attention each time they attend (i.e., the Lawton procedure; Lawton et al., 1991), or both.

Minimal Check. Within a minimal check procedure, parent-child interaction is placed on a time-based schedule, wherein parents attend to their child at set intervals (e.g., every 5 min) and withhold their attention (i.e., planned ignoring) in between these checks. When checking on their child, parents use the minimal amount of attention required to attend to their child, before leaving the room. The interval of time between each check is systematically increased (e.g., 5 min between checks is increased to 10 min), until no reinforcing attention is provided (France & Blampied, 2005; K. Turner & Johnson, 2013; Kodak & Piazza, 2008; L. Owens et al., 1999). In effect, this procedure modifies the contingency of reinforcement, the amount of reinforcement (large to minimal), and the frequency of reinforcement (the time between checks is systematically increased). Eventually, in graduated extinction and minimal check procedures, parent attention is revoked altogether, and (unmodified) extinction occurs, thereby strengthening the process.

Graduated extinction procedures are routinely used to treat sleep problems in TD children and have strong empirical support (Lawton et al., 1991; P. Moore, 2004). Graduated extinction procedures have also been used extensively within BSI for children on the autism spectrum throughout the research literature (e.g., Knight & Johnson, 2014; Malow et al., 2014; Montgomery et al., 2004; Reed et al., 2009), with an increasing evidence-base (Durand et al., 1996; K. Turner & Johnson, 2013; Richdale, 2013; Vriend et al., 2011).

For example, Moss, Gordon and O'Connell (2014) evaluated the effects of a programme called Sleepwise (O'Connell & Vannan, 2008), a group-based intervention involving sleep hygiene practice, graduated extinction, relaxation techniques and positive reinforcement, on sleep outcomes in 26 youths (15 on the autism spectrum), compared to a waitlist control. Parents were provided with information in a workshop setting regarding how to implement graduated extinction, including gradual distancing and planned ignoring with parental presence. Results showed an overall decrease in children's sleep disturbance in the

treatment group, and a reduction in levels of parent self-reported stress post-treatment (Moss et al., 2014).

Durand et al. (1996) used a graduated extinction procedure combined with sleep hygiene practice to reduce settling problems, NWs and co-sleeping in four children with DD, two of whom were on the autism spectrum. Parent attention in response to children's call out behaviour after children were bid goodnight was progressively reduced over subsequent nights throughout intervention, by systematically increasing the duration of time before parents attended to their child when in bed (e.g., waiting 3 min one night, then 5 min the next night before responding [i.e., the Ferber procedure]; Durand et al., 1996). Treatment successfully reduced settling problems, NWs and co-sleeping, and outcomes were maintained at a 6-month follow-up (Durand et al., 1996). Treatment adherence and parent satisfaction with treatment were also high (Durand et al., 1996).

In a study by Jin et al. (2013; described further below), a minimal check procedure was used to disrupt the reinforcing contingency of parent attention maintaining sleep interfering behaviour in a 9-year-old boy on the autism spectrum. After bidding the boy goodnight, parents visited him on a time-based schedule independent of his behaviour. Checks occurred frequently within the first three nights, with increasing periods between each check (i.e., 5 s, 10 s, 30 s, 1 min, 5 min, 10 min, 30 min). The frequency of checks was then systematically reduced by eliminating the first check every second night. Parent interaction during each check was kept to a minimum; parents restored the boy's sleep position and bid him goodnight while maintaining minimal language, eye contact, and a neutral facial expression. The minimal check procedure was one component within an individualised, multimodal intervention, which effectively improved the boy's sleep.

The Parental Presence Procedure. As discussed in Chapter 1, the presence of a parent (e.g., lying with a child at bedtime), with their highly salient S^D, can make it difficult

for a child to initiate and/or re-initiate sleep when the parent is absent because of inappropriate stimulus control, and is associated with sleep problems including increased and/or prolonged NWs in children of all ages (Mindell et al., 2009). While some parents place value on being present when their child falls asleep and other types of co-sleeping arrangements (Goldberg & Keller, 2007), for other parents this can occur in a reactive manner, as a strategy to cope with sleep difficulties (e.g., as a means to quickly resettle their child back to sleep; Liu et al., 2006). For children who are not able to self-settle to sleep (i.e., without parental assistance), and for parents for whom this is a problem, a parental presence procedure may be recommended (Bathory & Tomopoulos, 2017; Blampied & France, 1993; Burnham et al., 2002).

In the original parental presence procedure (Sadeh, 1994), once a child is put to bed the parent remains in the child's room, visible to the child, but in a separate bed or chair, and feigns sleep, until the child is asleep (France & Blampied, 2005; Sadeh, 1994). The parent may then sleep in the child's room overnight but does not engage with any of the child's disruptive behaviour unless necessary (e.g., to return the child to their bed, or for safety reasons; France & Blampied, 2005). Alternatively, the parent may leave the child's room (once the child is asleep) and sleep in their own bed, returning to the child's room if the child wakes. If the parent is required to respond to the child, interaction is kept to a minimum; for example, restoring the child's sleep position without verbal interaction (France & Blampied, 2005). The parent's presence in the child's bedroom is thought to continue to reassure the child and reduce the child's distress during the procedure, whilst simultaneously instituting extinction by removing or reducing parent-mediated reinforcement for sleep-interfering behaviours (France & Blampied, 2005; Sadeh, 1994).

After approximately one week, when the child is able to reliably fall asleep independently, the parent leaves the room as soon as the child is put to bed, so that the

presence of a parent is removed altogether (France & Blampied, 2005; Sadeh, 1994). In effect, the parental presence procedure is an unmodified extinction procedure that is supplemented by an additional procedure (i.e., the parent's presence) to mitigate distress that can be evoked by extinction, with the supplementary procedure withdrawn once a success criterion (e.g., one week of independent sleep onset) is achieved.

Systematic Fading of Parental Presence. With systematic fading of parental presence, the location where the parent waits for the child to fall asleep is gradually moved further away from the child's bed (i.e., towards the door) until the presence of the parent is fully eliminated (Vriend et al., 2011). For example, the parent may begin by sitting on the child's bed without providing any interaction while the child settles to sleep. Once the child is reliably falling asleep with the parent sitting on the bed, the parent may move to sitting on a chair close to the child's bed. This procedure is repeated, with the parent moving systematically further away from the child, until the parent can bid the child goodnight and immediately leave the room (Vriend et al., 2011). Systematic fading of parental presence constitutes an extinction procedure, because reinforcing parent attention (i.e., via interaction with the child) is revoked, and a stimulus control manipulation involving the gradual transfer of stimulus control from parent-provided S^D to bed-related S^D.

Studies show that a parental presence procedure can effectively reduce sleep problems in TD infants and children (France & Blampied, 2005; Mindell, 1999; Matthey & Črnčec, 2012). Only a few studies have employed this procedure (and systematic fading of parental presence) to treat sleep problems in children on the autism spectrum (Howlin, 1984; McLay, France, Knight, et al., 2019; Souders et al., 2017). Howlin (1984) used a stimulus fading technique to treat sleep problems (SOD, NWs and unwanted co-sleeping) in a 6-year-old boy on the autism spectrum. The parent's presence in the boy's bedroom was systematically

withdrawn, resulting in a reduction in SOD, NWs, and co-sleeping. In addition, parent-report indicated improvements in the martial relationship and maternal mood levels (Howlin, 1984).

More recently, McLay, France, Knight, et al. (2019) utilised systematic fading of parental presence within function-based BSI to address multiple sleep problems, including unwanted co-sleeping, in seven children on the autism spectrum. Initially, parents were instructed to sit on a chair next to the child's bed; the proximity of the parent to the child was then systematically faded over subsequent nights until the parent was out of the child's sight (McLay, France, Knight et al., 2019). During the procedure, parents were instructed not to engage with any of the child's attempts for attention, and to only respond with minimal interaction (e.g., returning the child to their bed without additional interaction) if necessary. Results showed improvements in sleep outcomes across all children, including the elimination of co-sleeping, with gains largely maintained at follow-up (McLay, France, Knight et al., 2019). In addition, parental satisfaction with treatment was high (McLay, France, Knight, et al., 2019). While the outcomes of studies utilising systematic fading of parental presence show promise (Howlin, 1984; McLay, France, Knight, et al., 2019; Souders et al., 2017), more research is needed to establish the efficacy of this procedure to reduce sleep difficulties in children on the autism spectrum (Vriend et al., 2011).

***The Bedtime Pass*.** A bedtime pass is a modified extinction procedure that can be used to help to reduce sleep-interfering behaviours (e.g., CCs) and encourage independent sleep (Reynolds & Malow, 2011). A bedtime pass is a small card or 'pass' that is given to a child each night, which can be exchanged for a permitted request (e.g., an extra cuddle) or trip out of the bedroom, following which a parent restores the child's sleep position and removes the pass (Freeman, 2006; Mindell & Owens, 2015). Once the pass is removed, all subsequent bids for parental attention are ignored (i.e., extinction; B. Moore et al., 2007; Freeman, 2006; Meltzer & McLaughlin Crabtree, 2015). The pass is often implemented with

a reward system whereby a child can earn rewards for appropriate use of the pass and/or settling to sleep without use of the pass (Malow et al., 2014). The bedtime pass therefore involves elements of both extinction and positive reinforcement (described below).

Since the bedtime pass was developed (Friman et al., 1999), studies have demonstrated its effectiveness in reducing bedtime resistant behaviours and SOD in TD children, with high social validity (e.g., B. Moore et al., 2007; Freeman, 2006; Friman et al., 1999). The findings of these studies suggest the pass may reduce the likelihood of an occurrence of an extinction burst, and improve parent adherence to extinction techniques (Freeman, 2006; Friman et al., 1999). The bedtime pass has also been used as part of wider behavioural strategies to reduce settling difficulties in children on the autism spectrum (Malow et al., 2014; Reed et al., 2009).

For example, in a study by Malow et al. (2014), 80 parents of children on the autism spectrum with sleep problems were taught graduated extinction techniques in combination with the use of a bedtime pass in parent sleep education workshops. The bedtime pass procedure followed that of Friman et al. (1999) but with a modification that children could exchange the pass for a reward in the morning if the pass had not been used during the night (Malow et al., 2014). In addition, the bedtime passes were individualised to reflect child preferences (e.g., a picture of the child's favourite movie character) and a social story was created to aid the child's understanding of how to use the bedtime pass. Results showed that the education workshops were associated with improved sleep outcomes, and parents reported an improved sense of parenting competency (Malow et al., 2014). Overall, research suggests that the bedtime pass may be a beneficial component of wider behavioural interventions to improve settling behaviour in children who have sufficient communicative ability, although the specific effects of the bedtime pass on sleep outcomes in children on the autism spectrum requires further research (Malow et al., 2014; Reed et al., 2009).

Overall, research suggests a modified extinction approach may be more suitable for children on the autism spectrum than unmodified extinction because it allows change to be introduced incrementally, which can minimise emotional and behavioural outbursts (i.e., reduces the risk of challenging behaviours) and supports a child's individual pace of learning (Kodak & Piazza, 2008; Richdale, 2013; Stores & Wiggs, 1998). Importantly, research suggests that parents may prefer modified procedures because they enable parents to continue to attend to their child during the night, and may be less emotionally demanding than unmodified extinction, particularly for parents without additional support (e.g., who are a solo care provider with other children; France & Hudson, 1993; K. Turner & Johnson, 2013; P. Moore, 2004; Pattison et al., 2020). Studies utilising modified extinction procedures within BSI for children on the autism spectrum largely report positive results; however, the contribution of modified extinction is often unclear, owing to the multicomponent nature of interventions in many studies (Durand et al., 1996; Malow et al., 2014; Moss et al., 2014). Further research is needed to establish the efficacy of modified extinction procedures in children on the autism spectrum (K. Turner & Johnson, 2013; Singh & Zimmerman, 2015; Vriend et al., 2011).

Positive Reinforcement. As discussed in Chapter 1, when a consequence immediately follows behaviour and increases the probability of that behaviour re-occurring, this is termed reinforcement (i.e., operant behaviour is strengthened; Hanley et al., 2003; Skinner, 1969). Positive reinforcement is a critical component of any behavioural intervention aimed at increasing desirable behaviour in children, including those with DD (Lang et al., 2013; Weston et al., 2018). Moreover, the incorporation of strategies that strengthen desirable behaviour within behavioural interventions can enhance social validity, compared to treatments aimed at reducing problem behaviour in isolation (e.g., extinction; Lanovaz & Sladeczek, 2012). Sleep itself is a primary reinforcer for the behaviour of falling

asleep (Blampied & France, 1993; Bootzin, 1977), but this source of reinforcement cannot be directly externally manipulated in order to strengthen the behaviour of falling asleep. External forms of positive reinforcement (e.g., praise and rewards) can instead be used to strengthen a child's sleep-conducive behaviour during the sleep onset chain (e.g., by rewarding compliance with various components of the bedtime routine), and during the night (e.g., for falling asleep quickly, for sleeping independently, for remaining in bed all night until a set wake time; Mindell & Owens, 2015; Pattison et al., 2020).

Typically, reinforcement should be delivered immediately following the desirable behaviour if it is to have an effect on that behaviour (Mindell & Owens, 2015). In the sleep context however, it is not always feasible to reward behaviour immediately; for example, it is not possible to reinforce a child for falling asleep once asleep (France & Blampied, 2005). Further, delivery of rewards can paradoxically result in arousal. Instead, rewards may be given to a child in the morning contingent on the previous night's sleep (e.g., sleeping all night in one's own bed). This may require the response-reward contingency to be modelled visually such as through a social story (e.g., McLay, France, Blampied, van Deurs, et al., 2021; P. Moore, 2004), and may entail the use of token reinforcement (e.g., receiving stickers on a chart to earn a larger reward). Rewards may also be used to reinforce adherence to other BSI strategies, such as correct use of the bedtime pass (Freeman, 2006; Friman et al., 1999; Malow et al., 2014). Rewards (which are relatively immediate and tangible) may be particularly beneficial for younger children, for whom intrinsic rewards (e.g., improved sleep) may be less salient and motivating than an external reward system (Pattison et al., 2020).

Alternative Therapies

Various types of non-traditional methods are commonly trialled by parents to treat sleep problems in children on the autism spectrum, including weighted blankets, massage

therapy, white noise, exercise, aromatherapy, and homeopathy (Buckley et al., 2020; Cortesi et al., 2010; Cuomo et al., 2017; McLay & France, 2016; McLay et al., 2020; P. Williams et al., 2006). Anecdotal claims that non-traditional treatment approaches are effective are widespread; for example, Schreck et al. (2013) found that in print media 75% of mentioned treatments (non-specific to sleep) for individuals on the autism spectrum were for non-empirically supported treatments. A parent-survey by P. Williams et al. (2006) indicated that many parents hold beliefs that non-traditional approaches have positive benefits for children on the autism spectrum.

Further, in a recent survey of 244 parents of children on the autism spectrum, McLay et al. (2020) found that non-empirically supported methods such as homeopathy, co-sleeping and exercise, were reported to be frequently used by parents to treat sleep problems, while empirically supported behaviour interventions (e.g., planned ignoring) were used relatively less frequently. Interestingly, McLay et al. (2020) found that frequency of use was strongly related to parent perception of treatment effectiveness, with higher frequency use associated with higher ratings of treatment efficacy. For example, parents who used exercise, weighted blankets, or white noise to treat sleep problems rated these treatments as effective in 74%, 67% and 54% of cases respectively, even though these strategies are not considered evidence-based practice. In some cases, white noise and weighted blankets were perceived to be effective despite parents having no direct experience using these strategies (McLay et al., 2020). These findings are in line with research non-specific to sleep (e.g., Miller et al., 2012) to suggest that parents frequently access and employ treatments for children on the autism spectrum for which there is no evidence-base, or for which the evidence-base has not yet been established.

Very few studies have investigated the efficacy of non-traditional approaches to treat sleep problems in children on the autism spectrum (Cortesi et al., 2010; McLay & France,

2016; Singh & Zimmerman, 2015). Extant research shows there is no evidence that weighted blankets (or specialised mattress technology) can effectively reduce sleep problems in children on the autism spectrum (Buckley et al., 2020; Gringas et al., 2014; McLay & France, 2016). A review by McLay and France (2016) examined the evidence for massage therapy, aromatherapy, and vitamin supplements on sleep outcomes in children on the autism spectrum; results were inconclusive, with a lack of methodological rigor limiting certainty regarding study outcomes. A later review by France, McLay, Hunter and France (2018) examining non-traditional approaches to improve sleep in clinical and non-clinical populations of children and young people, found that some approaches demonstrated promise in certain populations. This included white noise with TD infants and children (including those with sleep disturbance), bright light therapy with young people with delayed sleep phase, and massage therapy in TD infants and children, and children with DD (France et al., 2018).

There is a strong need for further research using rigorous methodology to examine the efficacy of non-traditional approaches to improving sleep and sleep problems, as well as the mechanisms for the effects (Cuomo et al., 2017; France et al., 2018; McLay & France, 2016). Of these approaches, white noise is considered further below, as it is relevant to the treatment of sleep-related vocal stereotypy in children on the autism spectrum, including participant children in this thesis.

White Noise

White noise is continuous sound that masks other sounds within the range of human hearing (20-20,000 Hz; France et al., 2018). It is typically implemented through an electronic device or machine that plays white noise through speakers (Borkowski et al., 2001; Forquer & Johnson, 2005). The use of white noise in relation to sleep is widespread (Reidy et al., 2021). It is thought to improve sleep by continually masking environmental noise that may

distract a child from sleeping, thereby helping to decrease arousal levels and allowing a child to focus on a single set of sounds (Brackbill, 1973; Forquer & Johnson, 2005; France et al., 2018; Singh & Zimmerman, 2015). It is also possible that white noise improves sleep in part by acting as an S^D for sleep (i.e., through a learned association between listening to white noise and falling asleep; Forquer & Johnson, 2005; France et al., 2018; Stores, 1996). If a child wakes during the night, then the stimulus remains present to set the occasion for returning to sleep (Jin et al., 2013). White noise is readily available, cost-effective, is easily implemented by parents in the home setting and is well-tolerated by parents and children (Forquer & Johnson, 2005; K. Turner & Johnson, 2013; Knight & Johnson, 2014; Rosalez et al., 2020).

There is some evidence that white noise may reduce SOD and the frequency of NWs in TD infants (Borkowski et al., 2001; Forquer & Johnson, 2005; France et al., 2018; Spencer et al., 1990) and children with ADHD (Rosalez et al., 2020). Only one study[2] (Knight & Johnson, 2014) has examined the effects of white noise on sleep in children on the autism spectrum. Knight and Johnson (2014) used white noise in conjunction with sleep hygiene (positive bedtime routines) and graduated extinction, to treat sleep problems including SOD and NWs in three children on the autism spectrum aged 4-5 years. Knight and Johnson (2014) reported the multimodal interventions effectively reduced sleep problems across all children, and treatment effects remained relatively stable at follow-up. It is noteworthy that parent adherence to treatment procedures was the highest (100%) for white noise, in comparison to the two other treatment components (Knight & Johnson, 2014). It is not possible to interpret the effect of white noise on sleep outcomes because white noise was not used in isolation (i.e., other behavioural treatment components were involved).

[2] At the time of writing this thesis, another relevant paper (McLay, France, Blampied, et al., 2019; for which I was a co-author) was available, but was here excluded because it includes data presented in Study 1 (Chapter 4) of this thesis.

Overall, research into the use of white noise to improve sleep problems in children, including those on the autism spectrum, is scarce, and consequently, further research is needed to establish its effect (France et al., 2018; McLay & France, 2016). The use of white noise in the treatment of sleep-related stereotypy is discussed in further detail in subsequent chapters in this thesis.

Functional Behaviour Assessment: The Missing Link

No single treatment approach is effective for all sleep problems across all children (Cuomo et al., 2017) and there are currently no published guidelines to direct clinical decision-making regarding the most appropriate intervention (or combination of interventions) for sleep problems in children with neurodevelopmental disabilities (Rigney et al., 2018; Vivanti et al., 2014). Further, research shows children on the autism spectrum can vary widely in terms of their response to treatment (Lombardo et al., 2019; Singh & Zimmerman, 2015; Vivanti et al., 2014). One reason why behavioural treatments may fail to produce change is that treatments often neglect to target the specific factors maintaining sleep problems for an individual (Ballester et al., 2020; Spruyt & Curfs, 2015; Stores & Wiggs, 1998). Causes of sleep problems in children on the autism spectrum are often multifactorial (Goldman et al., 2012; Krakowiak et al., 2008; Liu et al., 2006); it is essential therefore, that treatment is informed by assessment that comprehensively identifies modifiable factors that influence child and parent behaviour (Brown & Piazza, 1999; Hanley et al., 2014; Iwata & Dozier, 2008; Spruyt & Curfs, 2015).

One clear solution to the problem of which intervention(s) should be chosen and designed for whom is to align treatment to individual needs (Masi et al., 2017; Roussis et al., 2021; Singh & Zimmerman, 2015). An evidence-based method of achieving this is to select interventions based on the outcomes of FBA (Didden & Sigafoos, 2001; Hanley et al., 2014; Jin et al., 2013; Kodak & Piazza, 2008). FBA is a comprehensive assessment process that

identifies the role of the environment and learning in sleep problems; in particular, the purposes (i.e., functions) that a behaviour serves a person (Cooper et al., 2020). It provides a researcher or clinician with a clear hypothesis of *why* problem behaviour is occurring for an individual in a specific environment (Beavers et al., 2013; Blampied, 2013a; Brown & Piazza, 1999; McLay, France, Blampied, van Deurs, et al., 2021).

Specifically, FBA involves the systematic identification of the controlling factors for sleep problems, including: (a) antecedents (e.g., sleep environment and S^D); (b) motivational operations (i.e., states of satiation or deprivation affecting behaviour); and (c) the reinforcing consequences maintaining (sleep-conducive and sleep-interfering) behaviour. Knowledge of the factors controlling a child's sleep problems enables a clinician to develop a case conceptualisation regarding why sleep problems are occurring for an individual (i.e., to generate hypotheses regarding the function or purpose of a child's behaviour). The case conceptualisation is then used to directly select treatment strategies and inform the design of the treatment(s), so that evidence-based interventions are synthesised with the assessment process (Blampied, 2013a; Hanley et al., 2014). This process provides a clear rationale for the selection of any treatment for any individual (Weiskop et al., 2001; 2005).

Function-based interventions target a reduction in sleep problems by: (a) modifying the sleep environment and S^D to improve stimulus control for sleep; (b) manipulating the MOs for sleep (i.e., ensuring that motivation for sleep [sleep pressure] peaks at the scheduled bedtime, and that competition from other motivational states is low); (c) disrupting or weakening reinforcement contingencies maintaining sleep problems, and establishing new contingencies through which sleep conducive behaviour is strengthened; and (d) identifying appropriate, alternative behaviours to be taught to replace problem behaviour while continuing to serve the same function (Beavers et al., 2013; Blampied, 2013a; Hanley et al., 2014; Kodak & Piazza, 2008).

FBA was developed from experimental functional analysis, referring to pre-treatment assessment involving the direct, experimental manipulation of variables to isolate potential factors maintaining problem behaviour (Beavers et al., 2013; Iwata et al., 1982; 1994; Iwata & Worsdell, 2005; Schlinger & Normand, 2013). A robust body of evidence demonstrates that treatment for problem behaviour (e.g., self-injury, aggression) informed by functional analysis is more effective than a standardised approach that is not informed by such assessment (Heyvaert et al., 2014; McLay, France, Blampied, van Deurs, et al., 2021; S. Harvey et al., 2009; Scotti et al., 1991). Functional analysis (of this direct experimental sort) is not commonly used to test variables maintaining sleep problems however, because highly structured and controlled analysis is not necessarily feasible (e.g., would require implementation overnight) nor ethical (e.g., intentionally reinforcing problem behaviour) to implement in the sleep context (Blampied, 2013a; Blampied & Bootzin, 2013). Instead, FBA involves a range of comprehensive assessment processes that enable the identification of specific antecedents and consequences underpinning sleep problems in natural settings (Blampied, 2013a; Hanley et al., 2014; Schlinger & Normand, 2013).

Measures Included in Functional Behaviour Assessment

A comprehensive FBA includes several indirect and descriptive measures, used to inform both the identification and the description of the topography of sleep difficulties and the contingencies operating to maintain problem behaviour (Blampied, 2013a; Hanley et al., 2014; Iwata & Dozier, 2008; M. Moore et al., 2017). Indirect measures include interviews and parent-report questionnaires (e.g., the CSHQ, the Sleep Assessment and Treatment Tool [SATT, described below; Hanley, 2005]) with parents or caregivers who are highly involved with the child's sleep regime and life (Abel et al., 2018; Jan et al., 2008; Jin et al., 2013). Parent-report questionnaires are a commonly used method to help to categorise types of sleep difficulty and its typical frequency, duration, and intensity, and to identify sleep habits and

patterns (Blampied, 2013a; Jan et al., 2008; Knight & Johnson, 2014; M. Moore et al., 2017). Descriptive assessment involves direct observation of problem behaviour in the environment it occurs in and is usually achieved through parent-reported sleep diaries and night-time video recordings (i.e., videosomnography [VSG]; Astill et al., 2012; Hanley et al., 2014). The use of indirect (e.g., parent-report questionnaires) and direct (e.g., parent-reported sleep diaries) measures to assess sleep outcomes are described in further detail in the next chapter.

Functional Behaviour Assessment in the Autism and Sleep Literature

Function-based interventions are classified as evidence-based practice, meaning there is sufficient quality and replication of research demonstrating the efficacy and suitability of interventions informed by FBA to treat challenging behaviours in children on the autism spectrum (Beavers et al., 2013; Campbell, 2003; Hanley et al., 2014; Wadsworth et al., 2015). There is strong evidence that interventions that are informed by FBA are more effective at treating challenging behaviour than treatments that are arbitrarily selected based on topography (i.e., without consideration of the behavioural function; Cooper et al., 2020; Spruyt & Curfs, 2015). There are several reasons why FBA is particularly suitable for informing interventions for sleep problems in children on the autism spectrum. Sleep problems in children on the autism spectrum are heterogeneous, and vary in type, topography, and function across and within children. To treat sleep problems effectively, intervention must target the variables underpinning behaviour for an individual.

First, multiple children can experience the same type of sleep problem, but with different underlying functions for each child. For example, two children may experience SOD, but for one child SOD may be underpinned by engagement in automatically reinforced stereotypy, and for another child SOD may be perpetuated by socially maintained CC behaviour. The topography of the problem may therefore also differ across children (e.g., how SOD manifests). Conversely, multiple children may experience differing types of sleep

problems maintained by similar functions. For example, SOD in one child, and co-sleeping in another, may both be maintained by access to reinforcing parent attention.

Second, a single child can experience multiple sleep problems, for which the underlying functions may differ. For example, SOD may be maintained by access to tangible items (e.g., toys), but NWs may be maintained by access to parent attention. Further, each type of sleep problem may serve multiple functions for an individual. For example, SOD may be perpetuated by access to tangible items as well as parent attention and may be negatively reinforced via escape of the demand to go to sleep. Conversely, differing sleep problems may serve the same underlying function for an individual; for example, SOD and NWs may both be maintained via access to parent attention. For any of these cases, FBA is needed to assess the underlying functions for behaviour carefully and comprehensively to underpin the design of differential and multimodal treatment (Brown & Piazza, 1999; Campbell, 2003; Didden et al., 2002; Spruyt & Curfs, 2015).

Further, sleep problems do not occur in isolation and careful consideration must be given to the family context (Meltzer & Montgomery-Downs, 2011), so that interventions are responsive to the individual circumstances and needs of families. FBA enables the identification of parent goals and preferences for treatment, so that BSI are developed in conjunction with parents, modified to address individual family needs (Jin et al., 2013; Pattison et al., 2020). Given that parents must implement BSI within their own home, the inclusion of parents within the assessment and treatment process is critical to ensure treatments are acceptable to families, are implemented correctly (fidelity) and for sustained periods (maintenance; Hanley et al., 2014; K. Turner & Johnson, 2013; Pattison et al., 2020).

Although there is a robust body of literature evaluating the efficacy of function-based interventions to treat challenging behaviours (e.g., aggression, self-injury) in children on the autism spectrum, relatively fewer studies have examined the efficacy of function-based

behavioural interventions for the treatment of sleep problems (Jin et al., 2013; McLay, France, Blampied, van Deurs, et al., 2021). However, over the past 20 years, a number of studies have emerged. For example, Didden and colleagues (2002) used FBA to inform treatment for sleep problems in four children on the autism spectrum, including difficulty with settling, frequent NWs and EMW. FBA indicated that parental attention positively reinforced sleep problems in all four cases (Didden et al., 2002). An extinction procedure was implemented for each child to remove the reinforcement of parental attention maintaining behaviour. Following intervention sleep problems reduced across all children and results were maintained at a 6-month follow-up (Didden et al., 2002). Didden et al. (2002) concluded that FBA is a valuable means of informing intervention; however, there is a lack of studies utilising pre-treatment FBA in the treatment of sleep problems in children with DD.

P. Moore (2004; described above) used FBA to assess sleep problems in a 4-year-old boy on the autism spectrum with learning disabilities, across the home and school setting. FBA included interviews, direct observations, video recording, and parent-reported sleep diaries and questionnaires, to identify factors that precipitated and were maintaining sleep problems (P. Moore, 2004). A function-based behavioural intervention, including a social story reflecting a set bedtime routine, graduated extinction, visual aids, and reinforcement, was found to improve sleep quantity and quality for the boy and his mother (P. Moore, 2004).

In extension of research examining sleep problems in the home-setting, Friedman and Luiselli (2008) used FBA to inform an intervention for a 13-year-old boy on the autism spectrum who suffered from excessive daytime sleep in the school setting. FBA included questionnaires, independent behaviour observations and A-B-C checklists for the occurrence and duration of problem behaviour in a school setting (Friedman & Luiselli, 2008). The results of FBA indicated that excessive sleep was negatively reinforced through avoidance of non-preferred activities (Friedman & Luiselli, 2008). Function-based intervention involved

the removal of sleep-associated stimuli (a soft mat and chair), redirection by staff to highly engaging, preferred activities when daytime sleep was about to occur, and differential reinforcement (i.e., reinforcing a behaviour incompatible with sleep) in the form of praise for remaining awake (Friedman & Luiselli, 2008). Treatment effectively eliminated daytime sleep, with results maintained at a 6-month follow-up session (Friedman & Luiselli, 2008).

Jin et al. (2013) extended extant research regarding the utility of FBA to inform BSI for children on the autism spectrum by evaluating the use of the SATT (Hanley, 2005) to inform individualised, function-based treatments for sleep problems in three children (aged 7-9 years) on the autism spectrum. Sleep problems included SOD, NWs and EMW, and parents of the children reported high levels of stress owing to chronic disruption from the sleep problems (Jin et al., 2013). The SATT is an FBA measure specifically designed to identify the topography and function of a child's sleep problems with information obtained through parent interviews, including the antecedents and consequences maintaining sleep problems, history of sleep disturbance, and parent's goals for treatment. The information is then used to develop hypotheses regarding the factors contributing to sleep problems, to directly inform an individualised, comprehensive BSI for each child (Jin et al., 2013).

The individualised treatments included extinction to disrupt the reinforcement contingencies maintaining problem behaviour, and strategies to promote behavioural quietude and the onset of sleep (e.g., the faded bedtime procedure; Jin et al., 2013). Adaptations to each child's sleep environment, including the removal of reinforcers for problem behaviour, were made according to the outcomes of the SATT for each child (Jin et al., 2013). Treatment effectively reduced sleep problems and increased TST for all children (Jin et al., 2013). Jin et al. (2013) concluded that the study demonstrates the value of comprehensive, individualised assessment to inform intervention, so that treatment directly targets factors that serve to influence problematic sleep-related behaviour for each child.

More recently, a series of additional studies conducted by McLay and colleagues have evaluated the effectiveness of function-based, individualised multimodal interventions to treat sleep problems in children on the autism spectrum (McLay, France, Blampied, van Deurs, et al., 2021; McLay, France, Knight et al. 2019; van Deurs et al., 2019). These studies used a combination of indirect and descriptive measures to inform behavioural treatment, including parent-report questionnaires (e.g., CSHQ), sleep diaries and VSG. In a case analysis, McLay, France, Blampied, van Deurs, et al. (2021) investigated the efficacy of function-based behavioural interventions to treat sleep problems in 41 children on the autism spectrum. FBA was conducted using parent-reported questionnaires, sleep diaries and VSG, and was used to identify the environmental antecedents and the reinforcing consequences contributing to and maintaining sleep problems for each child. A behavioural case conceptualisation and individualised treatment plan (Blampied, 2013a) was developed for each child based on the outcomes of the FBA. Sleep interventions across children included a combination of antecedent- and consequence-based modifications, and the teaching of adaptive (sleep-conducive) replacement behaviours. Antecedent-based procedures included modifications to S^D (e.g., S^D for sleep/wake times, social stories, the location of sleep onset), sleep hygiene practices (e.g., consistent bedtime routine), and the faded bedtime procedure. Consequence-based procedures included disrupting the reinforcement contingencies for sleep-competing behaviour, including extinction and modified extinction, systematic fading of parental presence, reinforcement for sleep-conducive behaviours and the bedtime pass.

Results of the case analysis showed that the parent-delivered, function-based, multimodal interventions effectively reduced sleep problems across children. McLay, France, Blampied, van Deurs, et al. (2021) concluded there is support for the effectiveness of function-based BSI, and a strong need for future research involving rigorous methodology to

extend what is known regarding the effectiveness of function-based interventions to treat sleep problems in children on the autism spectrum.

Summary of the Evidence for Behavioural Sleep Interventions and Future Directions

The significant impact of sleep disturbance on quality of life for children and families creates an immediate need for effective interventions to treat sleep problems, and behavioural interventions are recommended as first line treatment (Keogh et al., 2019; Pattison et al., 2020; Phillips et al., 2020; Rigney et al., 2018; Roussis et al., 2021; Scantlebury et al., 2018). Research into the effectiveness of behavioural interventions to treat sleep problems in children on the autism spectrum is expanding (Keogh et al., 2019; Pattison et al., 2020). There is increasing evidence for overall effectiveness, irrespective of age, type of sleep difficulty, or severity of autism (Carnett et al., 2020; Cuomo et al., 2017; K. Turner & Johnson, 2013; Keogh et al., 2019; Pattison et al., 2020; Phillips et al., 2020; Richdale & Wiggs, 2005; Rigney et al., 2018; Schreck, 2001; Vriend et al., 2011; Wiggs & France, 2000).

A systematic review conducted by Rigney and colleagues (2018) assessed the impact of behavioural interventions on sleep outcomes in children with and without neurodevelopmental disorders (including autism) in 40 studies. Results showed that all 40 studies reported some level of improvement in sleep outcomes following at least one behavioural treatment component (Rigney et al., 2018). The most used strategies were sleep hygiene practices, particularly positive bedtime routines and parent education, extinction, modified extinction and positive reinforcement (Carnett et al., 2020, Keogh et al., 2019; Rigney et al., 2018), with the most empirical support for extinction and modified extinction procedures (Richdale, 2013; Richdale & Wiggs, 2005; Schreck, 2001; Vriend et al., 2011).

These behavioural strategies have largely been applied to children on the autism spectrum without modification (i.e., entail the same procedures as used for TD children; Keogh et al., 2019; Rigney et al., 2018). Critically, BSI entail the provision of information and training for parents, which increase parents' knowledge and skills to manage children's sleep-related behaviour in the home-setting (Cuomo et al., 2017; Hastings, 2002; Pattison et al., 2020; Prata et al., 2018). BSI are shown to increase parents' sense of control, competency, and overall wellbeing, to be valued by parents, and to support children's long-term learning in the child's natural environment (Kirkpatrick, Louw, et al., 2019; Pattison et al., 2020; Prata et al., 2018; Rigney et al., 2018; Vriend et al., 2011).

While the body of research demonstrating the efficacy and suitability of behavioural interventions to treat sleep problems in children on the autism spectrum is promising, there are a number of limitations and unanswered questions arising from extant research. First, compared to the evidence-base in TD children, there is a lack of studies investigating the efficacy of behavioural interventions to treat sleep problems in children on the autism spectrum, highlighting the need for further research in this area (Meltzer & Mindell, 2014; Spruyt & Curfs, 2015). In the absence of established guidelines to guide clinical decision-making, it is important that research further evaluate the relative utility of FBA to inform treatments for sleep problems in children on the autism spectrum. Function-based interventions are an evidence-based means for reducing problem behaviours in children on the autism spectrum and are particularly well suited to treat sleep problems given the heterogeneity in presentation and function of sleep problems in children on the autism spectrum.

Essential to the process of FBA is the importance of collaborating with parents to design interventions that are a good contextual family fit, which can increase the likelihood that treatments are implemented with fidelity, and that progress is sustained in the long-term

(K. Turner & Johnson, 2013; Jin et al., 2013; Moes & Frea, 2002). However, studies do not always report treatment fidelity, which is important to understanding treatment effects (i.e., if improvements can be attributed to reliable implementation) and parents' willingness and capacity to execute treatment procedures reliably and accurately (i.e., fidelity is important to the translation of evidence-based interventions into practice; Kirkpatrick, Louw, et al., 2019; Spruyt & Curfs, 2015). In addition, assessing social validity is critical to ensuring treatments are acceptable to and socially meaningful for families (Callahan et al., 2017; Finn & Sladeczek, 2001; Kazdin, 2000; P. Moore, 2004). Furthermore, there are major challenges in ensuring that practitioners are well informed about FBA and technically proficient in its use and in the design and implementation of function-based treatments.

Further, variation in core intervention features and methodological limitations within extant research limits certainty in the available evidence (Pattison et al., 2020; Scantlebury et al., 2018). For example, intervention studies vary in the mode of treatment delivery (e.g., individual or group level), the duration of clinical support (e.g., no support throughout implementation, or on-going support), measurement (e.g., parent-reported information, or recordings of sleep using instrumentation [e.g., VSG]), and follow-up periods (e.g., no follow-up, 1-month, 6-months; Kirkpatrick, Louw, et al., 2019; Spruyt & Curfs, 2015; Pattison et al., 2020). It is not clear whether particular methodologies improve treatment effects (Pattison et al., 2020). There is scarcity of controlled, clinical trials that investigate and compare efficacy of treatment types to address sleep problems in children on the autism spectrum (France & Blampied, 2005; K. Turner & Johnson, 2013; Meltzer & Mindell, 2014; Wiggs & France, 2000).

Studies are needed involving rigorous methodology, including controlled experimental design, blinded assessment of outcomes, interobserver agreement (IOA), and evaluation of long-term treatment effects (Cuomo et al., 2017; McLay, France, Blampied, van

Deurs, et al., 2021; Meltzer & Mindell, 2014; Pattison et al., 2020; Phillips et al., 2020; Richdale & Wiggs, 2005; Rigney et al., 2018; Scantlebury et al., 2018; Vriend et al., 2011). Where blinded assessment is not possible, the inclusion of instrumentation-based measures (e.g., VSG or actigraphy) is necessary to gather IOA data to reduce the potential for bias within parent-report (Rigney et al., 2018; Scantlebury et al., 2018). The evaluation of long-term effects in applied research is particularly important to assess the durability of meaningful changes in natural settings over time (Cooper et al., 2020; Kirkpatrick, Louw, et al., 2019).

Moreover, behavioural interventions used to treat sleep problems in children on the autism spectrum are predominantly approaches with established efficacy in TD children (Pattison et al., 2020; Spruyt & Curfs, 2015). While results suggest such strategies (e.g., extinction, modified extinction) are effective, the adaptations required to ensure behavioural interventions are responsive to the needs of children on the autism spectrum are not well understood (Pattison et al., 2020; Spruyt & Curfs, 2015). For example, sleep problems in children on the autism spectrum are often more chronic and severe than in TD children and can include autism-associated behaviour such as stereotypy (Phillips et al., 2020; Richdale & Schreck, 2009). Despite these differences, few studies have examined the types of modifications established interventions might require to effectively address autism-associated behaviour (Pattison et al., 2020; Phillips et al., 2020).

Relatedly, a majority of studies have examined the effectiveness of BSI in school-aged children on the autism spectrum (i.e., 5-13 years; Phillips et al., 2020; Spruyt & Curfs, 2015), with fewer studies focussing on children younger than five years (K. Turner & Johnson, 2013). It is important that research further evaluate the effectiveness of BSI, including the modifications that may be required, for younger children on the autism

spectrum, since early intervention is critical to preventing long-lasting problems in children and families (K. Turner & Johnson, 2013; Scantlebury et al., 2018; Phillips et al., 2020).

Chapter 3

General Methodology: Studies 1, 2 and 3

This chapter describes the general methodology common to Studies 1, 2 and 3. This includes an overview of the sleep research team, ethics and consent procedures, participant information, the setting, general materials, treatment acceptability, reliability and fidelity assessment, common dependent variables, procedures, and approach to data analysis. Methodological details relating specifically to Studies 1, 2 and 3 are described in Chapters 4, 6 and 7, respectively.

The Sleep Research Team

The sleep research team is based at Te Whare Wānanga o Waitaha/University of Canterbury, Christchurch, Aoeteroa NZ. This thesis is part of a larger research programme evaluating interventions for sleep problems in children on the autism spectrum. Three senior academics lead the research team: two of whom are registered[3] psychologists and the other is an experienced practitioner of ABA. The wider research team consists of registered psychologists, intern psychologists, postgraduate students, and research assistants. At the time of undertaking this research, the author was an intern psychologist and postgraduate student, responsible for providing all clinical assessment and intervention procedures with families, under the supervision of the senior academics.

Ethics and Participant Consent

Ethics Application and Approval

This thesis received ethical approval from the University of Canterbury Human Ethics Committee (#HEC 2018/47; see Appendix A).

[3] Note *registered* in New Zealand is the equivalent of *licensed* in other countries.

Screening and Consent

Immediately following referral or self-referral to the study (see Recruitment below), an initial screening process was completed by phone to ensure the research project was appropriate for the participants and that participants met eligibility criteria (described below). The process also ensured that there were no medical or physical factors contributing to children's sleep difficulties, which may have contraindicated use of a behavioural intervention. Information relating to confidentiality was conveyed verbally to families prior to the screening questions.

All parents were required to give consent for their child to participate in this research. Parents were fully informed by phone of what the research entailed, and information and consent forms were mailed to eligible families. Parents and children were provided with written information sheets (see Parent and Child Information sheets in Appendix B and C, respectively) before parents' written consent was obtained (see Parent Consent Form in Appendix D).

Consent was not able to be obtained directly from the child participants in this thesis because all were of a young age ($\leq$ 10 years) and had a developmental disorder that limited their ability to provide fully informed consent. Instead, children were verbally informed of the research project and what it involved (e.g., "Jolene will be working with my parent/s to help me to learn to sleep better"), and informed assent was obtained. Parents also signed a Child Consent Form (see Appendix E) on their child's behalf. Consent for audiovisual recording of children's sleep and the recording of clinical interviews was obtained from all parent participants (see Audiovisual Consent Form in Appendix F).

Participants

Recruitment

A flyer (see Appendix G) was shared with NZ organisations and community service providers for children and young people on the autism spectrum and their families via email and social media. These organisations were asked to share the flyer with families, and to refer suitable families to the research project. Interested parents were also able to self-refer.

Eligibility Criteria

Children were eligible for inclusion if they met the following criteria: they had (a) a formal diagnosis of ASD, as verified by a paediatrician, psychiatrist or psychologist (APA, 2013); (b) were between 3-12 years of age; and (c) had parent-reported sleep problems, including SOD, frequent or prolonged NWs, EMW and/or unwanted co-sleeping, later verified by sleep assessment tools (e.g., the SATT, sleep diaries, and video analysis). For Studies 2 and 3, eligibility criteria were extended to include: (d) engagement in at least one form of stereotypy (e.g., repetitive motor movements) after being bid goodnight. Participants were excluded from the study if they had any medical conditions that interfered with their sleep (e.g., sleep apnoea); if there was contraindication for a behavioural intervention (e.g., sleep apnoea, some stimulant medications); or if there were factors that made implementation of a behavioural intervention possibly unsafe (e.g., presence of nocturnal seizures).

Due to the clinical heterogeneity present in autism, no restrictions were placed on level of verbal ability, the presence of co-occurring disorders, or whether children were taking medication at the time of the study. Many children on the autism spectrum are medicated and still require sleep interventions, and thus there is reason not to exclude children on this basis. The use of single-case research design (discussed below) permits inferences about treatment effects to be made on an individualised basis, including

consideration of any potential confounds such as medication. Medication dosage was kept stable throughout baseline and treatment; if dosage was changed (i.e., melatonin for one child) then a new baseline was taken. Each child's verbal ability was assessed using the *Communication* domain on the Vineland Adaptive Behavior Scales, Second Edition, Caregiver Rating Form (VABS-II; Sparrow et al., 2005), administered during the assessment phase. Children were deemed to have difficulty with communication if their scores on the receptive and expressive communication subdomains of the VABS-II were more than one year below their chronological age.

Setting

Families were recruited throughout NZ; 4/12 families resided in the Canterbury region and the remaining resided outside of Canterbury. The clinical intake interview, treatment planning discussion, and treatment progress discussions were held either in the family home, at the Pukemanu Clinic at the University of Canterbury (Canterbury residents only), or via Skype. For five families who resided outside of Canterbury, the author travelled to the family home to conduct the intake interview. All interventions were implemented by parents in the home, with support provided via Skype, phone, and email contact. Pre- and post-treatment questionnaires were posted to families or completed over the phone.

General Materials

Sleep Diaries

Paper sleep diaries were provided for parents who were instructed on how to complete them. Further information about these sleep diaries is provided below, and an example of a standard sleep diary is included in Appendix H.

Video Equipment

Swann Advanced-Series DVR4-1200, night-time, time-lapse, infrared video cameras were placed in an elevated position, facing the bed in the child's bedroom. The camera was linked to a monitor, which was placed outside of the bedroom so that parents could view their child's behaviour while the camera was operating and could also ensure correct placement of the camera. Video equipment was supplied and set up by the author or was posted to families along with written instructions. The author was in regular contact with families to help them to manage the equipment and to resolve any technical issues. Video data was recorded to an internal hard drive, and later downloaded onto an external hard drive for coding.

Sleep Measures

Sleep data were collected using indirect and direct methods. Indirect methods included the clinical interview (described under General Procedure below) and the CSHQ. Direct means of data collection included parent-reported sleep diaries and visual analysis of VSG. FBA was undertaken using clinical interviewing, the SATT and sleep diary data.

The Children's Sleep Habits Questionnaire

The CSHQ (J. Owens et al., 2000) was administered pre- and post-treatment to evaluate change in the types and severity of sleep problems that children experienced. The CSHQ is a 33-item parent-report questionnaire used to categorise sleep difficulties, including bedtime resistance, SOD, sleep duration, sleep anxiety, NWs, parasomnias, sleep disordered breathing and daytime sleepiness. The CSHQ gives an overall total score and eight subscale scores, which reflect the different sleep domains that encompass the major sleep problems of preschool and school-aged children (J. Owens et al., 2000). Parents rate the frequency of how often a specific behaviour occurs within a typical week on a 3-point Likert scale (usually = 5-7 times/week; sometimes = 2-4 times/week; rarely = 0-1 times/week). Higher scores are

indicative of poorer sleep, and a total score $\geq$ 41 is indicative of clinically significant sleep disturbance (J. Owens et al., 2000).

The CSHQ is a commonly used measure of sleep problems in TD children as well as children on the autism spectrum, in both clinical and research settings (Goodlin-Jones et al., 2008; Malow et al., 2012; 2014). It has good internal consistency (α = .68 to .78) and test/re-test reliability (r = .62 to .79) for community and clinical populations (J. Owens et al., 2000). An abbreviated version of the CSHQ, the short-form CSHQ (SF-CSHQ: Bonuck et al., 2017) contains 23-items focussed on behaviourally-based as opposed to medically-based sleep problems; a cut-off score of 30 has correlations of 0.90-0.94 with the original CSHQ cut-off (Bonuck et al., 2017). The SF-CSHQ can discriminate between children with and without sleep disturbance, as measured by parent-report and actigraphy (Bonuck et al., 2017). The SF-CSQH was completed by the parents of 3/12 children in order to reduce the burden of written assessment material for these families, with the parents of the remaining children completing the full version CSHQ.

The Sleep Assessment and Treatment Tool

The SATT (Hanley et al., 2005; described previously in Chapter 2 under 'Functional Behaviour Assessment in the Autism and Sleep Literature') is a semi-structured interview that was used to guide questions about children's sleep problems within the clinical interview. The SATT features questions that assess and/or identify the child's: (a) history of sleep disturbance (e.g., 'how long have these problems occurred?'); (b) types of sleep problems (e.g., SOD, NWs; e.g., 'once in bed, does your child have difficulty falling asleep [e.g., it typically takes more than 15 min for him/her to fall asleep]?'), including descriptions of antecedents and consequences for the behaviour; (c) current sleep schedule (e.g., 'what time does your child typically go to bed?'/'what time does your child typically fall asleep?/'what time does your child wake in the morning?'); (d) sleep environment; (e) any

sleep dependencies; (h) sleep-interfering behaviours (e.g., ‘does your child repeatedly call out or engage in other behaviour that requires you to return to his/her bedroom?’); and (i) parent’s sleep goals (e.g., ‘describe your goals regarding your child’s sleep’).

Parent-Reported Sleep Diaries

Parents completed daily sleep diaries across study phases. Parents recorded information about their child’s sleep patterns and sleep-related behaviour, which included: (a) the setting (i.e., where the child fell asleep) and duration of any daytime sleep; (b) the setting of night-time sleep; (c) time put to bed; (d) frequency of CCs; (e) child behaviour during CCs, and parent-responses to child behaviour; (f) estimated time of sleep onset; (g) the time and duration of NWs; (h) child behaviour during NWs, and parent-responses to child behaviour; and (i) time awake in the morning (see Appendix H).

The same sleep diary was used for all participants and across all stages of the study; however, individual codes were assigned to capture the child’s behaviour, and parent responses to specific behaviour. For example, a code ‘BF’ was used in the case where a mother breast-fed in response to a NW. Codes were individualised for each child according to the specific presenting problems, and parents’ responses. This allowed parents to note behaviour quickly and consistently, and for ease of IOA. The researcher kept in regular contact with parents by phone to ensure that they were consistently completing the sleep diaries and these diaries were collected from families at least once per week to monitor progress.

Videosomnography

VSG is a commonly used instrumental measure used to assess children’s sleep patterns in both research and clinical settings (Astill et al., 2012). It enables direct, permanently recorded observation of a child’s sleep/wake patterns and night-time behaviour

(excluding behaviours occurring outside the range of the camera), as well as parental responses to behaviour. It is also useful in detecting child behaviours parents may otherwise be unaware of (e.g., quiet awakenings; M. Moore et al., 2017; Richdale & Schreck, 2009; Sadeh, 1999). When coded, VSG data is a reliable method with which to corroborate sleep diary data because it permanently captures a direct veridical record of behaviour for comparison with other records (France & Blampied, 2005; Richdale & Schreck, 2009).

For the present research, VSG data served as a reliability measure, by comparing VSG data with parent-reported sleep diaries. Furthermore, when there was uncertainty regarding the child's behaviour or parents' responses, VSG recording was able to be reviewed. Parents were instructed to begin recording each night at the time they put their child to bed, and to stop recording when their child got up for the day. Information obtained from VSG included: (a) topographies of awake behaviour post-bedtime and during the night if the child woke (e.g., play, stereotypy); (b) topographies of sleep behaviour (e.g., sleep position, limb movement, eye movement); (c) frequency of CCs; (d) duration of SOL; (e) frequency and duration of NWs; (f) co-sleeping (in child bed); and (g) time awake in the morning. The video recordings were only able to capture behaviour that occurred within the vicinity of the child's bed, so events occurring elsewhere (e.g., co-sleeping in the parents' bed) were not recorded. In addition, the video equipment that was used did not record audio. Video recordings were made during at least 30% of baseline and intervention phases.

Secondary Outcome Measures

Additional psychometric measures including the CBCL (Achenbach & Rescorla, 2000; 2001), the Gilliam Autism Rating Scale – Third Edition (GARS-3; Gilliam, 2013), the Pittsburgh Sleep Quality Index (PSQI; Buysse et al., 1989), the Depression Anxiety Stress Scales short-form version (DASS-21; Lovibond & Lovibond, 1995) and the Relationship Quality Index (RQI; Norton, 1983), were administered pre- and post-treatment to assess the

collateral (secondary) impact of a BSI on child (e.g., autism symptom severity) and parent (e.g., mental health) outcomes. These measures are described in Chapter 10.

Treatment Acceptability

Treatment acceptability was assessed using the Treatment Acceptability Rating Form Revised (TARF-R; Reimers et al., 1992) administered at the completion of treatment, and a post-treatment interview with parents. The TARF-R is a 20-item parent-report questionnaire; 17-items measure ratings of treatment acceptability, and 3-items measure respondents' understanding of the treatment approach and severity of the problem behaviour. Six subscales including Effectiveness, Reasonableness, Willingness, Cost, Negative Side-Effects, and Disruption/Time are summed to yield an overall treatment acceptability score. The TARF-R has good reliability ($\alpha = .92$) and clinical utility (Finn & Sladeczek, 2001; Reimers et al., 1992).

In addition to the TARF-R, a semi-structured, post-treatment interview (see Appendix I) was held with parents to obtain information about their perspective of and satisfaction with the assessment and treatment process. A member of the sleep research team other than the author (i.e., a person who did not work directly with the participant families) conducted the interview by phone. Parents were asked how they felt about the intervention and progress their child made, their attributions and understanding of treatment effects, and their level of satisfaction with treatment. Parents were also asked about any personal impact or additional changes for their child following the BSI. Parents were given an opportunity to provide any feedback about how the assessment and treatment process could have been improved.

Reliability and Fidelity

Interobserver Agreement

IOA was calculated by a member of the research team who viewed at least 30% of the VSG recordings taken of each child across each study phase. The observer was blinded to the diaries that were recorded by each family on the corresponding night. Sleep behaviours that were coded by the researcher included all items reported in parent-report sleep diaries, except for vocalisations (including vocal stereotypy) because the VSG technology did not support audio. The coded data was then compared to sleep diary data for the corresponding nights, to gain a measure of IOA. The frequency of behaviour (e.g., CCs, NWs detectable by parents) was recorded as an agreement if this were documented by both the parent and the researcher. Measures of duration (e.g., of SOL and NWs) were recorded as agreement if parent and researcher report were ± 15 min. Sleep phenomena that were not detectable by parents (e.g., quiet awakenings during which the child remained in their bed) was excluded from IOA calculations. The percentage of agreement for each of the coded behaviours was calculated by the equation [Agreement/ (Agreement + Disagreement)] x100%. Individual measures of IOA are reported in the results of each study.

Treatment Fidelity

A protocol checklist derived from a task analysis of each measurable parent-implemented treatment component (e.g., returning the child to bed) was made for each family. The prescribed treatment protocol was compared to treatment data, including sleep diary and VSG data, and the author's treatment progress contact notes, to ascertain parent adherence to each treatment component in order to calculate treatment fidelity. Treatment fidelity was calculated for 30-40% of nights across all participants and across all study

phases, using the formula (Completed tasks/Total tasks) ×100%. An aggregate score was then calculated by computing an average percentage for each participant.

Common Dependent Variables

Key dependent variables included awake, asleep, SOL, sleep-interfering behaviour, stereotypy, and NWs, and are defined as follows. The dependent variables were measured based on parent-report and/or VSG observation.

Awake

Awake was defined as an arousal > 2 min that involved purposeful gross motor movement (e.g., sitting up, looking around) for at least 15 s or vocalisation (e.g., calling out, crying) with eyes wide open (Camerota et al., 2018; Jin et al., 2013; Schwichtenberg et al., 2018). Two types of wakeful state were possible: signalled wakefulness and behavioural quietude (quiet awake). Behavioural quietude involved the child being in a wakeful but quiet state (i.e., parents were likely unaware the child was awake), whereas signalled wakefulness involved parents being alerted to the child's wakeful state (e.g., through noisy, disruptive behaviour; Blampied & France, 1993).

Asleep

Asleep was defined as the child lying down with an absence of purposeful gross motor movement or vocalisation, closed eyes, and chest moving up and down in steady rhythm, for a continuous period of > 5 min (Camerota et al., 2018; Jin et al., 2013).

Sleep Onset Latency

SOL referred to the duration of time (min) between when the child was bid goodnight, until the child fell asleep (Jin et al., 2013). SOL was considered problematic (i.e., SOD) if it exceeded the recommended/age-appropriate sleep onset duration (i.e., 15-30 min;

Hirshkowitz et al., 2015; Ohayon et al., 2017). SOL was calculated from the time the child was bid goodnight until the time of sleep onset.

Sleep-Interfering Behaviour

Sleep-interfering behaviour was defined as any behaviour after the child was bid goodnight that impeded a child's ability to lie quietly and initiate and/or re-initiate sleep (i.e., during SOL and/or NWs; Jin et al., 2013). Sleep-interfering behaviour included engagement in play activity (e.g., with toys), CCs, stereotypy (see below) and disruptive night-time behaviour such as roaming the house (Jin et al., 2013).

Stereotypy

Stereotypy was defined as any behaviour that appeared repetitive and fixed (Rapp & Vollmer, 2005a), and included verbal and motor behaviour as well as restricted and repetitive activities and manipulation of objects (i.e., RMO; Melo et al., 2020; Rapp & Lanovaz, 2016).

Night Wakings

NWs were defined as an arousal > 5 min (Ohayon et al., 2017) that occurred prior to the determined wake time. The duration of NWs was recorded from the time the child was considered to be awake until the time sleep recommenced. A minimum of five min of continuous sleep was necessary to code a new NW (Camerota et al., 2018).

Procedures

Research Design

Studies 1-3 employed a non-concurrent single-case research design, explained in further detail within each study. Single-case design uses repeated measurement of designated dependent variables across an extended period within study phases to draw inferences about the causal relation between variables (Barlow & Nock, 2009). First, a continuous baseline

phase is used to determine the trend, level, and variability of dependent variable data, and to establish its predicted future trajectory without intervention (Kazdin, 1982). The baseline phase provides a within-case control comparison for the subsequent treatment phase (Kazdin, 1982; L. Cohen, Feinstein, et al., 2014; Lane & Gast, 2014).

Changes to the trend, level and variability of data that correspond with the implementation of intervention allow inferences to be drawn regarding the effects of treatment (Kazdin, 1982; L. Cohen, Feinstein, et al., 2014). For example, a treatment effect may be obtained when data trends in the opposite direction in an intervention phase from the baseline phase, whereas a continuation in trend between baseline and treatment may be better attributed to naturally occurring patterns of behaviour change rather than treatment itself (Kazdin, 1982; L. Cohen, Feinstein, et al., 2014). Level refers to the position of data points relative to their possible minimum and maximum, which indicates whether behaviour is improving, deteriorating, or is unchanged in response to intervention (Lane & Gast, 2014). Further, little variability within a phase indicates stability of a behavioural response (L. Cohen, Feinstein, et al., 2014) and makes the detection of change in other phases easier and more reliable. Conversely, high levels of variability within and between phases make detection of and inferences about change difficult or impossible.

Inferences are strengthened when there is replication of treatment effects across participants, behaviours, and settings (Kazdin, 1982; L. Cohen, Feinstein, et al., 2014). For example, in a multiple baseline across participants design, the introduction of intervention is staggered across multiple participants, making it possible to observe a replicated ‘when and only when treatment is introduced’ pattern to changes occurring between baseline and treatment phases, thereby increasing confidence that changes in behaviour are a direct result of intervention, rather than owing to extraneous variables (Kazdin, 2000). Any inference that a treatment has caused a change in behaviour is strengthened by replication. This may be

direct replication, where the same (or highly similar) treatment is replicated within the initial case (as in reversal design) or across cases (or behaviours or settings) as in the multiple baseline design (Kazdin, 1982). Systematic replication may then be used to explore the reliability and generality of any treatment effect detected (Barlow, et al., 2009; Walker & Carr, 2021). In addition, to reduce threats to internal and external validity within single-case research, multiple sources and methods of data collection are used to triangulate findings, and consideration is given to alternative explanations that could otherwise account for data, so that attributions to treatment effects are rational (Kazdin, 1982; Vertue, 2011). For Studies 1-3 in this thesis, repeated measurement (i.e., taking multiple data points across time) helped to strengthen inferences regarding behavioural response to intervention (Kazdin, 1982).

Single-case design is amongst the most common research design in behavioural intervention literature for evaluating the effects of intervention on reducing problem behaviour in individuals on the autism spectrum (Campbell, 2003; Heyvaert et al., 2014), including in the sleep literature (Spruyt & Curfs, 2015). A single-case design was selected for Studies 1-3 in this thesis because of its idiographic approach to research; single-case research retains the richness of person-specific data, which enables meaningful interpretation of treatment effects for an individual (Blampied, 2013b; 2017; Kazdin, 1982). Single-case design is particularly suitable for examining the effectiveness of individualised BSI for children on the autism spectrum, as it allows in-depth analysis of the complexities associated with unique cases and is important to the purpose of using FBA given the heterogeneity in autism and in sleep problems across and within individuals (Blampied, 2013b).

Furthermore, a single-case research design enabled the author to monitor an individual's treatment progress and make adaptations to treatment plans as needed, in accordance with the child's individualised response to treatment (Lane & Gast, 2014). A between-groups design would not have been suitable for the purposes of this thesis because

the degree of experimental control (e.g., strict inclusion criteria, random assignment, manualised treatment protocols) and aggregated data analysis (e.g., null hypothesis testing) would have reduced generalisability to individuals (Barlow & Nock, 2009; Blampied, 2013b; 2017; Carter, 2013; Kazdin, 1982). Instead, rigorous guidelines were followed to measure the effects of intervention across participant children, which included graphing and visual analysis of the dependent variables, as well as the use of descriptive statistics and contemporary methods of analysis including modified Brinley plots (Blampied, 2017; see Data Analysis below).

General Procedure

For all participants, the general phases of each study were *assessment* (including FBA), *case conceptualisation, baseline, intervention, maintenance,* and *follow-up.*

Assessment

Upon obtaining informed consent, assessment commenced. The clinical interview was a semi-structured interview that followed the format of a standard intake interview used by Child and Family Psychologists at the Pukemanu Centre and was conducted by the author. In two-parent households, either one or both parents participated in the interview. The purpose of the intake interview was to comprehensively assess each child's sleep problems in the context of broader child and family factors. The interview took approximately 1-1.5 hours and began with a discussion of child and family-specific information (e.g., child's diagnosis and any co-occurring conditions, family members living in the household), and the child's presenting sleep problems. Discussion about the child's sleep problems included the type (e.g., SOD, NWs) and topography, frequency, intensity and duration of the problems, typical bedtime routine (e.g., time put to bed, steps in the bedtime sequence), and environmental variables affecting the child's sleep (e.g., changes in temperature or noise during the night).

Information was also obtained regarding the history of sleep disturbance (e.g., onset of the sleep problem, chronicity) and its impact (i.e., on child, parents, wider family).

A full developmental history and relevant child (e.g., interests, schooling) and family information (e.g., living situation including sleeping arrangements, employment, parental mental health and wellbeing, sources of support, wider culture) was also obtained; this ensured families met inclusion criteria and allowed for a holistic look at which family and child factors were potentially functionally significant as part of the setting within which the sleep problems existed. Parents were also asked about their goals for their child's sleep. Treatment goals were informed by parent's preferences in accordance with normative sleep recommendations (e.g., a reduction in SOD [i.e., $\leq$ 15 min]; Hirshkowitz et al., 2015). Finally, parents' questions were discussed.

The interview allowed for the assessment of any risk factors that may have endangered the child's safety in any way or posed challenges to the research process. The limits of confidentiality were discussed prior to the commencement of the interview. The SATT was used to guide the clinical interview; specifically, to help to categorise children's sleep problems (e.g., SOD, NWs), as well to identify the antecedents, consequences and environmental variables contributing to sleep problems for each child. An example of clinical interview content included is included in Appendix J[4]. Pre-treatment psychometric measures were administered following the completion of the interview.

Case Conceptualisation and Treatment Planning

The outcomes of the sleep diaries, clinical interview and SATT informed a case conceptualisation and treatment plan for each child. VSG-data was also used to inform the case conceptualisation for two children (in Study 1) where parent-report was not attainable

[4] Clinical interview questions pertaining specifically to stereotypy are presented in Appendix L

(e.g., parents were unaware of the child's behaviour). Parents were provided a written copy of the case conceptualisation and treatment plan for their child, and this was discussed with them to ensure consensus (Sanders & Burke, 2014), with modifications made to the treatment plan with parent input as required. Resources needed for intervention (e.g., social stories) were also discussed with families, then prepared and posted to them.

Baseline

Baseline commenced following assessment, except for two children in Study 1, where (owing to the pilot nature of the study) FBA was completed during baseline (i.e., baseline ran continuously from assessment to the start of intervention). Consistent with the non-concurrent multiple-baseline design, children were randomly allocated a baseline length of one, two or three weeks, which was sometimes amended for practical reasons (e.g., to accommodate family needs). Individual baseline lengths are recorded under the Method section for each participant within each study. During baseline, parents were instructed not to alter their child's sleep routine, nor their responses to their child's sleep-related behaviour.

Intervention

Function-based individualised BSI commenced immediately following the end of the baseline phase. The treatment components for each child are described in the Method section within each study. During intervention, parents were provided with intensive (daily-weekly as required) support through remote (phone, Skype or email) contact with the author, to document daily progress toward desired goals, to resolve any problems as soon as they occurred, and to enhance treatment adherence (Sanders & Burke, 2014). To revise a treatment plan, the original hypotheses developed through FBA were reconsidered as to what other factors were likely to be maintaining problem behaviour, and/or parents' ability to adhere to the treatment plan and/or child response to treatment were considered. The treatment plan

was then adjusted accordingly, in agreement with parents. The intervention phase ended when parents felt their sleep goals had been met. If a child became sick at any point throughout the study, parents were told to prioritise the needs of their child and to attend to them as they normally would during a course of sickness.

Child-specific social 'sleep' stories, following the conventions outlined by Gray and Garand (1993), were developed and used within the BSI for 10/12 participant children. Social stories were paper-made and included photos and text illustrating changes to the child's bedtime routine and expectations around sleep and placed an emphasis on sleep-conducive behaviours (e.g., lying quietly in bed). Parents read the social story to their child each night as part of the bedtime routine, or at any other time upon request.

Maintenance

Once the intervention phase was complete, participants moved into the maintenance phase. During this phase, parents were asked to respond to their child as they had been during treatment, and regular contact with the research team and data collection temporarily ceased. The maintenance phase allowed for both child and parent behaviour to continue to consolidate; that is, to allow the newly acquired behaviours to become practiced and habitual (Blampied, 2013b).

Follow-up

Gathering follow-up data is critical to assessing the durability of changes over time and is a recommended component of single-case research (Heyvaert et al., 2014). Short-term follow-up (STFU) and long-term follow-up (LTFU) data was gathered 3-8- and 10-12-weeks post-intervention, respectively; however, for some children, follow-up data were recorded outside of these times owing to extraneous circumstances (e.g., parental illness). Parents recorded a sleep diary for a period of one week during each STFU and LTFU period, and

VSG was recorded for 3-4 nights of STFU to provide IOA for the data collected. IOA was not calculated for LTFU because VSG was not recorded during LTFU.

Data Analysis

Data obtained through sleep diaries and VSG during the baseline, intervention and follow-up phases were graphed according to the different dependent variables for each child. The primary means of data analysis was systematic visual inspection of graphed data, as a comparison between the study phases (Blampied, 2013b; Carter, 2013; Hanley et al., 2003; L. Cohen, Feinstein, et al., 2014). Visual analysis is an effective means for assessing treatment outcomes and is commonly used within a single-case design (Blampied, 2013b; Hanley et al., 2003; Parker & Hagan-Burke, 2007). Visual analysis of graphs included assessment of change in the trend, level, and stability of data between phases (Kazdin, 1982; L. Cohen, Feinstein, et al., 2014; Lane & Gast, 2014). Numbers on the graphs in Studies 1-3 have been used to indicate the value of data points that are out of range; this practice was used to avoid compressing the Y-axis scale unduly owing to outlier scores. Treatment was considered effective when systematic changes in the measured variables were clearly related to the implementation of intervention, which may have included treatment modifications within phases, in comparison to the baseline condition (Kazdin, 2001).

In addition, descriptive statistics were employed to strengthen the inferences made regarding treatment effects. Effect size estimates (i.e., to determine the strength of the relationship between treatment and target behaviour change) were calculated using non-overlap methods, which assess the extent to which intervention and follow-up data do not overlap with baseline data (Carter, 2013; Parker & Vannest, 2009). The greater the percentage of non-overlapping data, the greater the behavioural change between the baseline and treatment conditions (Parker & Vannest, 2009). For Studies 1-3 in this thesis, computation of the percentage below the median (PBM) was used to examine the degree of

behaviour change (Ma, 2006; Parker et al., 2011). PBM was used instead of percentage exceeding the median (PEM) because the direction of therapeutic change was a reduction in scores. To calculate PBM, the baseline median is projected into the treatment phase, and then the number of intervention data points that fall below the median value is counted and expressed as a percent of all the data points in that phase. For example, if 20/20 data points fell below this line, PBM = 20/20 = 100%. If 10/20 data points fell below the line, PBM = 10/20 = 50%. These scores are interpreted as follows: > 90%: high effectiveness; 70%-90%: moderate effectiveness; < 70%: ineffective treatment (Ma, 2009).

An assumption of PBM is that the median is a good representation of baseline scores (Parker et al., 2011). Accordingly, if baseline data have high variability or a strong trend up or down, then PBM may not be an appropriate measure of effect size. In addition, a criticism of PBM is that it is insensitive to the magnitude of behaviour change; that is, data points are categorically calculated as falling above or below the median of the baseline phase, without consideration of how far above or below they fall (Carter, 2013; Ma, 2006; Wolery et al., 2010). This criticism is mitigated when PBM is used in conjunction with visual analysis, which examines the immediacy of treatment effects, trend, level, and stability of data, in the context of consideration of experimental control (Carter, 2013).

Despite its acknowledged limitations, PBM, which combines median level change statistics with non-overlap methods, is regarded as a useful measure and is a commonly used effect size in single-case research (Parker et al., 2011; Roth et al., 2014), including meta-analysis of single-case studies (Ma, 2009; Parker et al., 2011; Preston & Carter, 2009). Median level change is a more accurate effect size than mean level statistics, which often do not appropriately summarise data sets in single-case research (Parker et al., 2011). PBM was used in conjunction with visual analysis in Studies 1-3 to determine the degree of behaviour change and whether effects were functionally related to intervention (Carter, 2013; Kazdin,

2001). Change in pre/post-intervention psychometric measures were assessed using modified Brinley plots (Blampied, 2017), which are reported and discussed separately in Chapter 10.

Chapter 4: Study 1

An Evaluation of the Effectiveness of Function-Based Behavioural Interventions to Treat Sleep Problems in Children on the Autism Spectrum: A Pilot Study

This chapter presents Study 1, a pilot study examining the effectiveness of function-based BSI for six children on the autism spectrum[5]. In this study, FBA was used to identify the antecedent- and consequence-based factors maintaining sleep problems for each child. Stereotypy accompanied sleep problems in four children. FBA was then used to develop an individualised case conceptualisation and treatment plan. This study aimed to answer the following research questions:

1. Can function-based behavioural interventions effectively reduce sleep problems in children on the autism spectrum?
2. Are treatment effects maintained in the short- and long-term?
3. Are the selected treatment approaches acceptable to parents, and implemented with fidelity?

Method

Participants and Setting

Six families living in four urban centres in NZ participated; child participants included two girls and four boys, aged 3-10 years (mean = 5.5 years), who met the inclusion criteria described in Chapter 3. All children had communication difficulties (as defined in Chapter 3). One child (Nikolay) was not administered the VABS-II; however, clinical judgement in accordance with VABS-II criteria classified his communicative ability as low. Table 4.1 presents a summary of participant characteristics; all names are pseudonyms. Intake

[5] Data pertaining to two children 'Jorge' and 'Mirasol' in this study have been published in *Clinical Case Studies* (McLay et al., 2017) and *International Journal of Developmental Disabilities* (McLay, France, Blampied et al., 2019), respectively.

assessment, treatment planning discussions, and the administration of pre- and post-treatment questionnaires were conducted in accordance with the procedures outlined in Chapter 3. For 3/6 participant families, the author travelled to the family home to conduct the intake interviews.

Table 4.1. *Summary of Participant Characteristics at the Time of Pre-Treatment Assessment*

Participant	Age (Y-M)	Sex	Ethnicity	Diagnosis	VABS-II		Medication	Number of parents in household
					Receptive (Y-M)	Expressive (Y-M)		
Emma	7-6	Female	NZE	ASD, GDD	2-6	0-1	-	2
Jorge	3-9	Male	NZE	ASD	1-3	2-0	-	2
Mirasol	10-10	Female	Filipino/ NZE	ASD	2-6	1-9	Risperidone	1
Nikolay	6-2	Male	Russian	ASD	-	-	-	2
Davie	3-6	Male	NZE	ASD	1-2	1-8	Melatonin	2
Max	5-2	Male	Maōri/ NZE	ASD	2-2	3-7	-	1

Note. ASD: autism spectrum disorder; GDD: global developmental delay; M: months; NZE: New Zealand European; VABS-II: Vineland Adaptive Behavior Scales 2nd Edition; Y: years

Measures and Materials

Functional Behaviour Assessment

In addition to the FBA procedures outlined in Chapter 3, VSG was used to gather data to inform FBA for two children (Emma and Max). This data was not attainable via parent-report (e.g., child behaviour during SOD and NWs, the frequency and duration of NWs).

Stereotypy

VSG was also used to measure stereotypy for Emma and Max. Stereotypy was not measured for other children because it either did not occur (Jorge and Nikolay) or was not a

dependent variable (Mirasol and Davie). The duration (seconds) of all instances of stereotypy for each child were coded from the point at which repetitive behaviour began, until the child ceased the behaviour (e.g., the child lay still, or transitioned to another activity). The total duration of stereotypy in min (rounded to the closest min) was then calculated for each child; if the total duration of stereotypy was < 30 s (on a single night, across all phases) then it was considered non-problematic and was not graphed. Only stereotypy that was observable (i.e., motor stereotypy and RMO) could be recorded because the VSG technology used at the time did not support audio recordings.

Procedure

Design

A single-case AB (baseline [A], intervention [B]) design with short- and long-term follow-up phases was used to examine treatment effects, with replication of treatment effects across target variables including SOD, CCs, and NWs. Additional intervention phases (Jorge, Nikolay, Davie, and Max) are indicated by phase-change lines labelled Treatment 1 (T1) Treatment 2 (T2) and so on (see Figures 4.3, 4.5-4.7).

Baseline

Max was randomly allocated a baseline period of seven nights. For the other five children, baseline lengths were determined dependent on the time it took for a stable trend of data to materialise from any variability of behaviour (Blampied, 2013b; Kazdin, 1982) and/or to ensure that the conclusion of baseline was commensurate with the beginning of intervention (Carter, 2013). Jorge, Mirasol, and Nikolay were allocated a baseline length of 14 days. For Emma and Davie, baseline data were collected for 40 and 56 days, respectively, during which the FBA was completed. Davie's baseline was extended because of illness.

Function-Based Case Conceptualisation and Treatment Planning

Consistent with the general procedure described in Chapter 3, information gathered in the FBA was synthesised into a function-based case conceptualisation and used to develop a function-based individualised treatment plan for each child (Blampied, 2013a). The problem behaviours, precipitating and maintaining factors, hypothesised functions of the problem behaviour, and individualised treatment plan each child are summarised in Table 4.2, and discussed below. SOD and NWs were present for all six children, except Emma, for whom NWs were not a problem. Additional problem behaviours included bedtime resistance (*n* = 3), unwanted co-sleeping (n = 3), stereotypy (*n* =4), CCs (e.g., calling out to parents) and other disruptive behaviours (e.g., banging on the wall, roaming the house; *n* = 3). The FBA revealed that the problem behaviours served a variety of functions across and within children, including access to parental attention, access to tangible items, escape from the demand to go to sleep, and automatic reinforcement (i.e., motor and vocal stereotypy). Additional precipitating and maintaining factors included poor sleep hygiene (e.g., inconsistent bedtime) and insufficient sleep pressure.

Intervention

Parents' intervention goals included: (1) a reduction in sleep onset time (i.e., for SOL to be within 15-30 min); and (2) the elimination of sleep-interfering behaviours (including bedtime resistance); (3) NWs; and (4) co-sleeping. In addition, Jorge's parents wished to eliminate night-time breastfeeding, and Nikolay's parents wished for him to learn to sleep in his own bed in his own bedroom.

Table 4.2. *Summary of Problem Behaviours, Precipitating/Maintaining Factors, Hypothesised Functions and Method of Intervention*

	Problem behaviours	Hypothesised precipitating/maintaining factors	Hypothesised functions	Intervention
Emma	SOD	Access to preferred items; stereotypy	Automatic	Lightbulb removed; dresser drawers secured closed; magazines removed from bedroom (except one); finished box; pre-bedtime access to magazines (satiation); social story; positive reinforcement
Jorge	CCs	Inconsistent bedtime routine & bedtime; parent attention; breastfeeding	Escape (from bed); attention; tangible	Consistent routine & bedtime (SH); breastfeed prior to bed (satiation)/elimination of night-time breastfeeding; sleep item; PP; PI; social story; positive reinforcement
	NWs	Parent attention; breastfeeding	Attention; tangible	Sleep item; PP; PI; social story; positive reinforcement
Mirasol	SOD	Inconsistent bedtime routine and bedtime; insufficient sleep pressure; difficulty transitioning between tasks; parent attention	Escape from bed; attention	Consistent routine & bedtime (SH); calming pre-sleep activities; set wake time (sleep restriction); visual sign; PI; social story; positive reinforcement
	NWs	Decreased sleep pressure; access to preferred items; parent attention; stereotypy	Tangible; attention; automatic	Set wake time (sleep restriction); removal of lightbulb; prevented access to food & toys; visual sign; PI; white noise; social story; positive reinforcement
Nikolay	SOD	Inconsistent bedtime routine & bedtime; insufficient sleep pressure; tantrum behaviour; access to toys; parent attention	Escape from bed; tangible; attention	Consistent routine & bedtime (SH); set wake time (sleep restriction); visual schedule; sleep item; bedtime pass & PI; positive reinforcement

	CCs	Inconsistent bedtime routine & bedtime; insufficient sleep pressure; tantrum behaviour; access to toys; parent attention	Escape from bed; tangible; attention	Consistent routine & bedtime (SH); set wake time (sleep restriction); visual schedule; sleep item; bedtime pass & PI; positive reinforcement
	NWs	Insufficient sleep pressure; inconsistent S^D for sleep (multiple sleep locations); access to toys; parent attention	Tangible; attention	Consistent sleep location (SH); set wake time (sleep restriction); sleep item; Gro-clock™; bedtime pass & PI; positive reinforcement
Davie	CCs	Parent attention	Escape from bed; attention	Pre-bedtime access to reinforcement for sleep-interfering behaviour (satiation); faded bedtime procedure; PI; positive reinforcement
	SOD	Parent attention; stereotypy; disruptive behaviour	Attention; automatic	Pre-bedtime access to reinforcement for sleep-interfering behaviour (satiation); white noise; faded bedtime procedure; PI; positive reinforcement
	NWs	Parent attention; stereotypy; access to food & drink	Attention; automatic; tangible	White noise; faded bedtime procedure; PI; positive reinforcement
Max	SOD	Bedroom TV; insufficient sleep pressure; stereotypy	Automatic	Removal of bedroom TV (night light replacement); pre-bedtime access to preferred TV show (satiation); faded bedtime procedure & set wake time (sleep restriction); Gro-clock™; social story; positive reinforcement
	NWs	TV playing in bedroom; insufficient sleep pressure; stereotypy	Automatic	Removal of TV from bedroom (night light replacement); pre-bedtime access to preferred TV show (satiation); faded bedtime procedure & set wake time (sleep restriction);

	Gro-clock™; social story; positive reinforcement

Note: BR: bedtime resistance; CCs: curtain calls (i.e., bids for parental attention); Co-S: co-sleeping; PP: parental presence procedure; PI: planned ignoring; NWs: night wakings; SH; sleep hygiene; SOD: sleep onset delay; TV: television

Emma. Emma had a consistent bedtime routine, bed and wake time; however, she had substantial SOD. After being bid goodnight, Emma turned on her bedroom light and engaged in RMO involving flipping magazine pages, mouthing rolled up magazines, and browsing a pile of magazines in her bedroom. She would also pull out the drawers from her bedroom dresser, remove the clothing contents, stack the drawers and climb on them. Before falling asleep, she wrapped herself in her duvet and body rocked in bed until sleep onset occurred. Emma's parents woke her each morning and reported that she was typically difficult to wake. They expressed concern that a lack of sleep was affecting her daily functioning.

The FBA indicated that Emma's sleep onset was primarily delayed because of her engagement in stereotypy, which actively interfered with behavioural quietude and the onset of sleep. Stereotypy (RMO and body-rocking) was hypothesised to be maintained by automatic (i.e., non-social) reinforcement, as the behaviours occurred when she was alone in her bedroom. Access to antecedent stimuli in her bedroom (magazines, dresser drawers) afforded opportunities for manipulation (page-flipping, stacking, and climbing). Turning on the bedroom light increased the salience of these antecedent stimuli, and a brightly lit bedroom was not conducive to quality sleep.

Emma's treatment ran for 55 days. Environmental modifications included the removal of her bedroom lightbulb to ensure her bedroom remained dark (except for a small amount of natural light). This meant she was no longer able to see objects that provided her with

stimulation, and improved natural cues for sleep (i.e., darkness). In addition, her parents secured her dresser drawers closed, to revoke access to her stacking and climbing on the drawers. Finally, all magazines were removed from her bedroom and relocated to the lounge, except for one that she selected to take to bed in accordance with parents' preferences.

Emma's parents were instructed to allow her to engage in RMO with magazines in the lounge prior to bed, in order to increase the likelihood of her satiating on the magazines (i.e., reducing the value of the reinforcer), thereby decreasing her motivation to engage in RMO after being bid goodnight (i.e., an AO; Jin et al., 2013; Michael, 1982). Emma was taught to then tidy the magazines into a storage box with a 'finished' symbol on the lid. Access to the magazines was removed until the following morning, at which time she was given access to the magazines, non-contingent on the previous night's sleep.

A social story was introduced, which reflected the environmental changes of intervention. For example, the story included how her parents would keep her magazines in a safe place over night, and that she could choose one special magazine to take to bed.

Jorge. Jorge's bedtime and bedtime routine were inconsistent, and he resisted going to bed. His sleep problems included CCs, and frequent and prolonged NWs. Prior to initial sleep onset and during NWs, he would leave his bed, demand attention, demand to be breastfed, and would scream if he was not responded to. During NWs, he would seek out his mother who would typically lie with him and breastfeed him to resettle him to sleep. At the time that the study commenced, Jorge was breastfed throughout both the day and night.

The FBA revealed multiple factors were likely to be contributing to Jorge's sleep problems. Antecedent factors included an inconsistent bedtime routine and bedtime (i.e., poor sleep hygiene) contributing to bedtime resistance and CC behaviour. CCs and NWs were positively reinforced through parent attention. In addition, CCs were negatively reinforced

through bedtime avoidance, and NWs were complicated by his reliance on parental presence to settle to sleep (inappropriate stimulus control). The presence of Jorge's mother may have also provided S^D for breastfeeding demands. In the initial FBA it appeared that breastfeeding provided tangible reinforcement that maintained his disrupted sleep. However, owing to the continuation of NWs despite the elimination of night-time breastfeeding, the FBA was later revised to identify parent attention as the primary reinforcer for NWs. Jorge's treatment ran for 53 days. Procedural changes were made during intervention, resulting in four main treatment phases.

Phase 1. Phase 1 included establishing a consistent bedtime and bedtime routine in which Jorge was breastfed before bed to satiate his need to be breastfed after going to bed (i.e., an AO). A sleep item in the form of a favourite soft toy was introduced, which Jorge could be redirected to cuddle if he woke during the night. It was agreed that night-time breastfeeding would be eliminated (extinction) so that Jorge would no longer be breastfed upon being put to bed or during NWs. Jorge was able to be breastfed when he woke each morning after 6:00 a.m. If he requested breastfeeding at night, his parents were instructed to resettle him into bed and redirect him to cuddle his soft toy.

A parental presence procedure was used to eliminate co-sleeping, so that he could learn to settle to sleep independently. During this procedure, Jorge was bid goodnight then a parent remained on a mattress in his room without engaging with him, until he settled to sleep. If he left his bed, the parent returned him immediately to his bed with a verbal prompt to redirect him to sleep (e.g., "sleep time now Jorge"). Once he was asleep, the parent left his room to resume their evening, returning to sleep on the mattress in Jorge's room overnight. If he woke during the night, the parent feigned sleep without interaction until he resettled independently or returned him to his bed (with verbal redirection for sleep) as required.

A social story was introduced that reflected the changes under intervention (e.g., the parent on the mattress) and portrayed him sleeping all night in his own bed, with a parent present and without breastfeeding. The social story also described a reinforcement procedure, wherein he received social praise and a voucher (for 15 min "special fun time" with parents) contingent upon a successful night sleep.

Phase 2. Phase 2 involved further modification to the parental presence procedure; specifically, verbal prompts were removed to further control for reinforcement that Jorge may have received through interaction with parents. Instead, parents returned him to bed using only physical gestures and prompting, without verbal interaction.

Phase 3. A barrier to sleep onset was identified wherein Jorge consistently passed a bowel motion and was then showered by his parents after being put to bed, delaying his sleep onset; this appeared to be maintained by parent attention. Procedural modifications were made and reflected in the social story, wherein he toileted prior to bed and showered in the morning. Jorge was given small rewards (e.g., stickers) for sitting on the toilet before bed.

Phase 4. Final procedural modifications included the full elimination of parental presence, with this change reflected in a new social story. Instead of sleeping on the mattress, his parents bid him goodnight and left the room, ignoring any bids for attention, and sleeping in their own bed overnight. If he awoke during the night, his parents redirected him to sleep without verbal interaction, before returning to sleep in their own bed.

Mirasol. Mirasol's bedtime routine and bedtime were highly variable, and she was noncompliant with parent requests to get ready for bed. Her sleep problems included SOD, NWs, and unwanted co-sleeping. Mirasol's mother would typically lie with Mirasol until she fell asleep during the sleep onset period for 1-2 hours. Mirasol woke frequently during the night for prolonged periods and was unable to return to sleep unless co-sleeping occurred

with her mother lying in Mirasol's bed. During NWs, she engaged in vocal stereotypy and disruptive behaviour, including turning on the lights, roaming the house, searching for food, playing with toys, and bouncing on the bed. She woke at inconsistent times as her mother typically allowed her to sleep in.

The FBA indicated that Mirasol's sleep difficulties were likely to be multiply determined. Bedtime resistance, contributing to SOD, appeared to be perpetuated through variation in the bedtime routine activities, requests to transition from a preferred (e.g., watching television [TV]) to a non-preferred (i.e., getting ready for bed) task, and an inconsistent bedtime. In addition, decreased sleep pressure owing to an inconsistent wake time (i.e., being able to sleep in) contributed to difficulties initiating and maintaining sleep on subsequent nights. NWs were deemed likely to be positively reinforced by access to toys, food, and parent attention, and engagement in stereotypy. Stereotypy was hypothesised to be automatically maintained as it occurred when she was alone; however, as it occasionally resulted in parent attention (e.g., Mirasol's mother would verbally interact and lie with her), a second possible function of gaining parent attention was hypothesised. NWs were further complicated by a reliance on parental presence to resettle to sleep (inappropriate stimulus control).

Mirasol's treatment ran for 35 days and included establishing a set bedtime routine, bedtime, and morning wake time (i.e., sleep hygiene practices). Specifically, Mirasol's mother ensured that she did not engage with any stimulating activities (e.g., watching TV) that were difficult for her to transition away from prior to bed. Instead, her routine involved quiet, calming pre-sleep activities (e.g., doing a puzzle and reading with her mother). A consistent bedtime of 9.00 p.m. was determined based on the assessment results, which indicated this was the time closest to her typical sleep onset (i.e., she was likely to fall asleep

within 15 min of going to bed at that time). In addition to the delayed bedtime, a set wake time of 7.00 a.m. was introduced to increase physiological sleep pressure.

Environmental modifications included preventing access to food and toys overnight. The lightbulb was removed from her bedroom to eliminate sleep-interfering behaviour (i.e., repeatedly turning the light on and off) and to improve natural cues for sleep. In addition, a visual symbol (a picture of a bed) was placed on Mirasol's mother's door overnight, to signal that it was night-time and her mother was sleeping. This was replaced with a picture of a sun in the morning, to indicate that Mirasol could enter her mother's bedroom.

Planned ignoring (extinction) was used to eliminate the reinforcing effects of parental attention prior to sleep onset and during NWs. After bidding her goodnight, Mirasol's mother left the room and only returned if necessary for safety reasons. If Mirasol left her bedroom prior to sleep onset or during NWs, her mother redirected her back to bed with minimal interaction (e.g., without verbal interaction) and then left the room. Mirasol's mother ignored any behaviour that she engaged in whilst in bed, including stereotypy. In addition to planned ignoring, white noise was used to reduce the reinforcing value of vocal stereotypy by masking auditory feedback. A white noise generating device (an Audio Box FM MP3 player with speakers which played an MP3 file of the white noise) was provided to the family. Mirasol's mother was instructed to play the white noise at a starting level of 50 dB, and to gradually adjust to a maximum of 75 dB to determine a noise level which Mirasol was comfortable with (Knight & Johnson, 2014). Mirasol's mother turned on the device when Mirasol was in bed and turned off the device when she woke in the morning.

Finally, a social story was developed which laid positive emphasis on her sleeping independently through the night in her own bed and ended with her being rewarded in the morning (i.e., playing with toys with her mother) for a successful night sleep.

Nikolay. Nikolay had an inconsistent bedtime routine, bed and wake time. His sleep problems were CCs, SOD, NWs, and unwanted co-sleeping. Nikolay engaged in tantrum behaviour (e.g., shouting, throwing toys) in response to parent requests to return to bed. In response, his parents typically allowed him to continue to play with toys until he complied. Once in bed, he frequently demanded parent attention by leaving the bed or calling his parents into the room. He woke 1-3 times during the night for prolonged periods, during which he played with toys in his bedroom and in the lounge. Co-sleeping often occurred in response to NWs. Nikolay slept in one of three locations: his bed, his parent's bed, or a separate bed in his parent's bedroom. He woke on his own in the morning at inconsistent times.

The FBA determined that multiple factors were likely to be contributing to Nikolay's sleep problems. Antecedent factors included an inconsistent bedtime routine and bedtime, and insufficient sleep pressure owing to an inconsistent wake time. Positive reinforcement in the form of access to preferred items (toys) and parent attention, and negative reinforcement in the form of demand avoidance, were thought to be maintaining sleep problems. Further, the variation in sleep locations (e.g., his bed, his parent's bed) and his dependence on a parent's presence to resettle to sleep meant there were inappropriate and inconsistent S^D for sleep.

Nikolay's treatment lasted 45 days. From the start of intervention, he began sleeping in his own bed in his own bedroom, so that he could learn to settle to sleep independently and to improve stimulus control for sleep (i.e., by establishing his own bed as S^D for sleep). In treatment phase one (T1) a sleep item was introduced, whereby he was able to choose one of two soft toys presented by his parents to take to bed each night. Access to other toys was removed to reduce reinforcement for bedtime resistance and NWs. A visual schedule (depicting a 'first/then' contingency, e.g., *first* eat dinner, *then* brush teeth) was developed to

support him complying with the bedtime routine. This depicted two steps at a time. Parents provided social reinforcement for completing steps in the bedtime routine. The visual schedule ended with a picture of him going to sleep, followed by a picture of a reward to be gained when he woke in the morning.

A consistent bedtime of 9.00 p.m. was established, as this was the time that he was likely to fall asleep within 15 min of going to bed. In addition, a set wake time of 8.00 a.m. was selected to maintain sleep pressure A Gro-clock™ (a digital clock that displays a star/sun on its face, to represent sleep/wake times) was placed next to his bed each night, which acted as a visual cue for him to return to sleep if he woke during the night. Nikolay's parents verbally instructed him each night that he could get out of bed when he saw the sun on the Gro-clock™ in the morning (i.e., at 8.00 a.m.).

A bedtime pass procedure (Friman et al., 1999) was implemented to address sleep-interfering behaviour after Nikolay was put to bed. He was given access to two bedtime passes per night; initially, one pass was intended for use during a NW, however, Nikolay's parents chose to use both passes for behaviour that occurred prior to sleep onset. If Nikolay required a pass (e.g., leaving his bedroom or calling a parent into his room) a parent would resettle him, then state "you have used your bedtime pass now" and ensure Nikolay saw that the pass was removed. Once both passes were used, Nikolay's parents ignored subsequent bids for attention. If he left his bedroom, his parents immediately redirected him to his own bed without verbal interaction.

In the morning, his parents presented him with a menu of three highly preferred items as reinforcement contingent on Nikolay remaining in his own bed all night. A second treatment phase (T2) involved providing parents with further instructions to reduce social interaction at bedtime (i.e., they were instructed to bid him goodnight with a clear, simple instruction, "it's time to sleep now") and to leave the room.

Davie. Davie had a consistent bedtime routine, bedtime, and morning wake time. His sleep problems included CCs, SOD, NWs, and vocal stereotypy. After being bid goodnight, he engaged in call out requests, vocal stereotypy, and disruptive behaviour such as kicking the wall and jumping on the bed. He regularly woke at least once during the night for a prolonged period, during which he engaged in vocal stereotypy, repeating words and song phrases to himself. Davie's parents responded to his NWs in a variety of ways, including ignoring the behaviour, changing his nappy, offering food and drink in the lounge, lying with him, and rubbing his back.

The FBA indicated that Davie's sleep problems were likely to be multiply determined. Sleep-interfering (stereotypic and disruptive) behaviours during the sleep onset period were likely to be reinforced by parent attention and automatic (non-social) consequences. NWs were thought to be maintained through his engagement in vocal stereotypy, intermittent tangible reinforcement (e.g., food, drink) and parent attention. Vocal stereotypy was hypothesised to be automatically reinforced, and in addition, may have functioned to produce social consequences given the behaviour (particularly during NWs) frequently resulted in parent attention (e.g., rubbing his back).

Davie's treatment ran for 53 days and involved two treatment phases. First, white noise was used to target vocal stereotypy in isolation, as vocal stereotypy was a primary concern for Davie's parents that precipitated their response to his NWs. White noise was played on a portable device, following the guidelines as outlined for Mirasol above (Knight & Johnson, 2014). The second phase of treatment involved a faded bedtime procedure in which his bedtime was delayed until 8.45 p.m. During this time, he received a light snack, a drink of water, a nappy change, and undivided attention from his parents, intended to satiate his motivation for sleep-interfering behaviours (i.e., an AO) and to ensure any needs he had were addressed prior to bed.

Once in bed, Davie's parents reduced the variety and intensity of their responses to sleep interfering behaviours. For example, his parents guided him back to bed without giving him food. The introduction of phase two was aligned with Davie moving into his own bedroom, which presented an opportunity to establish the new bedroom as S^D for sleep. Social reinforcement (e.g., praise from his parents) was provided upon waking, contingent upon a good night sleep.

Max. Max had a consistent bedtime routine and bedtime. His sleep problems were SOD and NWs. A large TV faced Max's bed in his bedroom, which played a single episode from a preferred show on repeat all night. Max took approximately two hours to fall asleep, during which time he watched TV and infrequently engaged in motor stereotypy (i.e., moving/waving his hands and arms in front of his face, and body rocking). He woke at least once during the night for prolonged periods, during which time he remained in bed, again watching TV, and engaging in stereotypy. He woke on his own between 9.00–10.00 a.m.

FBA indicated that a primary factor likely to be maintaining Max's sleep problems was the presence of a TV in his bedroom. Audio and visual stimulation, and blue light emitted from the television, were non-conducive to quality sleep. In addition, Max spent up to 15 hours in bed (7.00 p.m.–10.00 a.m.), resulting in fragmented sleep and reduced sleep pressure, and a lack of stimulus control for sleep (i.e., the bed and bedroom environment were not established S^D for sleep).

Max's treatment ran for 106 days. Treatment phase one entailed removing the TV from his bedroom, to improve sleep hygiene (e.g., to eliminate blue light, audio and visual stimulation). A personalised night light was provided to the family, as a more adaptive (sleep-conducive) replacement for the TV. Max was permitted to watch his preferred show on the TV in the lounge up until he commenced his bedtime routine, to help to satiate his desire for TV (i.e., an AO). A faded bedtime procedure and a set wake time were used to increase

physiological sleep pressure (i.e., an EO, by preventing the dissipation of sleep pressure owing to sleeping in), and to improve stimulus control for sleep (i.e., via reduced time spent in bed awake).

A Gro-clock^{TM} was used to clearly signal the morning wake time, and his parent helped to rouse him at this time (e.g., by opening the curtains and bidding him good morning). He was given immediate access to a preferred activity in the lounge upon waking, to help him to transition out of bed without resistance. Finally, a social story and a reinforcement procedure where Max was given immediate access to a preferred activity in the lounge upon waking were included to reinforce appropriate behaviour. A second treatment phase involved tightening up the sleep restriction procedure to reduce variability still occurring with Max's bed and wake time, by re-establishing the time Max went to bed (8.00 p.m.) and was woken (7.15 a.m.)

Maintenance and Follow-up

A maintenance phase was established once parents felt their treatment goals had been met, as described in Chapter 3. STFU data was collected (as described in Chapter 3) for seven nights at 44, 44, 43, 27, 174, 26 days post-intervention for Emma, Jorge, Mirasol, Nikolay, Davie and Max, respectively. LTFU was collected at 79, 79, 81 and 208 days post-intervention for Emma, Jorge, Davie and Max, respectively. LTFU was not collected for Mirasol or Nikolay because the families were unable to be contacted.

Data Analysis

As described in Chapter 3, data were examined using visual analysis of the graphed data with assessment of change in level, trend and variability across phases and cases (L. Cohen, Feinstein, et al., 2014). The treatment results for Emma, Jorge, Mirasol, Nikolay, Davie2, and Max are presented individually in a case study format in Figures 4.1-4.6,

respectively. PBM was used as an effect-size measure of single-case data to supplement visual analysis. For children who had more than one treatment phase (Jorge, Nikolay, and Davie), the final treatment phase was used to calculate PBM. PBM was calculated using sleep diary data, however, where data was unavailable then VSG data was used, as indicated in the results section for each child. The PBM scores for the six children are presented in Table 4.3.

Results

Data Quality

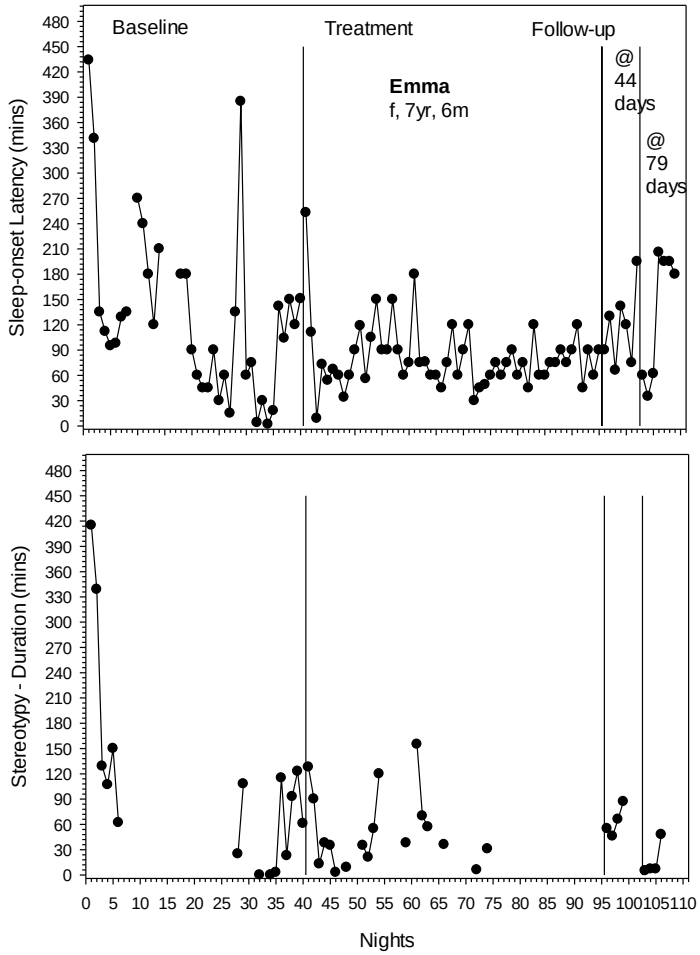
Parents were asked to record sleep diaries daily; however, diaries were sometimes incomplete or were not recorded by parents, who cited time demands as the primary reason for failing to complete diaries. For Jorge, CC and SOD data were incomplete or absent during baseline, and only 4/7 nights of data were recorded for all variables during LTFU. For Emma, IOA was unable to be calculated owing to the absence of parent-reported data (i.e., parents were unaware of her behaviour). Missing data points during baseline represent nights where VSG data were not recorded. Stereotypy data for Emma were scarce as she often moved out of the range of the camera and her behaviour was therefore not always recorded. For Max, VSG was used to assess sleep outcomes because his solo parent struggled to maintain sleep diaries and Max did not always signal when he was awake (e.g., his quiet awakenings). For this reason, IOA was limited to 8% of the treatment phase. Missing data points during treatment represent nights where VSG data were not recorded. Follow-up data for Max were recorded by diary (without VSG) which is why stereotypy data were unable to be reported during his follow-up.

Emma

The dependent variables for Emma were the duration of SOL and duration of stereotypy (RMO with magazines, body rocking), and are presented in Figure 4.1. During

baseline, SOL was highly variable (range = 2-434 min), with a trend that appeared to be increasing toward the end of baseline, although not to a level exceeding that observed earlier in baseline. During treatment, the variability and level of SOL reduced to an average range of 60-90 min, with a PBM score of 86% demonstrating a moderate treatment effect. This effect was not maintained during the follow-up phases (PBM = 43%). During treatment, the duration of stereotypy reduced compared to baseline, with a moderate treatment effect (PBM = 83%). Stereotypy remained at reduced levels throughout follow-up, with a large treatment effect (PBM = 100%).

Figure 4.1.



Sleep Outcomes for Emma: Duration of Sleep Onset Latency and Stereotypy across Baseline, Intervention, and Follow-up Phases. Treatment = environmental modifications, finished box, pre-bedtime satiation (magazines), social story, positive reinforcement

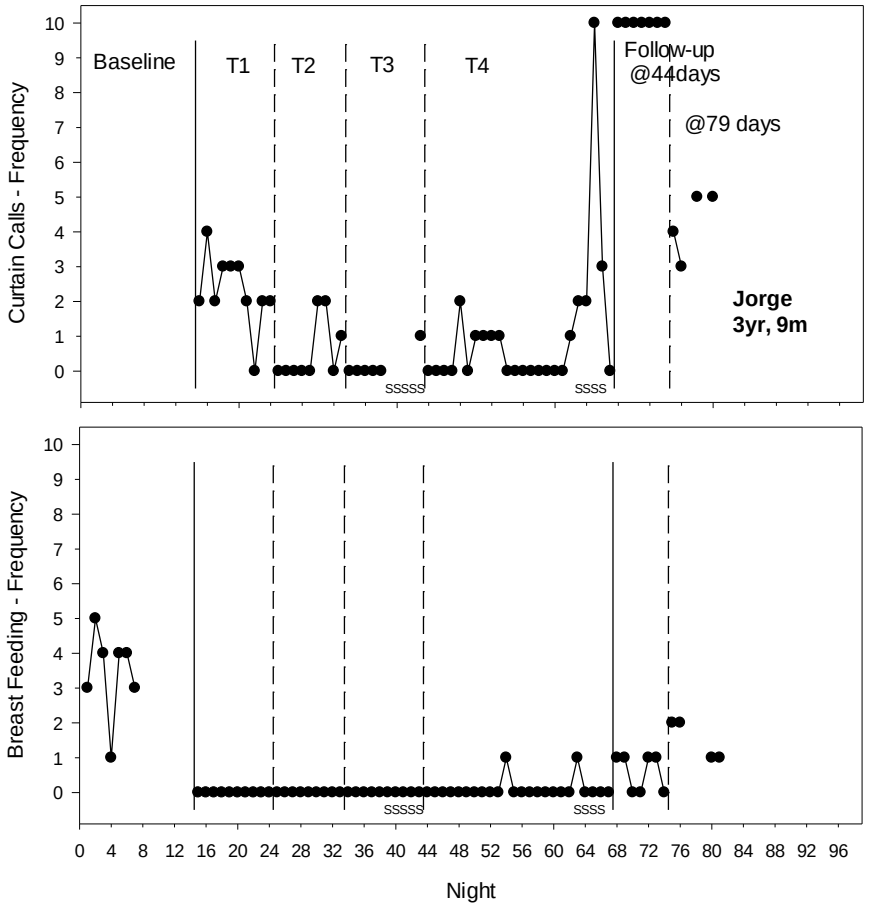
Jorge

The dependent variables for Jorge were the frequency of CCs and night-time breastfeeding, and the frequency and duration of NWs (see Figures 4.2 and 4.3). The 'S' represents nights when Jorge was sick. Night-time breastfeeding occurred frequently during baseline (range = 1-5/night). Results show the immediate elimination of night-time breastfeeding throughout treatment (PBM = 100%), although data are absent from the second half of baseline. Night-time breastfeeding was evident during follow-up, however, the frequency (range = 0-2, median = 1) remained clearly reduced compared to baseline (range = 1-5, median = 4; PBM = 100%).

Interpretation of the frequency of CC data is limited by baseline data being missing. However, the trend during treatment is for a reduction in the level recorded in treatment phase one (T1), except during the period of sickness at the end of phase four (T4). At STFU, however, the frequency of CCs was elevated to a level worse than at baseline (10 per night) and remained frequent (range = 3-5/night) during LTFU.

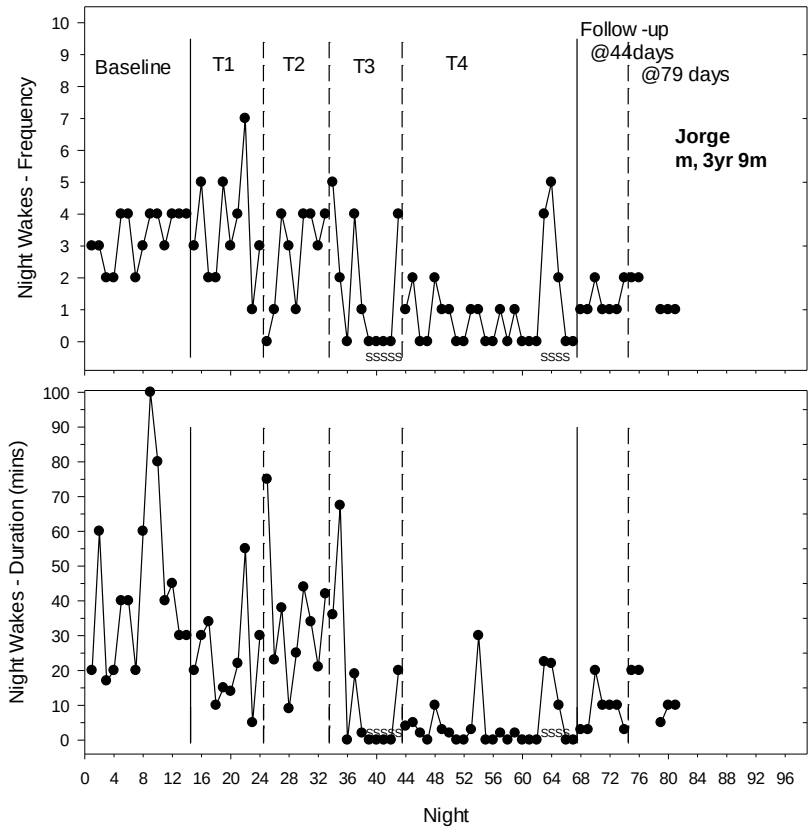
There were clear treatment effects evident for a reduction in the frequency and duration of NWs during intervention (PBM = 100%), with these effects maintained throughout follow-up (PBM = 100%). During baseline, Jorge woke three times per night (range = 2-4) on average for a variable duration (range = 15-100 min). The frequency of NWs increased in variability during treatment phases one (T1) and two (T2), although the overall level of waking remained unchanged. In treatment phase three (T3) the trend was a reducing frequency, and this trend was sustained in treatment phase 4 (T4) except during a period of sickness. The overall pattern of the duration of NWs mirrored that of frequency, but with greater variability.

Figure 4.2.



Sleep outcomes for Jorge: Frequency of Curtain Calls and Frequency of Night-Time Breastfeeding across Baseline, Intervention, and Follow-up Phases. The 'S' represents nights when Jorge was sick. T1 = sleep hygiene, sleep item, pre-bedtime satiation (breastfeeding), extinction (breastfeeding), parental presence, social story, positive reinforcement; T2 = modification to parental presence; T3 = toileting before bed; T4 = planned ignoring

Figure 4.3.



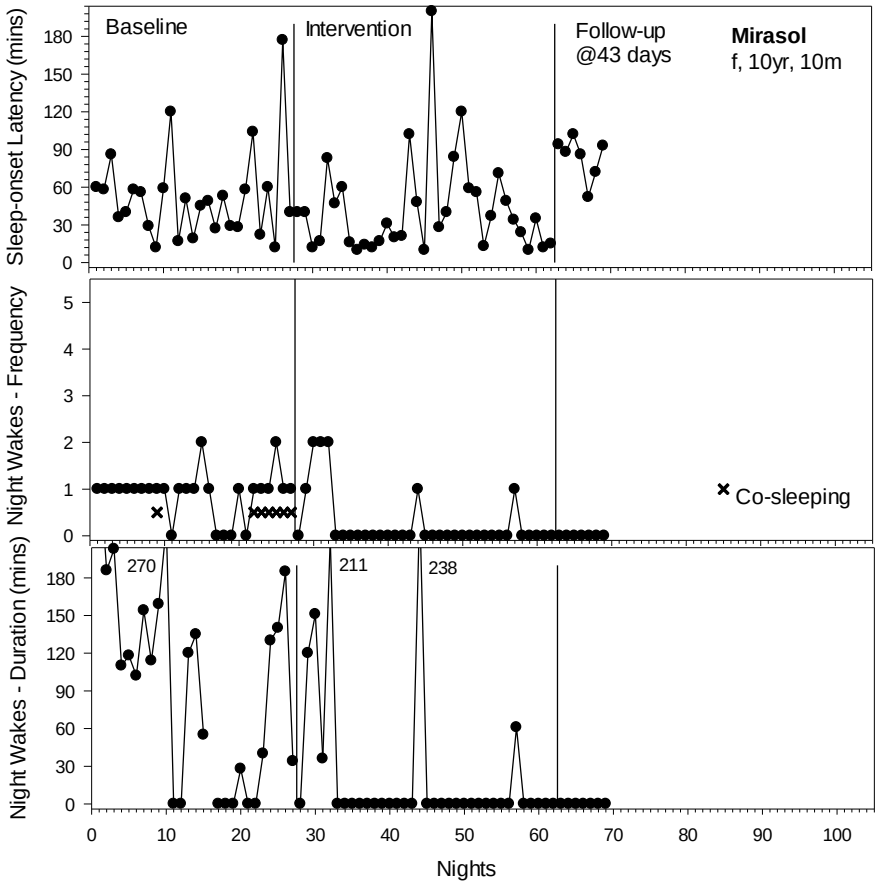
Sleep Outcomes for Jorge: Frequency and Duration of Night Wakings across Baseline, Intervention, and Follow-up Phases. The 'S' represents nights when Jorge was sick. T1 = sleep hygiene, sleep item, pre-bedtime satiation (breastfeeding), extinction (breastfeeding), parental presence, social story, positive reinforcement; T2 = modification to parental presence; T3 = toileting before bed; T4 = planned ignoring

Mirasol

The dependent variables for Mirasol were duration of SOL, and the frequency and duration of NWs (see Figure 4.4). The 'X' signifies co-sleeping during NWs. The numbers on the graph represent the value of data points that are out of range. During baseline, the average duration of SOL was 52 min (range = 10-175 min). There was a slight decrease in the overall duration of SOL during treatment, with the PBM score (71%) demonstrating a moderate treatment effect. This effect was not maintained during STFU (PBM = 0%). It is of note however, that Mirasol's mother ceased lying with her to settle Mirasol to sleep under intervention, which may have affected the time taken for her to fall asleep.

During baseline, Mirasol woke on average once per night (range = 0-2), and NWs often resulted in co-sleeping. The duration of NWs during baseline was highly variable (range = 30-270 min). Five nights into treatment, NWs were eliminated, with the exception of two nights (one of long latency) and remained eliminated during STFU. The PBM scores for frequency (83%) and duration (89%) of night wakes showed a moderate treatment effect, with a large treatment effect at STFU (PBM frequency and duration = 100%). In addition, co-sleeping was eliminated upon the introduction of intervention and remained eliminated throughout treatment and follow-up.

Figure 4.4.



Sleep Outcomes for Mirasol: Duration of Sleep Onset Latency, Frequency and Duration of Night Wakings across Baseline, Intervention, and Short-Term Follow-up Phases. The numbers on the graph represent the value of data points that are out of range. Intervention = sleep hygiene (including calming pre-sleep activities), environmental modification, sleep restriction, visual sign, no access to food and toys, planned ignoring, social story, positive reinforcement

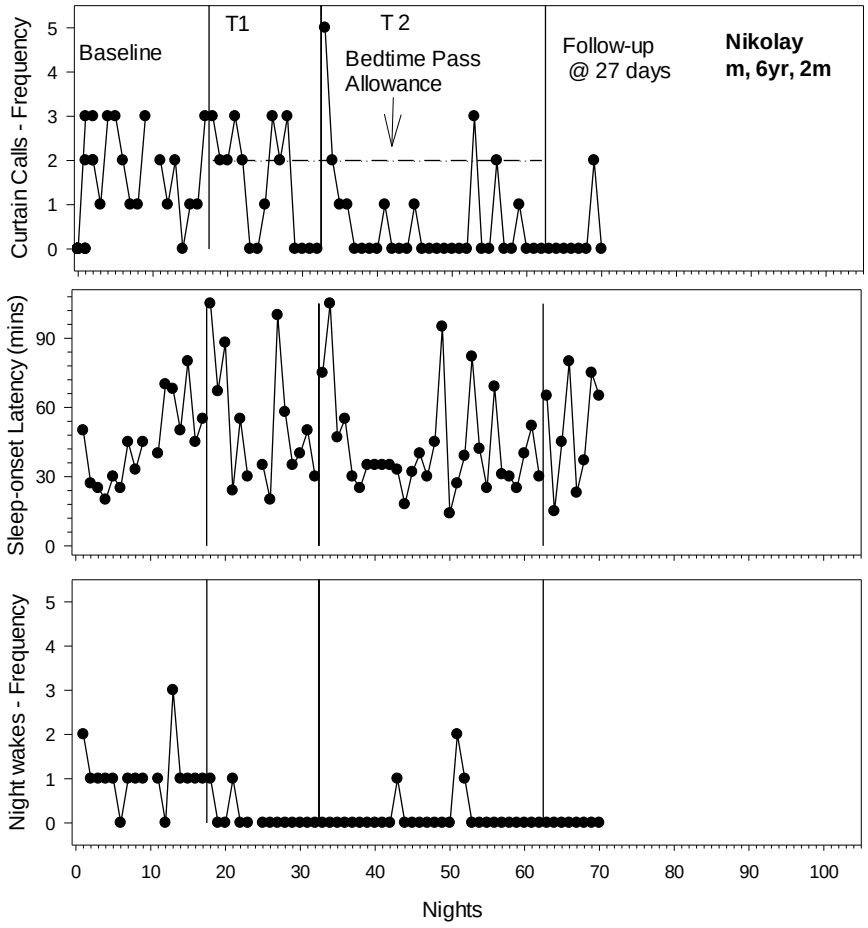
Nikolay

The dependent variables for Nikolay were the frequency of CCs, the duration of SOL and the frequency of NWs, and are presented in Figure 4.5. The line on the curtain call graph shows the cut-off for sanctioned (two with the bedtime pass) versus unsanctioned curtain calls. During baseline, Nikolay slept in his parent's bedroom, however under intervention he slept independently in his own bedroom. SOL trended upward in baseline; during treatment phase one (T1) and phase two (T2) there was high variability, although the level of SOL reduced in T2 compared to baseline with a moderate treatment effect (PBM = 70%). This treatment effect was no longer evident at STFU (PBM = 38%).

During his settling period Nikolay engaged in frequent CCs (range = 1-3) during baseline. These decreased in T1 and were eliminated in T2 (PBM = 97%) and at STFU (PBM = 100%). Figure 4.5 also shows use of the bedtime pass in relation to CC data; the horizontal line indicates the allowance of call outs as per the procedure (i.e., 2 passes = 2 sanctioned call outs); any CCs above this line were unsanctioned. Results show that across intervention when Nikolay began sleeping in his own bed, frequency of CCs and usage of bedtime passes often converged on zero.

During baseline, Nikolay woke once per night on average (range = 0-3). His NWs ceased almost immediately when treatment began and continued to be infrequent throughout intervention (PBM = 90%) and were eliminated during STFU (PBM = 100%).

Figure 4.5.



Sleep Outcomes for Nikolay: Frequency of Curtain Calls, Duration of Sleep Onset Latency and Frequency of Night Wakings across Baseline, Intervention, and Short-Term Follow-up Phases. T1 = sleep hygiene (including consistent sleep location), sleep item, sleep restriction, visual schedule, Gro-clock™, bedtime pass and planned ignoring, positive reinforcement; T2 = reduced parental interaction

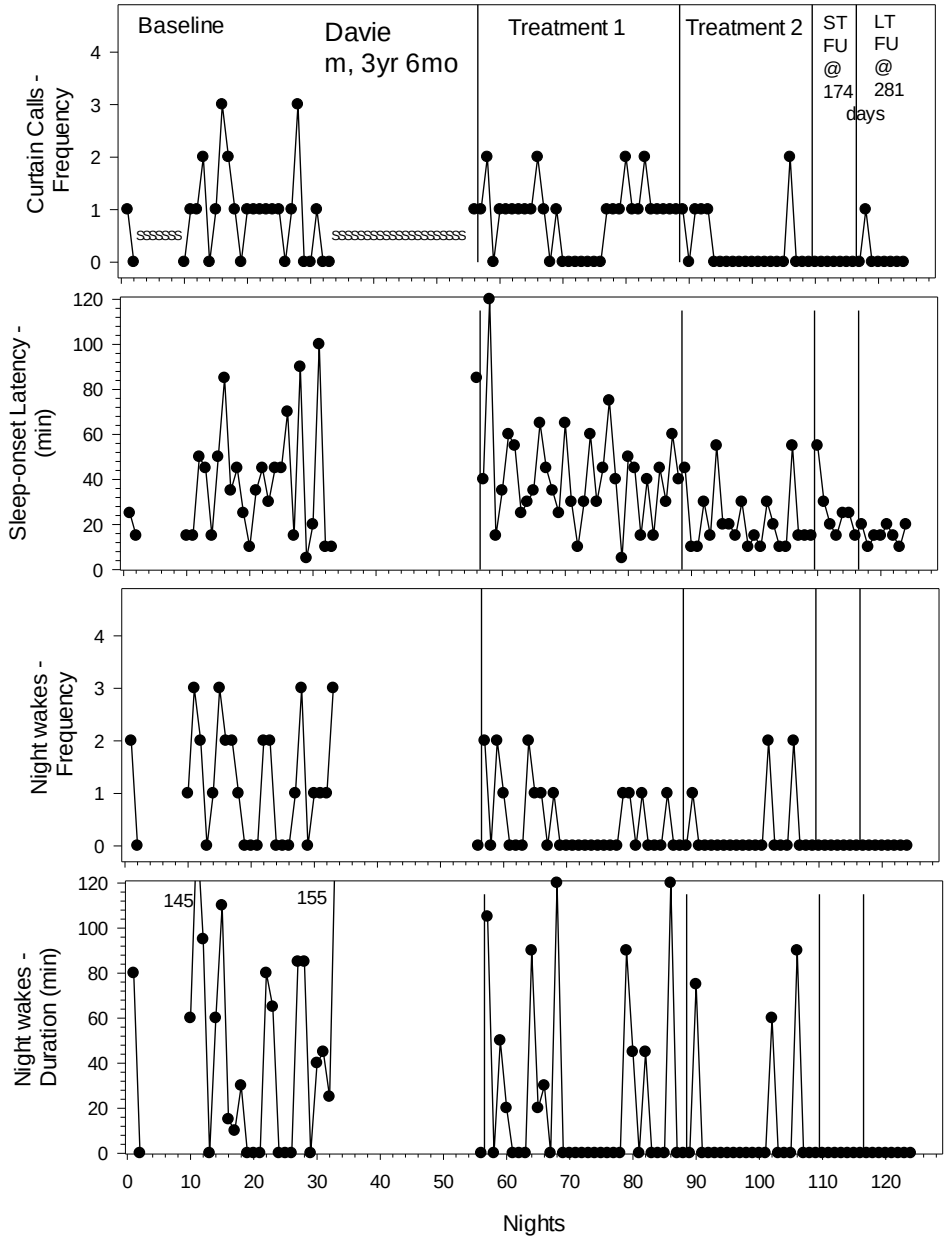
Davie

The dependent variables for Davie were the frequency of CCs, duration of SOL, and the frequency and duration of NWs (see Figure 4.6). The 'S' represents nights that Davie was sick. The numbers on the graph represent the value of data points that are out of range. Davie engaged in frequent CCs during baseline (range = 0-3). No treatment effect is evident during treatment phase one (T1). During treatment phase two (T2), however, CCs were near eliminated, with the PBM score demonstrating a moderate treatment effect (81%) and

remained eliminated except for one night during follow-up (STFU PBM = 100%, LTFU PBM = 86%). Unfortunately, a period of sickness precluded data collection immediately prior to the start of treatment. Davie's SOL during baseline was highly variable, ranging from five to 100 min. There is no evidence of a treatment effect for SOL during T1, however, during T2 the level and variability of SOL were reduced, with a moderate effect size (86%). Further improvement was evident during follow-up, with all nights' SOL $\leq$ 30 min (PBM = 100%).

During baseline, Davie woke up to three times per night. The duration of his NWs was highly variable, ranging from 10 to 155 min. During T1, no treatment effect was obtained for the frequency of NWs, however, the duration of NWs was reduced with a moderate sized treatment effect. In T2, NWs became less frequent, with longer periods compared to baseline where no NWs occurred. The PBM scores for frequency and duration of NWs in T2 showed a moderate treatment effect (86%). During follow-up, NWs were eliminated (PBM = 100%).

Figure 4.6.



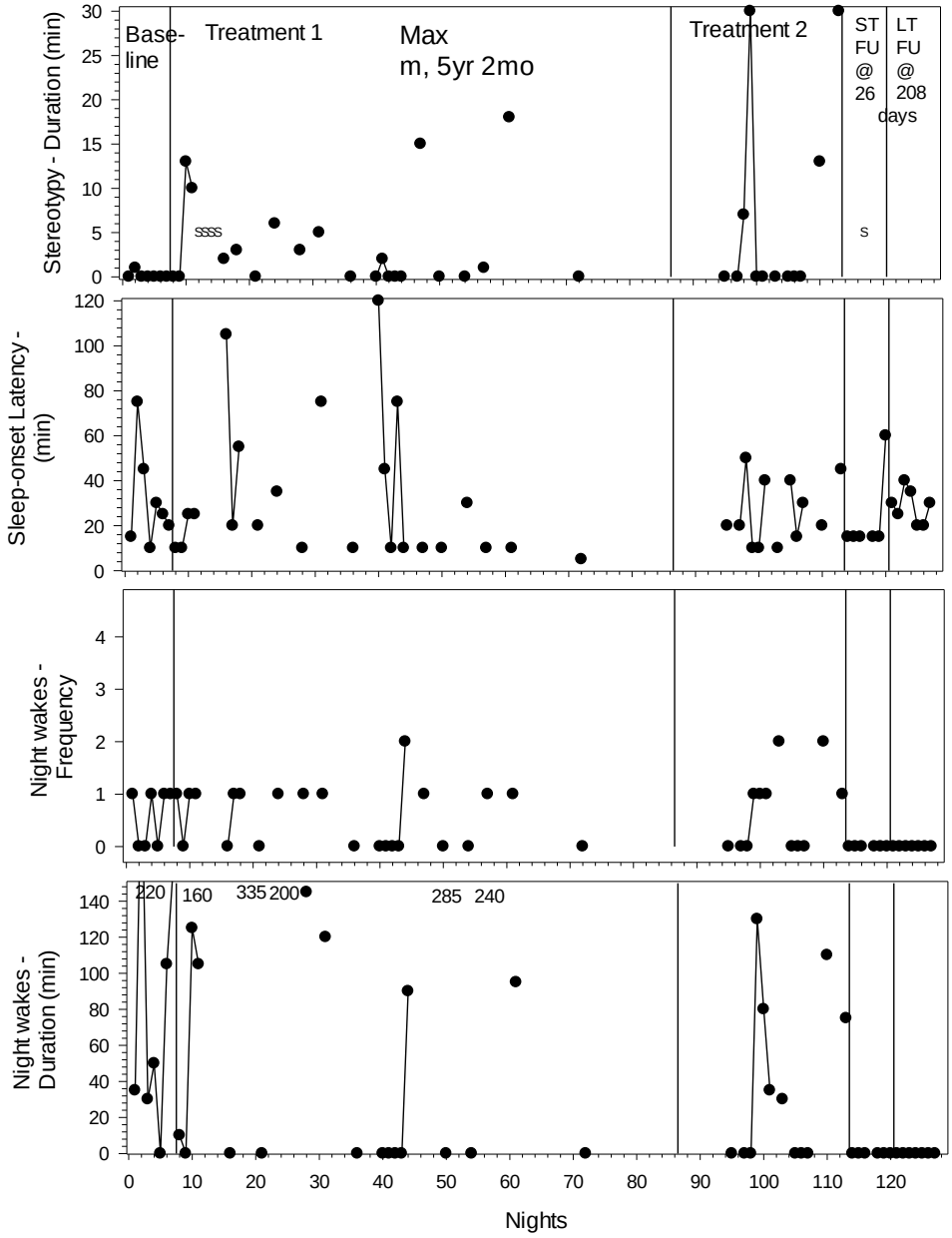
Sleep Outcomes for Davie: Frequency of Curtain Calls, Duration of Sleep Onset Latency and the Frequency and Duration of Night Wakings across Baseline, Intervention, and Follow-up Phases. The 'S' represents nights that Davie was sick. The numbers on the graph represent the value of data points that are out of range. Treatment 1 = white noise; Treatment 2 = (new bedroom) faded bedtime, pre-bedtime satiation (food, drink, attention), positive reinforcement

Max

The dependent variables for Max were the duration of SOL, the frequency and duration of NWs, and the duration of stereotypy (see Figure 4.7). The 'S' shows the nights that Max was sick. The numbers on the graph represent the value of data points that are out of range. Stereotypy was almost non-evident during baseline, occurring once for a matter of minutes. PBM could not be calculated for stereotypy because of its low occurrence during baseline, but it is evident that stereotypy worsened (duration range of 2-30 min) during intervention compared to the baseline phase. During baseline, SOL ranged from 15-75 min. Treatment did not effectively reduce SOL during treatment phase one nor two (PBM = 58%), however a reduction in SOL was evident during STFU, with a moderate treatment effect (PBM = 83%). This effect was not maintained at LTFU (PBM = 29%).

The frequency of NWs during baseline was 0-1, with the duration of NWs ranging from 30-220 min. There is no evidence of a treatment effect on NWs during intervention (frequency PBM = 50%, duration PBM = 67%). A significant improvement is evident at follow-up, during which no NWs were reported (PBM = 100%), however, it should be noted that follow-up was assessed through sleep diary data rather than VSG, and it is possible that NWs occurred that Max's parent was unaware of (i.e., Max did not signal that he was awake). It is also noteworthy that Max's parent reported that he suffered anxiety relating to attending school; Max's parent reported that although the set wake time improved his morning routine, his anxiety increased relating to increased hours at school (i.e., from arriving on time). Max's parent believed Max's worries might have interfered with his sleep onset. Max was receiving treatment for anxiety from an external service provider.

Figure 4.7.



Sleep Outcomes for Max: Duration of Stereotypy, Duration of Sleep Onset Latency and the Frequency and Duration of Night Wakings across Baseline, Intervention, and Follow-up Phases. The 'S' shows the nights that Max was sick. The numbers on the graph represent the value of data points that are out of range. Treatment 1 = TV replaced with night light, pre-bedtime satiation (TV), sleep restriction, Gro-clock™, social story, positive reinforcement; Treatment 2 = sleep restriction modification

Table 4.3. *Percentage below the Median Scores*

	DV	Treatment PBM		STFU PBM	LTFU PBM
Emma	SOD	86%		43%	43%
	Stereotypy	83%		100%	100%
Jorge	Freq NWs	100%		100%	100%
	Dur NWs	100%		100%	100%
	BF	100%		100%	100%
Mirasol	SOD	71%		0%	-
	Freq NWs	83%		100%	-
	Dur NWs	89%		100%	-
Nikolay	CCs	97%		100%	-
	SOD	70%		38%	-
	Freq NWs	90%		100%	-
Davie		T1	T2		
	SOD	38%	86%	100%	100%
	CCs	28%	81%	100%	86%
	Freq NWs	69%	86%	100%	100%
	Dur NWs	78%	86%	100%	100%
Max	SOL	58%		83%	29%
	Freq NWs	50%		100%	100%
	Dur NWs	67%		100%	100%

High effectiveness (>90%)	Moderate effectiveness (70-90%)	Ineffective (<70%)

Note. BF: (night-time) breastfeeding; CCs: curtain calls; Dur: duration; DV: dependent variable; Freq: frequency; LTFU: long-term follow-up; NWs: night-wakings; PBM: percentage below the median; SOD: sleep onset delay; STFU: short-term follow-up; T1: treatment phase 1; T2: treatment phase 2

Children's Sleep Habits Questionnaire Scores

The CSHQ and SF-CSHQ total pre- and post-treatment scores for all children are presented in Table 4.5. Davie and Max's CSHQ scores reduced post-treatment; however, their scores remained above the clinical cut-off (41). Emma, Jorge, and Mirasol's scores also reduced post-treatment on the SF-CSHQ. For Jorge and Mirasol, these changes were clinically significant, falling below the clinical cut-off (30). Emma's pre- and post-treatment

scores were below this cut-off. Insufficient information was available to calculate Nikolay's scores.

Table 4.4. *Children's Sleep Habits Questionnaire Scores*

	Emma	Jorge	Mirasol	Nikolay	Davie	Max
Pre-treatment	13(SF)*	36(SF)	40(SF)	-	53	50
Post-treatment	7(SF)*	23(SF)*	17(SF)*	-	45	47

Note. SF: short-form CSHQ; *Below clinical cut-off

Interobserver Agreement

For Jorge, Mirasol, and Nikolay, IOA was calculated for 26%, 34% and 42% of sessions across phases, and was 88%, 92% and 89%, respectively. For Davie, IOA was calculated for 57% of STFU only, and was 87%. For Max, IOA was calculated for 8% of the treatment phase only, and was 82%. IOA was not able to be calculated for Emma.

Treatment Fidelity

Treatment fidelity scores are presented in Table 4.4. Treatment fidelity during the intervention phase was calculated for 30% of sessions for Emma, Jorge, Davie, and Max, and 35% of sessions for Nikolay; scores ranged from 78-100%. There was insufficient information to calculate treatment fidelity for Mirasol. Treatment fidelity during STFU was calculated for 43%, 33%, 57% and 43% of sessions for Jorge, Nikolay, Davie and Max, respectively; scores ranged from 56-100%. Treatment fidelity during LTFU was calculated for 60%, 38%, 57% and 43% of sessions for Jorge, Nikolay, Davie and Max, respectively; scores ranged from 0-100%. Insufficient information prevented treatment fidelity from being calculated during follow-up for Emma and Mirasol.

The overall treatment fidelity scores ranged from 77-100% across children indicating that in general, parents implemented intervention with moderate-high fidelity, particularly during the intervention phase. In general, fidelity scores tended to decrease during follow-up compared to intervention.

Table 4.5. *Treatment Fidelity Scores across Phases*

	Intervention	STFU	LTFU	Overall score
Emma	100%	-	-	100%
Jorge	91%	56%	20%	78%
Mirasol	-	-	-	-
Nikolay	78%	78%	67%	77%
Davie	100%	75%	0%	100%
Max	79%	100%	100%	82%

Note. LTFU: long-term follow-up; STFU: short-term follow-up.

Social Validity

Post-Treatment Interview

Overall, parents felt satisfied with the treatment process and with the outcomes regarding their own child's sleep patterns. All parents perceived the intervention to be beneficial, with some reporting that it exceeded their expectations. Specifically, parents felt they learnt valuable strategies for managing children's sleep-related behaviour, and/or that the child learnt to change their own behaviour (e.g., to sleep independently, lie quietly in bed) and were pleased their child received the recommended amount of sleep for their age (Ohayon et al., 2017). Parents also described satisfaction relating to the level of collaboration (i.e., inclusion of parent input throughout the assessment and treatment process) and that the programme was adapted to the child and family's individual needs (e.g., *"a good family fit"*).

All parents felt the interventions were effective. Interestingly, Mirasol's mother perceived white noise to be the most effective treatment component, and reported it was well liked by Mirasol and herself. Similarly, Davie's parents reported Davie enjoyed the white noise and would request it prior to bed by making static sounds. Disadvantages of treatment noted by parents included the length of time needed to carry out treatment and recording data daily. One downside for Jorge's parents was their conflicting emotions about whether ceasing night-time breastfeeding would affect Jorge's nutritional needs, which they were seeking advice from a nutritionist about. Outside of sleep, perceived positive effects included improved routines (e.g., morning routine before school), tidiness of the child's bedroom (i.e., items previously used during sleep-interfering behaviours were removed), improved daytime behaviour (e.g., fewer challenging behaviours such as tantrums) and improvements in parents' own sleep quality, maternal mood levels (e.g., *"feeling calmer")* and less strain in the marital relationship.

The Treatment Acceptability Rating Form - Revised

TARF-R scores are presented in Table 4.6. The total acceptability score has a possible range of 17-119; higher scores indicate higher acceptability. Overall, parent ratings indicated a high level of treatment acceptability (range = 87-119). Parents viewed treatment as effective and reasonable, and indicated high willingness to implement treatment. Jorge's parents were less satisfied overall with treatment proceedings, particularly owing to perceived negative side effects and disruption, as did one of Nikolay's parents. Parents generally viewed the treatments as easy to understand and having no financial cost.

Table 4.6. *Treatment Acceptability Rating Form-Revised Scores*

	Emma		Jorge		Mirasol	Nikolay		Davie		Max	Maximum score
Scale	P1	P2	P1	P2	P1	P1	P2	P1	P2	P1	
Effectiveness	20	18	20	18	18	21	21	19	-	21	21
Reasonableness	18	20	17	20	18	21	18	20	-	21	21
Willingness	19	20	20	15	21	20	15	21		21	21
Cost	7	7	14	14	14	14	14	14	-	14	14
Negative side-effects	18	17	8	16	21	20	19	9	-	21	21
Disruption/time	17	14	13	11	21	17	20	10	-	21	21
Problem severity*	-	7	10	8	14	14	12	12	-	14	14
Understanding of treatment*	7	7	6	6	6	7	4	6	-	7	7
Total acceptability	**106**	**103**	**87**	**94**	**113**	**113**	**107**	**111**	**-**	**119**	**119**

Note. P1: parent one; P2: parent two; *Not included in total acceptability score

Discussion

The purpose of this study was to evaluate the effectiveness of function-based BSI for children on the autism spectrum, the short- and long-term maintenance of treatment effects, and the social validity of treatment. Stereotypy was observed in 4/6 children which permitted a preliminary examination of the feasibility of addressing stereotypy using function-based interventions where it appeared to interact with sleep problems. All children in this study experienced multiple sleep-related problem behaviours in line with research showing that sleep problems frequently co-exist in children on the autism spectrum (Cotton & Richdale, 2006; Liu et al., 2006; Roussis et al., 2021). In addition, FBA revealed the sleep problems were multiply determined, indicating the need for multimodal and individualised treatment (Didden et al., 2002; Goldman et al., 2012; McLay, France, Blampied, van Deurs, et al.,

2021; Spruyt & Curfs, 2015). Even when two children experienced the same type of sleep problem, the underlying factors contributing to the sleep problems differed across individuals. For example, FBA indicated that for Emma SOD was maintained via engagement in automatically reinforced stereotypy, whilst for Nikolay, SOD was maintained by access to toys and parent interaction in response to his avoidant bedtime behaviour. Further, the same functions maintained different sleep problems across children; for example, parent attention was a common reinforcing consequence maintaining bedtime resistance, CCs, SOD and NWs across and within children. FBA highlighted not only what types of intervention were likely to be effective and why but allowed for a personalised approach that comprehensively addressed multiple factors unique to each child.

All treatments in this study were multimodal involving antecedent and consequence-based strategies. Stimulus control for sleep was improved via modifications to children's sleep environment and/or instructional context, including improved sleep hygiene practices (e.g., a clear, calm, consistent bedtime routine), sleep/wake scheduling, the establishment of sleep-conducive S^D (e.g., a sleep item) and the removal of inappropriate S^D (e.g., bedroom TV). Visual aids (e.g., a Gro-clock™, social stories) were used to provide S^D for sleep and to instruct children on desirable sleep-related behaviour (e.g., lying quietly in bed).

Stimulus control was also improved via manipulation of the MOs for sleep, to ensure children's physiological state at bedtime was sleep-conducive. Specifically, sleep restriction in the form of the faded bedtime procedure and set wake time was used to increase the value of sleep (via mild sleep deprivation, i.e., an EO) and accordingly increase children's motivation to fall asleep and stay asleep. For three children (Emma, Davie, and Max), pre-bedtime access to putative reinforcement for sleep-interfering behaviour was used to satiate the child (i.e., an AO) and decrease competition from other motivational states after they were bid goodnight. This procedure was coupled with extinction, wherein access to the same

putative reinforcement after being bid goodnight was removed. Although its effects on sleep-interfering behaviour are unclear (e.g., whether the child was satiated), this component highlights the types of strategies that may be indicated for use based on the outcomes of FBA. Satiation was not identified by the review in Chapter 2 of behavioural sleep treatments but may be an important factor when broadly considering the motivational variables that affect children's sleep, as discussed in Chapter 1. The use of satiation as a procedure to reduce sleep-interfering stereotypy is discussed further in Chapter 5.

Finally, in this study, modified and unmodified extinction procedures were used to eliminate parent attention maintaining problem behaviour (e.g., CCs). Despite the concerns outlined in Chapter 2 regarding the undesirable effects of extinction (e.g., an extinction burst), it is noteworthy that one parent preferred an unmodified extinction procedure which was implemented with high fidelity, emphasising the importance of parent choice within a behavioural sleep programme, and supporting the findings of Weiskop et al. (2005).

Results showed that the function-based interventions effectively reduced SOD in 4/5 children (for Jorge SOD was not a problem) with a moderate treatment effect. CCs reduced with a moderate-large treatment effect for 2/3 children. Of note, a reduction in Nikolay's CCs did not reduce SOL. This suggests that Nikolay left his bedroom or demanded parent attention less often compared to baseline, but that he remained awake (e.g., lying quietly in bed) during this time. Lengthy and/or frequent NWs were problematic for all children except Emma. Following intervention, NWs reduced in frequency and duration in 4/5 children, with a moderate-large treatment effect.

Additional dependent variables were co-sleeping (Mirasol and Nikolay), night-time breastfeeding (Jorge) and stereotypy (Emma and Max). Following intervention, all children were sleeping independently and in their own bed. Jorge's parents were able to eliminate night-time breastfeeding during intervention. The return to breastfeeding during follow-up

likely reflects the indecision parents reported throughout the study regarding Jorge's nutritional needs, for which they received advice for from a professional outside of the study. Despite the return of breastfeeding, it remained at a significantly reduced frequency compared to baseline.

Interestingly, following intervention, stereotypy reduced for one child (Emma) with a moderate sized treatment effect, but worsened for another (Max). Emma's sleep intervention appeared to successfully reduce her SOL and engagement in RMO, however, she continued to engage in motor stereotypy (e.g., body rocking) during SOL. This finding raises a critical challenge regarding the treatment of stereotypy in the sleep context, specifically, how to disrupt the contingency between sleep-interfering behaviour and its reinforcer, when reinforcement is (presumably) automatic and sensory-based, and not amenable to direct, external manipulation. It is also unclear whether a reduction in one form of stereotypy (e.g., RMO) may have increased engagement in another (e.g., body rocking). Further, for Max, stereotypy intensified when other sleep problems were targeted during function-based treatment. It is possible that the absence of audio and visual stimulation (previously obtained from the TV) increased the salience and value of reinforcement from stereotypy, resulting in increased engagement in stereotypic behaviour. These findings strongly suggest there are associated complexities that require further investigation to enhance our understanding of stereotypy within the sleep context.

Vocal stereotypy was problematic for two children (Mirasol and Davie); however, it was not possible to determine the impact of treatment on these behaviours owing to the inability of VSG to capture audio. It is interesting to note that for Davie, vocal stereotypy was most problematic during NWs, which reduced in duration with a moderate sized effect upon the implementation of white noise. In comparison, white noise had no effect on his SOL nor CCs. It is possible, but impossible to determine, that white noise reduced his vocalisations

enabling him to resettle to sleep more efficiently. However, Davie's parents also reported that they closed their own bedroom door to reduce their exposure to continuous sound which they disliked, which resulted in them feeling less disturbed by Davie's NWs during intervention. It is possible therefore, that Davie's vocalisations did not reduce but rather that parents became less aware of his stereotypic behaviour during NWs. This is an important line for further research to investigate the impact of white noise on vocal stereotypy (e.g., whether it effectively reduces such behaviour), as well as the relationship between stereotypy and sleep problems, including whether a reduction in stereotypy can reduce the duration of NWs.

Overall, reported social validity was high, indicating that parents were generally satisfied with the treatment procedures and outcomes. These ratings occurred despite not all sleep goals being ideally met; for example, Emma's SOD did not reduce below a 60-90 min timeframe, but her parents were highly satisfied with this outcome. Some families specifically noted during the post-treatment discussion that they were particularly pleased with the collaborative nature of the assessment and treatment process; this is in line with research emphasising the value of shared decision making on treatment outcomes (Jin et al., 2013; K. Turner & Johnson, 2013; Sanders & Burke, 2014; Weiskop et al., 2005). A strong advantage of FBA is the ability to tailor treatment to be a good fit for each family; this is an important consideration, as the families in this study diverged in terms of how autism affected their child, parental goals and motivations, and family context (Hastings, 2002; Waddington et al., 2020). Parents were an active and continual part of the research process, which ensured that family goals, preferences, strengths (including parent expertise) and unique needs were incorporated into intervention (Jin et al., 2013; Moes & Frea, 2002; Sanders & Burke, 2014).

In addition, treatment fidelity was moderate-high demonstrating that parents were generally willing and able to adhere to the behavioural programme. In general, however,

fidelity was found to decrease during follow-up in comparison to intervention. The reasons for this are unclear, but the cessation of on-going contact with the research team is one possibility (e.g., there was decreased accountability to maintain the behavioural strategies). Of note, treatment effects were inconsistently maintained when assessed during follow-up, which is possibly a result of decreased fidelity over this time.

Limitations and Directions for Further Research

A limitation of this study is that owing to the quasi-experimental (AB) design, confident inferences cannot be made about the treatment causing the changes observed. Further, the small number of participants also limits the extent of replication and the generalisability of results. Another key limitation of this study was the difficulty some parents had collecting sleep diary data, resulting in missing or incomplete data sets. For example, Jorge's CC results were limited by the absence of baseline data, which occurred despite close contact between the researcher and Jorge's family. In the case of Emma and Max, parents were limited in their ability to record sleep diary data because they were unaware of behaviour that occurred whilst the child was not visible or audible. This emphasised the importance of methods of data collection involving instrumentally recorded observations (e.g., VSG) to triangulate data and circumvent bias, and to help quantify treatment effects (Blampied, 2013b; Blampied & Bootzin, 2013; Knight & Johnson, 2014; Pattison et al., 2020; Spruyt & Curfs, 2015).

On the other hand, heavy reliance on VSG (Emma and Max) was also a limitation, because on nights when video data was not recorded (for reasons unknown), or the child moved out of the range of the camera, the sleep variables (e.g., duration of sleep-interfering behaviour) remained unknown. The fact that data can be lost for a variety of reasons underscores the need for multiple sleep-related measures to allow for triangulation of findings. Furthermore, this limitation emphasises the importance of close contact with

families to help circumvent problems with data recording, preferably before they arise, which may include remote support (Pattison et al., 2020).

Finally, the results of this study were limited by an inability to comment on the effectiveness of individual modes of treatment. FBA directed the need for multimodal treatment, to effectively address the multiple determinants of sleep problems for each child (Kodak & Piazza, 2008; McLay, France, Blampied, van Deurs, et al., 2021). However, the combination of multiple treatment components limited the ability to understand how each component contributed to the overall outcomes, or whether effects were due to the accumulation of treatment effects over the treatment sequence (Cuomo et al., 2017; Knight & Johnson, 2014; Pattison et al., 2020; Vriend et al., 2011). One way to investigate this even when the need for multiple treatments is indicated may be to stagger the introduction of treatment components so that each component may be individually assessed, as was done for Davie with white noise. This specific line of inquiry is investigated in Chapter 7. The finding that white noise alone significantly reduced the duration of NWs for Davie highlights the need for further investigation into the use of white noise on sleep outcomes; particularly as the mechanism underlying this change remains unclear.

A central finding to have emerged from this study was that stereotypy, as a core characteristic of autism, was a feature of sleep disturbance in four (Emma, Mirasol, Davie, and Max) of the six participant children. This emphasises how the outcomes of FBA may highlight autism-related factors contributing to sleep disturbance (Mazzone et al., 2018; Phillips et al., 2020; Reynolds & Malow, 2011), and raises important questions regarding how to treat sleep-related stereotypy. For example, for Max, stereotypy unexpectedly worsened in the context of the BSI. It is generally unclear whether established behavioural interventions strategies can effectively reduce sleep-related stereotypy, or the types of modifications or additional strategies that are required to address autism-associated

behaviours (Pattison et al., 2020; Phillips et al., 2020), and the efficacy of such strategies. The evidence-based interventions available to treat stereotypy, and the potential application of such strategies to the treatment of sleep problems, are investigated in the next two chapters.

Chapter 5

The Assessment and Treatment of Sleep-Related Stereotypy[6]

The outcomes of Study 1 underscore the need to examine sleep-related stereotypy in children on the autism spectrum in greater depth, including investigating the ways in which stereotypy can be treated when it occurs in the context of sleep problems. This chapter begins with a focus on the definition of stereotypy and its underlying functions, as well as discussing the types, prevalence, aetiology, and development of stereotypy in children on the autism spectrum. Consideration is then given to the conditions under which stereotypy is problematic, and briefly to sleep-related movement disorders in autism. Following this, the relationship between stereotypy and sleep problems is explored, with a focus on the specific ways in which stereotypy may interfere with sleep in children on the autism spectrum. This is followed by an overview of how FBA may be used to assess and then inform treatments for sleep-related stereotypy. Finally, a summary of available treatments for stereotypic behaviour and the potential application of these treatments to sleep problems in children on the autism spectrum is discussed. This chapter highlights the complexities associated with the treatment of sleep-related stereotypy and will provide an overview of the extant research.

Definition of Stereotypy

There is no clear consensus on how to define stereotypy, and variations exist in research and clinical practice (Bodfish et al., 2000; Melo et al., 2020; Rapp & Vollmer, 2005a; Singer, 2009). Stereotypy is typically determined on the basis of observation in that behaviour that appears repetitive and fixed may be defined as stereotypic (Cunningham &

[6] Sections of this chapter are published with permission from Springer Nature from a book chapter with myself as the first author: Hunter, J. E., McLay, L. K., France, K. G., & Blampied, N. M. (forthcoming). The assessment and treatment of stereotypy in the sleep context. In L. K. McLay, K. G. France, & N. M. Blampied (Eds.), *Clinical handbook of behavioral sleep treatment for children on the autism spectrum.* Christchurch, NZ: Springer International

Schreibman, 2008; M. Turner, 1999). Aside from sharing a response topography of repetition and invariance, stereotypic behaviours in children on the autism spectrum are heterogeneous, varying not only in form but also in duration, frequency and intensity across settings and time (Cunningham & Schreibman, 2008; Rapp & Vollmer, 2005a). Stereotypy is therefore an umbrella term, which encompasses a wide variety of repetitive and fixed behaviours in children on the autism spectrum (Cunnigham & Schreibman, 2008; M. Turner, 1999).

It is widely accepted that stereotypies are operant behaviours (Cunningham & Schreibman, 2008; DiGennaro Reed et al., 2012; Rapp & Vollmer, 2005a) that are often maintained, at least in part, by the automatic sensory consequences produced by the behaviour itself (Ahearn et al., 2007; Lanovaz & Sladeczek, 2012; Rapp & Vollmer, 2005a). Automatic reinforcement is either positive (i.e., produces a sensory consequence) or negative (i.e., reduces or eliminates a sensory experience, e.g., scratching to attenuate an itch). Stereotypic behaviours are commonly deemed *self-stimulatory* as the behaviour occurs in the absence of obvious external events (e.g., social consequences) that might constitute reinforcement and regardless of an individual's environment (DiGennaro Reed et al., 2012; Vollmer, 1994). For example, a child may make repetitive vocalisations that serve no apparent social function, but which are presumed to have reinforcing sensory consequences (e.g., auditory stimulation) that are intrinsic to the individual (Lanovaz & Sladeczek, 2012).

Sensory consequences may be auditory, tactile, visual, olfactory, proprioceptive (arising from innervation of muscles and joints) and interoceptive (arising from internal organs [e.g., vestibular, or gustatory sensations]), or can occur in combination (Cunningham & Schreibman, 2008). Further, specific aspects of sensory stimulation may be reinforcing; for example, oral stimulation of mouthed items may be reinforcing, but it may be the taste or the texture of the mouthed item that is specifically reinforcing (Piazza et al., 1998).

Historically, the terms *self-stimulatory* and *stereotypy* have been used interchangeably in both research and clinical practice, as all stereotypic behaviour was presumed to fall under the response class of automatic reinforcement (Ahearn et al., 2007; Cunningham & Schreibman, 2008; Rapp & Vollmer, 2005a). The notion that *stereotypy* should refer only to automatically reinforced behaviour, is still advocated for in the research literature (e.g., Cook et al., 2018; Lanovaz & Sladeczek, 2012; Rapp & Lanovaz, 2016; Rapp & Vollmer, 2005a). However, while automatic reinforcement does appear to be the most frequently maintaining function (DiGennaro Reed et al., 2012), there is also evidence to show that stereotypy can be maintained by socially mediated contingencies of positive (e.g., attention) and negative (e.g., escape from demands) reinforcement (Cunningham & Schreibman, 2008; Durand & Carr, 1987; Kennedy et al., 2000; Roantree & Kennedy, 2006). For example, Wilke et al. (2012) assessed the function of stereotypy in 53 children and adolescents on the autism spectrum and found that aside from automatic reinforcement, stereotypy was maintained by social consequences (e.g., attention) in 10% of individuals.

In another study by Scalzo et al. (2015), vocal stereotypy was hypothesised to be maintained by multiple functions in a 12-year-old male on the autism spectrum, including social consequences (e.g., attention, access to tangible items) in addition to an automatic function. In summary, stereotypy has at least one of four maintaining functions: positive or negative social reinforcement (e.g., attention or escape), or positive or negative non-social reinforcement (i.e., automatic reinforcement from experience of or escape from sensory conditions; Cunningham & Schreibman, 2008; DiGennaro Reed et al., 2012). Stereotypy may also be multiply determined across individuals (Kennedy et al., 2000; S. Patterson et al., 2010) since there is no reason to expect a single contingency to maintain behaviour.

Types of Stereotypy

There is evidence to suggest that the broad category of stereotypy, including repetitive motor movements, vocalisations, RMO, insistence on sameness, and circumscribed interests (APA, 2013), may consist of at least two distinct subcategories of behaviour (Lam et al., 2008; Hundley et al., 2016; MacDuffie et al., 2020). These include Repetitive Sensory Motor (RSM) behaviours, sometimes called 'lower-order' behaviours, and Insistence on Sameness (IS), sometimes called 'higher-order' behaviours (Cunnigham & Schreibman, 2008; Hundley et al., 2016; Lydon et al., 2017; Singer, 2009). The RSM subcategory is thought to comprise repetitive motor movements, vocalisations and RMO, whilst the IS subcategory includes insistence on sameness, restricted interests, resistance to change, and cognitive behavioural rituals and compulsions (Bishop et al., 2013; Cunningham & Schreibman, 2008; DiGennaro Reed et al., 2012; Hundley et al., 2016; MacDuffie et al., 2020).

These subcategories may also have distinct genetic or neurobiological aetiologies (Langen et al., 2014; Lam et al., 2008; Wolff et al., 2013). RSM behaviours are shown to be associated with younger age and lower intellectual ability (Goldman, Wang, et al., 2009; Hundley et al., 2016; Hollway & Aman, 2011; Szatmari et al., 2006), while IS behaviours are often associated with anxiety (Hundley et al., 2016; Lydon et al., 2017; M. Turner, 1999). Further research is needed to improve our understanding of subtypes of stereotypy (Hundley et al., 2016), including whether such subcategories actually exist (Bodfish et al., 2000; Langen et al., 2011; M. Turner, 1999), and the number of distinct subtypes (Lam et al., 2008).

All participant children within this thesis happened to engage in repetitive motor movements, vocalisations and/or RMO, which fall under the RSM subcategory of behaviour. Within this subcategory, stereotypic behaviour is often broadly distinguished as either motor or vocal behaviour (Cook et al., 2018; D. Wang et al., 2020); each with its own distinct body of treatment literature (discussed under 'Treatment' below; Akers et al., 2020; D. Wang et al.,

2020). Motor stereotypy has been further characterised in the research literature according to body part (e.g., Cook et al., 2018; Goldman, Wang, et al., 2009). For example, Goldman, Wang, et al. (2009) counted and characterised the types of stereotypies occurring in 129 children on the autism spectrum compared to 148 cognitively matched developmentally delayed children (not on the autism spectrum) in terms of body parts used to perform the behaviour. Children were observed during 15 min standardised play sessions, with the observers blinded to children's diagnosis. The forms of motor stereotypy observed included the: (a) face (e.g., grimacing, lips, tongue movements); (b) head, trunk and shoulders (e.g., head shaking, body rocking, shrugging the shoulders); (c) arm/leg (e.g., flapping, stamping feet); (d) hand/finger (e.g., shaking, tapping, twirling an object); (e) gait (e.g., pacing, jumping, spinning); (f) self-directed movements (e.g., mouthing, slapping self, object or surface); and (g) visual fixation (e.g., staring at an object or fingers out of the corner of the eyes; Goldman, Wang, et al., 2009).

Results showed that autism and low nonverbal IQ (i.e., NVIQ < 80) independently contributed to the frequency of stereotypy, with stereotypy occurring more frequently in the autism group (irrespective of NVIQ) than the DD group, and more frequently for those with low NVIQ (with or without autism) than for those with higher NVIQ (Goldman, Wang, et al., 2009). Additive effects were found regarding the frequency and variety of stereotypies for children on the autism spectrum who also had low NVIQ (Goldman, Wang, et al., 2009). Gait and hand/finger stereotypies in particular were strongly associated with autism (Goldman, Wang, et al., 2009).

Prevalence, Aetiology and Development of Stereotypy

Most individuals on the autism spectrum engage in at least one form of stereotypy (APA, 2013; Chebli et al., 2016). Melo et al. (2020) reviewed the prevalence of motor stereotypies in 37 studies of individuals on the autism spectrum and concluded a median

prevalence of 52% (range = 22-98%). A review by Chebli et al. (2016) of the prevalence of stereotypy (inclusive of motor and vocal behaviours) found a median prevalence of 88% in individuals on the autism spectrum, in comparison to 61% of individuals with other types of DD. The prevalence of vocal stereotypy is relatively unclear and likely to be underestimated owing to a lack of items on common measures of stereotypy specific to vocal stereotypy (Chebli et al., 2016). For example, the Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al., 2000) which is a frequently used measure of stereotypy does not include any items pertaining to vocal stereotypy (Chebli et al., 2016). However, a survey of parents of children on the autism spectrum suggests that up to 85% of these children and young people engage in vocal stereotypy (Mayes & Calhoun, 2011).

The aetiology of stereotypy in children on the autism spectrum, as well as how stereotypy changes across the lifespan, is not well understood (Langen et al., 2011; MacDuffie et al., 2020; Wolff et al., 2014). Stereotypy is not specific to autism and appears in other neurodevelopmental, medical, and psychiatric conditions such as intellectual disability, schizophrenia, obsessive compulsive disorder and Giles de la Tourette syndrome (Bodfish et al., 2000; Langen et al., 2011; M. Turner, 1999; MacDuffie et al., 2020; Péter et al., 2017). Further, stereotypy is a robust and a key part of early typical development in humans and animals and is thought to play an adaptive role in early learning or organisation of the environment (Langen et al., 2011; Goldman, Wang et al., 2009; Singer, 2009). For example, human infants and toddlers are commonly observed to engage in a range of repetitive behaviours (e.g., body rocking, lining up objects) that tend to dissipate with age, although engagement may persist in a minority of TD individuals throughout the lifespan (D. Wang et al., 2020; Goldman, Wang, et al., 2009). For individuals on the autism spectrum, stereotypy tends to increase in frequency and intensity over time (Bodfish et al., 2000). Research suggests that the extent and severity of repetitive behaviour in children as young as

12 months of age may be greater in children who go on to develop autism, compared to TD children (Wolff et al., 2014).

The fact that stereotypy occurs across a range of human conditions and animal species highlights the role of neurobiological mechanisms underpinning such behaviour (Goldman, Wang, et al., 2009; Langen et al., 2011; 2014; Péter et al., 2017). Although not mutually exclusive, neurological and operant interpretations of stereotypy in autism have historically been examined independently in the research literature, each with distinct assumptions, research questions and methodologies (Lanovaz, 2011; Rapp & Vollmer, 2005b). In essence, the operant model focusses on the role of reinforcement in the maintenance of stereotypy and the manipulation of external (environmental) events through which to treat behaviour (Lanovaz, 2011). Neurological research has implicated the role of the basal ganglia and neurotransmitter systems associated with the basal ganglia (particularly dopaminergic systems) in the emergence and maintenance of stereotypy in autism (Lanovaz, 2011).

An integrated account that combines evidence from operant and neurological models of stereotypy could provide a more comprehensive understanding of stereotypy; however, such integrated explanations have thus far only been theorised (e.g., Lanovaz, 2011; Rapp & Vollmer, 2005b). Lanovaz (2011) proposed that neurological and operant interpretations of stereotypy might overlap in the key area of reinforcement; for example, reinforcement is directly associated with the firing of dopamine cells in the basal ganglia. Differences in central nervous system functioning in autism, particularly in the basal ganglia, may lead to the release of dopamine when stereotypy occurs, which may automatically reinforce and perpetuate such behaviour (Lanovaz, 2011). Further research is needed into how neurological and operant models of stereotypy may interact.

Studies show that severity of stereotypy is associated with younger age, lower NVIQ and symptom severity of autism (Boyd et al., 2012; Goldman, Wang, et al., 2009; Melo et al.,

2020; Péter et al., 2017). Nevertheless, the extent and severity of stereotypy may change over time, with evidence suggesting that some subtypes may increase or decrease in severity, while others remain more stable (MacDuffie et al., 2020). These changes may reflect underlying changes in brain growth and development as children on the autism spectrum mature (Langen et al., 2014). It is also possible that the factors that perpetuate stereotypy are distinct or become divorced over time from factors associated with its origin; for example, repetitive behaviour may serve a (perhaps advantageous) physiological function during infancy but become established and maintained through learned contingencies as children mature (Gwyther et al., 2017). Thus, the aetiology of stereotypy may involve complex and dynamic neurobiological and learned (behavioural) processes. Nevertheless, there is strong evidence that stereotypy falls under operant control (Lanovaz, 2011; Rapp & Vollmer, 2005b), highlighting the crucial role of behaviour management techniques for reducing stereotypy in children on the autism spectrum.

When is Stereotypy a Problem?

Stereotypy tends to occur for individuals on the autism spectrum during times of boredom, stress, excitement, fatigue, or anxiety (McCarty & Brumback, 2021; Melo et al., 2020; Péter et al., 2017). Stereotypy is not necessarily problematic in and of itself; in fact, individuals on the autism spectrum may report experiencing stereotypy as calming and enjoyable (McCarty & Brumback, 2021), and it may serve as a coping mechanism for some individuals by helping to regulate thoughts and emotions (Gabriels et al., 2005; 2013; McCarty & Brumback, 2021). However, stereotypy can be problematic when the behaviour impedes an individual's ability to engage with their environment or interferes with adaptive functioning (Akers et al., 2020; Cook & Rapp, 2020; Didden et al., 2012; S. Patterson et al., 2010). For example, stereotypy can impede learning in the classroom, and can interfere with social interaction and be socially stigmatising (Chebli et al., 2016; Coon & Rapp, 2019; Melo

et al., 2020). In addition, stereotypy can affect family functioning and is associated with severe parental stress (Boyd et al., 2012; Gabriels et al., 2005; Johnson et al., 2018; Lydon et al., 2017). For these reasons, stereotypy is one of the most common intervention targets for children on the autism spectrum (Akers et al., 2020; Horner et al., 2002).

The Relationship between Stereotypy and Sleep Problems

As discussed in Chapter 1, studies show that sleep disturbance can intensify the core symptoms of autism, including stereotypy (Abel et al., 2018; Gabriels et al., 2005; H. Adams, Matson, Cervantes, et al., 2014; Mayes & Calhoun, 2009; Reynolds & Malow, 2011; S. Cohen, Conduit, et al., 2014; Sannar et al., 2018; Schreck et al., 2004; Tudor et al., 2012). Specifically, children on the autism spectrum who have sleep problems are significantly more likely to engage in repetitive behaviours than those without sleep problems (Gabriels et al., 2005: Goldman, Surdyka, et al. 2009; Hundley et al., 2016; Mutluer et al., 2016; Park et al., 2012; Schreck et al., 2004; Zachor & Ben-Itzchak, 2016). The mechanism by which this occurs is yet to be determined (S. Cohen, Conduit, et al., 2014; Schreck & Richdale, 2020); however, it is thought that sleep disturbance increases daytime fatigue, which impairs neurocognitive function and behaviour regulation, making challenging behaviours in general more likely (Gregory & Sadeh, 2012).

Recent research suggests that variability in sleep patterns (Bangerter et al., 2020), specifically in average sleep patterns (i.e., in duration measures of sleep [e.g., TST] across five nights of recording) rather than night-to-night fluctuations (Abel et al., 2018), may be particularly associated with increased repetitive behaviour. There may also be differences for whom stereotypy increases; as shown in a study by Saré and Smith (2020) which found subtle sex differences may exist within this relationship, with stereotypy being more strongly associated with sleep problems in females on the autism spectrum than males.

Subcategories of stereotypy may also be differentially associated with sleep problems; a study by Hundley and colleagues (2016) of 532 participants on the autism spectrum aged 2-17 years found that RSM behaviours were significantly associated with sleep problems after controlling for anxiety, whilst IS behaviours were not. By contrast, MacDuffie et al. (2020) found that sleep problems at age 4-years in children on the autism spectrum were associated with trajectories of higher-order (IS) but not lower-order (RSM) behaviours, after controlling for anxiety, IQ, and social-affective problems. These findings highlight the under-studied complexities of the sleep and stereotypy relationship. Better understanding of this relationship is likely to yield more precise treatment targets to improve sleep and wider functioning in children on the autism spectrum (Hundley et al., 2016; MacDuffie et al., 2020; S. Cohen, Conduit, et al., 2014).

Stereotypy as Sleep-Interfering

As described in Chapter 1, sleep problems can develop and be maintained when children engage in problem behaviour (e.g., calling out, leaving the bed) that disrupts the behavioural chain of falling asleep (Blampied, 2013a). Further, the core characteristics of autism may interfere with the behavioural chain of falling asleep, including that children may experience difficulty transitioning between tasks and interpreting social expectations related to going to bed and to sleep (Mazzone et al., 2018; Reynolds & Malow, 2011; Richdale & Schreck, 2009; Schreck, 2021). As noted above, stereotypy in children on the autism spectrum is pervasive, occurring across settings and time (APA, 2013). It is therefore possible that children who engage in stereotypy during the day will engage in stereotypy at night (Hundley et al., 2016; Johnson, 1996) providing various opportunities for sleep and stereotypy to coincide.

Little consideration has been given in the research literature as to how stereotypy may interfere with the behavioural chain of falling asleep. As for daytime stereotypy, sleep-related

stereotypy may be considered problematic when perseverance in repetitive behaviour incurs a clinical consequence including interference with adaptive functioning (Didden et al., 2012; Gwyther et al., 2017). Specifically, after a child is bid goodnight, stereotypy has the potential to be sleep-interfering by inhibiting a child's ability to establish and maintain the behavioural quietude necessary to initiate and/or re-initiate sleep (i.e., if the child wakes and engages in stereotypy; Blampied & France, 1993; Jin et al., 2013). For these reasons, stereotypy may contribute to sleep problems in children on the autism spectrum by delaying initial sleep onset or the resumption of sleep following NWs. There is strong reason therefore, to assess for the presence of stereotypy in relation to sleep disturbance in children on the autism spectrum.

In addition to the findings of Study 1 (Chapter 4), extant research suggests that stereotypy can accompany sleep problems (e.g., SOD, NWs) in children on the autism spectrum (Jin et al., 2013; Malow, Marzec, et al., 2006; Richdale & Schreck, 2009; Weiskop et al., 2005). For example, in a case analysis by McLay, France, Blampied, van Deurs, et al. (2021; described in Chapter 2) of 41 children and adolescents on the autism spectrum with sleep problems, 27% of participants exhibited stereotypy in the sleep context. The role of stereotypy in relation to sleep in children on the autism spectrum, however, is unclear. While there is reason to conceptualise stereotypy as sleep-interfering (e.g., to delay sleep onset), this may not always be the case. For some children, stereotypy may serve a self-soothing function, perhaps in preparation for sleep. For example, repetitive movements such as head-banging and body-rocking prior to the onset of sleep are behaviours that can be quite commonly observed in infants and young children and are thought to be a possible means of self-settling (Gwyther et al., 2017; Haywood & Hill, 2012). It is also possible that stereotypy occurs as a product of sleep disturbance (i.e., in virtue of children lying in bed awake),

without impacting sleep. Given the heterogeneous nature of stereotypy, it may differentially affect sleep across and within children on the autism spectrum.

Sleep-related stereotypy has not been well-characterised in the research literature, including types of behaviour specific to individuals (Gabriels et al., 2005), and whether night-time stereotypy differs in topography and/or function from that which children engage in throughout the day. Importantly, regardless of its underlying function in relation to sleep (e.g., if it is self-soothing or otherwise), stereotypy may still be problematic if a child's ability to settle or re-settle to sleep efficiently is impaired through perseverance with repetitive behaviour (Gwyther et al., 2017). Parents may also report stereotypy as problematic if it disturbs other family members, such as through noise emitted by repetitive behaviour (Haywood & Hill, 2012). Research is needed to better understand the interplay between sleep problems and stereotypy in children on the autism spectrum (Martin et al., 2019; S. Cohen et al., 2018).

Sleep-Related Movement Disorders in Autism

Several sleep-related movement disorders are associated with autism, including restless leg syndrome (RLS), periodic limb movement disorder (PLMD) and RMD (Richdale & Schreck, 2009; Veatch et al., 2015). The distinction between different types of repetitive movements in relation to sleep disturbance is important to knowing how to treat such behaviour (Veatch et al., 2015; Weiskop et al., 2005). RLS is characterised by an urge to move the legs owing to discomfort that is worse during the evening and periods of rest, and which is (temporarily) relieved by leg movement (Greydanus et al., 2015; Kanney et al., 2020). RLS commonly occurs during the wake-sleep transition and may involve repetitive movement of larger muscle groups involving the legs, such as body-rocking (Kanney et al., 2020; Veatch et al., 2015). RLS is frequently associated with periodic limb movements in

sleep (PLMS) which involves repetitive movement of the limbs during sleep (Veatch et al., 2015).

Although these conditions can disrupt sleep, they are considered primarily neurological or medical in nature (Kanney et al., 2020) and are unlikely to fall under operant control. RLS is a heritable condition associated with underlying dopaminergic dysfunction and low serum ferritin levels, commonly treated with supplemental oral iron (Kanney et al., 2020; Rana et al., 2021). PLMS is a form of parasomnia that occurs during sleep and thus is not under operant control nor is amenable to behavioural treatment. This thesis focusses on the role of the environment and learning (i.e., functions) in sleep problems, particularly whilst children are awake and making the wake-sleep transition (at the start of and during the night if the child wakes). As such, sleep-related movement disorders that lack clear operant qualities to behaviour, including RLS and PLMS, are not discussed further within this thesis.

Comparatively, RMD is a type of parasomnia involving rhythmic, stereotyped movements (e.g., body-rolling or rocking) that can occur during sleep, but often occur during the wake-sleep transition, and can disrupt sleep by delaying sleep onset (Gwyther et al., 2017; Veatch et al., 2015). Although the aetiology of RMD is unclear, it is understood to involve multiple determinants including that RMD may function as operant behaviour (Gwyther et al., 2017). The potential overlay between RMD and sleep-related motor stereotypy as identified in participant children within this thesis is discussed in Chapter 8.

Assessment of Stereotypy

Variation in the function of stereotypy across individuals underscores the importance of FBA prior to any intervention because it provides a basis for case conceptualisation and treatment planning that meets the needs of each individual child (Akers et al., 2020; Boyd et al., 2012; Cunningham & Schreibman, 2008; DiGennaro Reed et al., 2012). Viewed as

operant behaviour, stereotypy must be understood within the framework of contextual variables that serve to occasion and maintain the behaviour (Rapp & Vollmer, 2005a). Assessment of stereotypy using FBA permits the identification of the antecedent environmental conditions and variables that establish the motivation for stereotypy, as well as contingencies of reinforcement that maintain it (Cunningham & Schreibman, 2008; Rapp & Vollmer, 2005a).

Examples of antecedent variables within the sleep context that may control stereotypy include access to items that elicit RMO (e.g., preferred objects or toys) and environmental cues non-conducive to sleep, such as being put to bed too early or in a brightly lit bedroom. If stereotypy (e.g., rocking) has consistently occurred in a particular setting (e.g., in bed) then this particular stereotypy may come under stimulus control in that setting (viz lying in bed). Factors that may increase a child's motivation to engage in stereotypy include the absence of external stimulation in the bedroom, and a state of tiredness (Péter et al., 2017). Conversely, if a child is not motivated to fall asleep (i.e., when there is inadequate homeostatic sleep pressure), then the reinforcement value of stereotypy may be more salient and appealing compared to the delayed reinforcer of sleep. Sources of reinforcement that may maintain stereotypy include the automatic sensory consequences produced by the behaviour itself, or external events such as parent attention and/or the opportunity to escape/avoid a non-preferred activity or sensation (e.g., escape from the demand to go to sleep, avoidance of the dark, avoidance of physical discomfort).

Behaviour is presumed to be automatically reinforced when it persists in the absence of social consequences (i.e., when emitted by an individual experiencing extended periods of time alone [e.g., a no-attention condition in functional analysis procedures]; Akers et al., 2020; Iwata et al., 1982; 1994; Kennedy et al., 2000; Querim et al., 2013; Rapp & Lanovaz, 2016). This might be the case, for example, when a child engages in vocal stereotypy when

alone in their bedroom. In addition to the context automatically reinforced behaviour occurs in (e.g., an alone condition), the structural characteristics of the behaviour may help to inform hypotheses regarding specific sensory reinforcement (e.g., automatically reinforced hand-mouthing may be maintained through tactile mouth (oral) or hand-stimulation, or both; Piazza et al., 2000). The term *automatic reinforcement* implies that, in the absence of obvious external consequences, further assessment is (ideally) required to uncover the specific sensory reinforcers for the behaviour (Kennedy, 1994; LeBlanc et al., 2000). It may be unrealisable, however, to identify all sensory consequences that are reinforcing behaviour, particularly given that multiple modes of sensory stimulation may occur simultaneously (Shore et al., 1997).

FBA of stereotypic behaviour is a particularly important part of this process for multiple reasons. First, stereotypy can appear topographically diverse across different children but serve the same underlying function. Such is the case where repetitive vocalisations for one child and repetitive hand-flapping for another may both be maintained by automatic positive reinforcement. Second, stereotypic behaviours across different children may appear topographically similar but serve different functions (S. Patterson et al., 2010). For example, two children may engage in repetitive hand-flapping; for one hand-flapping may be maintained by automatic reinforcement, and for the other it may serve an escape function (e.g., if a parent habitually stopped asking their child to comply with instructions while hand-flapping occurred).

Third, an individual may engage in diverse forms of stereotypy that serve the same function. For example, a child's stereotyped motor movements such as head-shaking, hand-flapping and jumping may all be reinforced by the automatic sensory consequences. Finally, stereotypy can occur in singular form but serve multiple functions for an individual (Kennedy et al., 2000). For example, repeatedly bouncing on the bed may be reinforced by automatic

consequences, escape from the demand to go to sleep, and social reinforcement (e.g., a parent may lie down with a child in attempt to enforce behavioural quietude). In addition to identifying possible function/s, FBA can help to determine the level of interference stereotypic behaviours cause in relation to sleep (e.g., through consideration of the frequency, intensity, and duration of the behaviour) in relation to the timing of other, sleep-related events. Overall, function cannot be inferred by form alone (Kennedy et al., 2000), so it is imperative FBA is conducted prior to intervention for stereotypic behaviour (Cunningham & Schreibman, 2008; DiGennaro Reed et al., 2012; Horner et al., 2002), including that occurring in the sleep context.

Treatment of Stereotypic Behaviour in Children on the Autism Spectrum

No single treatment approach can effectively reduce all types of stereotypy for all children on the autism spectrum (Cunningham & Schreibman, 2008; Neely et al., 2016). Instead, the type of interventions chosen should depend upon which function or combination of functions the behaviour serves (Cunningham & Schreibman, 2008), in accordance with the unique needs of the individual and family (S. Patterson et al., 2010). One of the notorious challenges in treating stereotypy is how to interrupt reinforcement contingencies that are automatic (i.e., intrinsic to the individual) and resistant to external social manipulation (Boyd et al., 2012; Cunningham & Schreibman, 2008; D. Wang et al., 2020; Gover et al., 2019). One such example is vocal stereotypy maintained by automatic sensory consequences, making it a particularly challenging behaviour to modify, because reinforcement is concomitant with the performance of the response and highly immediate (Lanovaz & Sladeczek, 2012; Rapp & Vollmer, 2005a). It may be impossible or unviable to identify and truly eliminate all sensory consequences that are reinforcing that behaviour (particularly if multiple modes of sensory stimulation [e.g., both sound and vibration] occur; Shore et al., 1997).

Despite such substantial difficulties, there is now a robust body of literature investigating the effectiveness of interventions for daytime stereotypy in children on the autism spectrum (Cunningham & Schreibman, 2008; DiGennaro Reed et al., 2012; Rapp & Vollmer, 2005a). A wide range of antecedent- and consequence-based procedures, based on the principles of ABA, have been used effectively to reduce autism-related stereotypy in children, including stereotypy maintained through automatic reinforcement (Boyd et al., 2012; Cunningham & Schreibman, 2008; Mulligan et al., 2014; Neely et al., 2016; Rapp & Lanovaz, 2016; Rapp & Vollmer, 2005a). Antecedent-based interventions involve modification to the environment or instructional context to prevent or reduce the likelihood of behaviour occurring. Consequence-based interventions involve disrupting the relationship between the behaviour and its reinforcer (Boyd et al., 2012; Lanovaz & Sladeczek, 2012), including the use of differential reinforcement procedures. Treatments that combine multiple components (i.e., antecedent and consequence-based strategies) are found to be more effective at treating stereotypy than either strategy used alone (DiGennaro Reed et al., 2012; Mulligan et al., 2014). An overview of ABA-based strategies with the most empirical support for treatment of daytime stereotypy is presented below.

Antecedent-Based Interventions

Research shows that stereotypy can be modified through antecedent processes (i.e., MOs) that alter the reinforcing value of the events and experiences (i.e., the putative reinforcers) obtained as a consequence of stereotypic behaviour (Rapp & Lanovaz, 2016). As described in Chapter 1, reinforcer value is influenced by the level of satiation or deprivation an individual experiences with regard to a particular event or experience (Lang, O'Reilly, et al., 2010; Michael, 1982; Vollmer & Iwata, 1991). Thus, it may be the case that preventing a child from engaging in stereotypy for a prolonged period of time promotes a state of relative deprivation with respect to that usually received from the behaviour. This deprivation

increases the value of the reinforcer, and, therefore, the child's motivation to engage in stereotypy; in this instance the deprivation is an EO (Michael, 2004). Conversely, allowing a child to engage in stereotypy for a prolonged period may lead to satiation which reduces the value of the reinforcer, thereby, in the immediate future, reducing the child's motivation to engage in the behaviour (i.e., it is an AO; Lang, O'Reilly, et al., 2010; Michael, 2004; Rapp et al., 2017). MOs may provide an important means by which to reduce stereotypy or to enhance the effectiveness of an intervention (Lang, O'Reilly, et al. 2010; Rapp et al., 2017).

Noncontingent Reinforcement (Matched Stimulation). The antecedent intervention with the most empirical support for treatment of stereotypy is noncontingent reinforcement (NCR; Akers et al., 2020; DiGennaro Reed et al., 2012; Mulligan et al., 2014; Rapp et al., 2017; Rapp & Lanovaz, 2016). NCR (sometimes called 'matched stimulation') involves modifying a child's environment to provide access to stimuli (i.e., items/events/experiences) that 'match' the putative sensory consequences for stereotypic behaviour. Identification of matched stimuli is typically done through a preference assessment followed by a competing stimulus assessment. The individual is then provided with continuous access (sometimes on a time-scheduled basis) to the matched stimuli (D. Wang et al., 2020; Rapp & Lanovaz, 2016).

NCR has been primarily applied to vocal stereotypy reinforced through automatic sensory consequences (Gover et al., 2019; Rapp & Lanovaz, 2016). In several studies, noncontingent access to sound-producing toys or music has been shown to reduce vocal stereotypy in individuals on the autism spectrum (Lanovaz & Sladeczek, 2012; Rapp & Lanovaz, 2016). It is assumed that the alternative auditory stimulation generated by music or sound-producing stimuli may satiate the child (i.e., an AO), thereby reducing the motivation to vocalise and the reinforcing effect of the self-produced sound from the vocal stereotypy (Lanovaz & Sladeczek, 2012). Another explanation is that the auditory stimulation masks the

sound produced by the child themselves, thereby disrupting the feedback loop (i.e., sensory extinction; Aiken & Salzberg, 1984).

Although the underlying mechanism is unclear, it is noteworthy that to effectively mask self-produced sound, the alternative auditory stimulation (e.g., music) would need to be at a high enough intensity (i.e., measured in dB) to block the sound. Lanovaz, Sladeczek and Rapp (2011) investigated the effects of manipulating the volume of music on vocal stereotypy in two children on the autism spectrum. They found that noncontingent access to listening to the music decreased immediate engagement in vocal stereotypy for both children and that altering the music intensity did not differentially affect the levels of vocal stereotypy for either child. This suggests that sensory extinction may not be the underlying mechanism for matched stimulation. Research suggests that the efficacy of NCR may increase when coupled with strategies such as social reinforcement for desirable behaviour (differential reinforcement of other behaviour; DRO) and withdrawal of reinforcement for undesirable behaviour (extinction; Boyd et al., 2012; Gover et al., 2019).

Physical Exercise. Another example of manipulation of MOs to reduce stereotypy is physical exercise. Physical exercise such as jogging prior to an activity associated with stereotypy is found to likely function as an AO by reducing subsequent engagement in stereotypy in individuals on the autism spectrum, although the effects may be short-lived (Boyd et al., 2012; Lang, Koegal, et al., 2010; Lanovaz & Sladeczek, 2012). The mechanism by which physical exercise can reduce stereotypy is unclear, however, it may suppress stereotypy via strong response competition and engagement with alternative sources of reinforcement (Lang, Koegal, et al., 2010). There is moderate evidence for physical exercise decreasing motor stereotypy (Rapp & Lanovaz, 2016).

Consequence-Based Interventions

Consequence-based, followed by multicomponent (consequence and antecedent-based) interventions, are the most common approaches employed in the research literature (DiGennaro Reed et al., 2012). Most empirical support has been found for DRO, sensory extinction (response blocking) and punishment procedures, as well as response interruption and redirection (RIRD; Lanovaz & Sladeczek, 2012; Rapp & Lanovaz, 2016).

Differential Reinforcement of Other Behaviour. DRO involves the reinforcement of desirable behaviour while reinforcement of undesirable behaviour is withheld. For example, rewarding an individual with a preferred item for engaging in behaviour other than vocal stereotypy (e.g., communicative verbalisations) for a set amount of time, in the place of vocal stereotypy (Lanovaz & Sladeczek, 2012; Rapp & Lanovaz, 2016). For children with motor stereotypy, the most commonly reinforced alternative behaviour are appropriate play skills (Akers et al., 2020). DRO can also involve the provision of functionally matched stimuli contingent on the absence of stereotypy; B. Taylor, Hoch and Weissman (2005) found that providing access to sound-producing toys as a reward for the absence of vocal stereotypy effectively reduced vocalisations in a child on the autism spectrum and was more effective than noncontingent access to the same toys. DRO has a moderate (Rapp & Lanovaz, 2016) to strong (Akers et al., 2020) evidence-base for treatment of stereotypy in individuals on the autism spectrum.

Punishment. Punishment (positive or negative) refers to any consequence that immediately follows behaviour that serves to reduce the strength of that behaviour (Rapp & Vollmer, 2005a). Examples of positive punishment include a verbal reprimand, brief restraint, and overcorrection, while an example of negative punishment is a response cost (e.g., removal of a preferred item or preventing access to a preferred activity; DiGennaro Reed et al., 2012). One form of overcorrection with high empirical support is positive

practice overcorrection (PPOC; Akers et al., 2020; Rapp & Lanovaz, 2016). PPOC involves being required to repeatedly perform an appropriate behaviour as a consequence of emitting a target response such as stereotypy. For example, when a therapist physically guides an individual to operate a sound-producing toy for a set amount of time as a planned consequence for vocal stereotypy (Rapp & Lanovaz, 2016). Despite a substantial evidence-base for its effectiveness, PPOC, like all positive punishment procedures, is conventionally regarded as intrusive and socially undesirable, and therefore not typically used as a strategy on its own nor recommended as first-line practice (Akers et al., 2020; Rapp & Lanovaz, 2016).

Sensory Extinction. Sensory extinction involves blocking the sensory consequences maintaining a behaviour, such as the use of gloves to prevent a child from sucking their thumb (Rapp & Vollmer, 2005a). Sensory extinction is an evidence-base strategy for the treatment of stereotypy where the function of the behaviour is identified as automatic (B. Taylor et al., 2005; Piazza et al., 2000), and is considered an efficacious treatment for motor stereotypy (DeRosa et al., 2019). Blocking the sensory consequences of the behaviour may decrease the value of the automatic reinforcement for the behaviour because it disrupts the sensory feedback loop (Aiken & Salzberg, 1984; Lanovaz & Sladeczek, 2012). Note however, that sensory extinction, as in the case of any extinction procedure, has the potential to induce an extinction burst, where problem behaviour increases in intensity, duration, and/or frequency before it improves (Lerman & Iwata, 1995; Rapp & Vollmer, 2005a).

In addition, a reduction in a targeted form of stereotypy may lead to an increase in an untargeted form of stereotypy (sometimes called displacement behaviour; Lanovaz et al., 2013), therefore, any intervention that aims to reduce stereotypy needs to ensure that adaptive replacement behaviour is targeted and strengthened (Cunningham & Schreibman, 2008; Lanovaz et al., 2013; Lydon et al., 2017; Rapp & Vollmer, 2005a). For these reasons, most

empirical support has been amassed for treatments that utilise a combination of two or more treatment strategies (DiGennaro Reed et al., 2012). Sensory extinction or punishment procedures, for example, are often incorporated as one component within a multimodal intervention that also includes the teaching of adaptive alternative behaviour (DiGennaro Reed et al., 2012; Rapp & Vollmer, 2005a).

Multicomponent Interventions

Response Interruption and Redirection. RIRD is an intervention comprised of sensory extinction and DRO components, which was developed for and has been primarily used to treat vocal stereotypy (Lydon et al., 2013; Martinez & Betz, 2013). RIRD involves disrupting stereotypy with a request (e.g., in the case of vocal stereotypy, "say ball") until the child complies with the request on three consecutive occasions without stereotypy (Ahearn et al., 2007). Differential reinforcement (DRO) is given through social reinforcement (e.g., praise) for each instance of compliance (Ahearn et al., 2007; Liu-Gitz & Banda, 2010; Vollmer, 1994). RIRD may also be considered to involve an element of punishment, given that instructional requests typically continue until the target compliance rate has been met (Akers et al., 2020; DeRosa et al., 2019). There is strong evidence that RIRD requiring a vocal response is effective for the treatment of vocal stereotypy (Akers et al., 2020; Rapp & Lanovaz, 2016), although variations of RIRD exist in the research literature making the evidence for RIRD as a standardised treatment difficult to interpret (Rapp & Lanovaz, 2016). There is also evidence that RIRD requiring a motor response can effectively reduce motor (Akers et al., 2020; DeRosa et al., 2019; Rapp & Lanovaz, 2016) and vocal (D. Wang et al., 2020) stereotypy.

Inhibitory Stimulus Control Procedures. Another intervention comprised of antecedent- and consequence-based components are inhibitory stimulus control procedures (ISCPs). ISCPs involve bringing stereotypy under stimulus control through discrimination

training, wherein an individual is reinforced for engaging in stereotypy in the presence of a particular stimulus (e.g., a green card), and redirected and not reinforced for engagement in stereotypy in the presence of a different stimulus (e.g., a red card; Akers et al., 2020). Ideally, reinforcement and punishment strategies are eventually faded out, bringing behaviour under the sole control of a specific stimulus (Lydon et al., 2017). If successful, the individual learns to discriminate those circumstances when it is acceptable to emit stereotypy versus those where it is unacceptable (Akers et al., 2020). This procedure would ideally include naturally occurring stimuli such as specific people, places, or activities (Lydon et al., 2017). The evidence-base for ISCPs shows it has promise as an emerging intervention, but further research is needed (Akers et al., 2020; Lydon et al., 2017; Rapp & Lanovaz, 2016).

Summary of Treatments for Stereotypy

In sum, a variety of evidence-based interventions, including antecedent (e.g., NCR), consequence-based (e.g., DRO) and multicomponent (e.g., RIRD) strategies, are available to treat vocal and motor stereotypy in individuals on the autism spectrum (Akers et al., 2020; Mulligan et al., 2014; Rapp & Lanovaz, 2016). A combination of intervention strategies is often employed to treat stereotypy and found to enhance treatment efficacy (Akers et al., 2020; DiGennaro Reed et al., 2012; Gover et al., 2019). For instance, NCR is often used in combination with consequence-based strategies such as reinforcement or response blocking (Gover et al., 2019). Aside from including multiple treatment components within an intervention, preference is given to treatments that enhance adaptive behaviour (i.e., produce desirable collateral effects) and have high social validity, rather than treatments considered to be intrusive, socially undesirable or resource intensive (Lanovaz et al., 2013; Lanovaz & Sladeczek, 2012; Lydon et al., 2017).

More research is needed into specific treatments (and treatment packages) that effectively decrease stereotypy and increase alternative adaptive behaviours in children on

the autism spectrum (DiGennaro Reed et al., 2012; Mulligan et al., 2014; Rapp & Lanovaz, 2016). In particular, research into function-based interventions is needed in order to determine the efficacy of treatments that target a range of behavioural functions (S. Patterson et al., 2010), as well as comparative studies that compare the effects of one or more treatments together (DeRosa et al., 2019). Practitioners in a therapy or education setting implement most interventions for stereotypy in children on the autism spectrum, and there is a need to utilise parents as natural intervention agents to support children's learning long-term in the home setting (Akers et al., 2020; D. Wang et al., 2020). Finally, the effects of treatment on stereotypy over time are largely unknown (Akers et al., 2020).

Treatment of Sleep-Related Stereotypic Behaviour

There is a paucity of research that investigates the effect of treatments for stereotypy within the sleep context (Hundley et al., 2016; Jin et al., 2013), providing a particular impetus for the present research. It is critical that this research is undertaken, as many daytime intervention strategies for stereotypy are impractical to implement in the sleep environment. This is the case for DRO and RIRD as they are complex and intensive interventions, requiring implementation on a continuous basis (e.g., interruption of each occurrence of stereotypy for RIRD; Rapp & Lanovaz, 2016). In the sleep context, a parent would therefore be in the untenable position of needing to be aware of and able to respond to as many instances of night-time stereotypy as possible (D. Wang et al., 2020; Lanovaz & Sladeczek, 2012). As parents are not typically present in a child's room overnight (except for co-sleeping), a major challenge of treating stereotypic behaviour in the sleep context is that children are required to manage their behaviour independently.

This requirement for self-management by the child brings its own challenges, not least that the reinforcer of sleep may be less salient, less desirable, and more delayed than the reinforcement available from sleep-competing activities (Blampied & France, 1993). This

difficulty with self-management may be particularly challenging for children of a younger age and/or limited intellectual ability, who may struggle to refrain from activities that generate immediate reinforcement in favour of the appropriate alternative response. Further, the requirement for parents to act (where possible) as primary interventionists to treat stereotypy within the sleep context is an additional challenge (Hanley et al., 2014; Jin et al., 2013). Research suggests that parents can be effectively taught strategies to reduce stereotypy in the home setting (Gerow et al., 2019; Lanovaz et al., 2016; Specht et al., 2017), however, research is yet to determine strategies that are both suitable to the sleep environment and realisable by parents (D. Wang et al., 2020).

Conversely, some of the limitations and considerations attached to interventions for daytime stereotypy may not pertain to the sleep context. To give one such example, a disadvantage of NCR is that continuous access to preferred stimuli (e.g., music) has limited social validity in daytime settings (e.g., the classroom) where it may decrease learning opportunities (e.g., being unable to hear the teacher; D. Wang et al., 2020; Lanovaz & Sladeczek, 2012; Rapp & Lanovaz, 2016). However, in the sleep context, as discussed in Chapter 2, continuous sound (e.g., white noise) is commonly used in relation to sleep (Reidy et al., 2021). An important consideration for the use of NCR in the sleep context is that the introduced stimuli should be less sleep-disruptive than the stereotypy itself (e.g., stimulating music would not be appropriate; Akers et al., 2020).

Likewise, an important focus of treatment for daytime stereotypy is shaping desirable replacement behaviours (e.g., functional communication in the place of vocal stereotypy), and care must be taken to not inadvertently reduce appropriate behaviour when treating stereotypy (Lanovaz & Sladeczek, 2012; Machalicek et al., 2007). This aspect of intervention (e.g., the 'redirection' component of RIRD used to teach an adaptive behaviour) is less critical in the sleep context where the target response is behavioural quietude and the

transition into sleep. Research is needed to investigate the modifications that established interventions for stereotypy require to be applicable to the treatment of stereotypy within the sleep context, as well as the efficacy of such treatments.

In the absence of research into the effectiveness of treatments for stereotypy in the sleep context, treatments can be selected based upon: (1) what is effective for treating daytime stereotypy; (2) what it is feasible to implement in the sleep context; and (3) the outcomes of FBA. First, effective daytime strategies that may be feasible for use (or adapted for use) to treat stereotypy in the sleep context include manipulation of MOs (e.g., NCR and physical exercise) and RIRD (appropriately adapted for use in the context). Regarding MOs, it may be possible to decrease a child's motivation to engage in stereotypy after being bid goodnight by reducing the reinforcement value of stereotypy prior to the child being put to bed. For instance, allowing a child to freely engage in stereotypy in an appropriate place (e.g., the lounge) prior to going to bed may lead to a state of satiation (Lang, O'Reilly, et al. 2010) thereby reducing the likelihood of stereotypy once the child is in bed (Jin et al., 2013). Further, it may be more reasonable when aiming to decrease stereotypy in the sleep setting to enable and encourage a child to still engage in the behaviour, albeit under more appropriate parameters (e.g., the lounge prior to bed; Akers et al., 2020).

Allowing a child to engage in an activity that provides a competing source of reinforcement may also have an abolishing effect on the value of stereotypic reinforcement after a child is bid goodnight. Physical exercise (e.g., jogging or jumping prior to the commencement of the bedtime routine) may serve as a brief AO for subsequent stereotypy (Lang, Koegal, et al., 2010; Rapp & Lanovaz, 2016). A competing source of reinforcement using matched stimuli could also be applied on a continuous basis (NCR) within the bedroom to reduce the value of reinforcement typically obtained through stereotypy. Thus, preferred music or white noise played in a child's bedroom may help to reduce engagement in vocal

stereotypy by providing a continuous source of alternative reinforcement through the auditory channel. As discussed in Chapter 2, research suggests that white noise may help to improve sleep in children (e.g., France et al., 2018). Thus, manipulation of MOs using one or more of these strategies may be an effective and practical way to reduce stereotypy, particularly vocal stereotypy, within the sleep context.

Concerning motor stereotypy, it may be feasible for a parent to interrupt such behaviour (i.e., response blocking/sensory extinction) in the sleep context and to redirect the child toward falling asleep (i.e., as a modified version of RIRD). For example, a parent could sit next to a child in bed and place a hand on the child when motor stereotypy occurred, as a reminder and encouragement for the child to lie still. This could be coupled with a redirection component, for example, when interrupting instances of stereotypy (e.g., with a hand to lie still), the parent could also give a verbal prompt as redirection toward sleep (e.g., "lie still and go to sleep"). Unlike RIRD, the redirection component would not require consecutive responses from the child since the target behaviour in the sleep context is behavioural quietude. Although it would not be feasible for a parent to interrupt stereotypy throughout the night (i.e., during NWs), it may be possible for a parent to do so during the onset of sleep, until the child learnt to settle to sleep without engagement in stereotypy. The parent's presence could then be systematically faded out of the bedroom.

In general, removal of reinforcement (i.e., extinction) is considered more suitable to the sleep context than procedures that use reinforcement to shape desired behaviour (e.g., DRO), because it is not possible to reinforce a child for falling asleep more efficiently (i.e., without engagement in stereotypy) once asleep (France & Blampied, 2005). Any time extinction is a component of treatment, however, the risk of inducing an extinction burst or unintentionally increasing an alternative form of stereotypy demands consideration in

treatment planning (Lanovaz et al., 2013). Sensory extinction (as one example) may therefore be used as one component of a multimodal intervention (DiGennaro Reed et al., 2012).

In general, treatment strategies for stereotypy such as ISCPs and punishment are not regarded as suitable for use in the sleep context, given the need for intensive involvement from a parent and the socially undesirable nature of punishment. However, it may be useful to include discriminative stimuli (as in ISCPs) as an antecedent component of intervention for night-time stereotypy. A cue card (e.g., a picture of a sun) could be placed on the child's bedroom door to signal when it is acceptable to engage in stereotypy, and an alternate cue card (e.g., a picture of a moon) to signal when it is time to sleep and to not engage in stereotypy. Such visual cues may be a pragmatically useful instructional strategy to incorporate alongside other intervention strategies, such as manipulation of MOs (e.g., allowing a child to freely engage in stereotypy prior to bed) and NCR. Cue cards could also be incorporated in a social story, possibly enhancing the development of stimulus control.

The process of FBA should inform selection of treatment strategies to address stereotypy in the sleep context (Akers et al., 2020; Cunningham & Schreibman 2008; Hanley et al., 2014). Jin et al. (2013; described in Chapter 2) examined the effects of function-based individualised intervention packages to treat sleep problems in three children, two (one on the autism spectrum) of whom engaged in sleep-interfering stereotypy. Stereotypic behaviours included motor stereotypy (e.g., body rocking, head shaking) and RMO (e.g., with clothing items, paper, magazines). Based on the outcomes of FBA, stereotypy was hypothesised to be maintained by access to items that provided reinforcement, which actively competed with the reinforcement of sleep. Motor stereotypy was theorised to be maintained through automatic consequences (Jin et al., 2013). Treatment included the manipulation of MOs; specifically, both children were given the opportunity to freely engage in stereotypy (with full access to the putative reinforcers for RMO) for at least 20 min prior to going to bed. This strategy was

theorised to act as an AO by satiating the child for stereotypy, thereby reducing the likelihood of the behaviour after being bid goodnight. Then, once in bed, access to putative reinforcers for RMO was restricted. In addition, for the child with motor stereotypy, any occurrences of this behaviour were interrupted by parents and the child was redirected to bed (Jin et al., 2013). Treatment was found to effectively reduce sleep problems in all three children, including sleep-interfering stereotypy in two children (Jin et al., 2013). The study by Jin et al. (2013) highlights the importance of tailoring treatment to the functions underlying sleep-interfering behaviour, inclusive of stereotypy.

Summary and Directions for Future Research

Despite high rates of autism-associated stereotypy and of sleep problems, there is a significant lack of research that has explored the impact that stereotypy may have on sleep in children on the autism spectrum. In addition, there is a dearth of research into how to assess and treat stereotypy when it occurs in the context of sleep. Established treatments for daytime stereotypy may not always be appropriate or practical to implement in the sleep context or require modification for use in those circumstances. The process of FBA should be used to inform interventions for sleep-related stereotypy (Akers et al., 2020; Cunningham & Schreibman, 2008; Jin et al., 2013). Some treatment strategies (e.g., NCR) show promise as feasible interventions for sleep-interfering stereotypy, but further research is needed. The next chapter will extend this line of research by investigating the effectiveness of function-based behavioural interventions to treat sleep-interfering stereotypy and sleep problems in children on the autism spectrum.

Chapter 6: Study 2[7]

Sleep-Interfering Stereotypy and Sleep Problems in Children on the Autism Spectrum: The Effectiveness of Function-Based Behavioural Treatment

The outcomes of Study 1 (Chapter 4) revealed that stereotypy was a feature of sleep disturbance in 4/6 participants. This raised the important question of how to assess and treat sleep-related stereotypy, particularly when it is maintained by automatic reinforcement. Chapter 5 underscored the need to develop treatments based on the outcomes of FBA and highlighted several evidence-based strategies for the treatment of daytime stereotypy that may be feasible for implementation in the sleep context. The aim of this study is to extend previous research (Jin et al., 2013) and the findings of Chapters 4 and 5 by investigating:

(1) What is the impact of function-based behavioural treatment on both sleep and stereotypy?
(2) Are treatment effects maintained in the short- and long-term?
(3) Are the selected treatment approaches acceptable to parents, and implemented with fidelity?

Method

Participants and Setting

Three families living in two urban centres in NZ participated. Child participants included two girls and one boy aged 4-5 years, who met the inclusion criteria described in Chapter 3. All three children had communication difficulties (as defined in Chapter 3). Table 6.1 presents a summary of participant characteristics; all names are pseudonyms.

[7] A brief report based on this study has been published in *Sleep Medicine* with myself as first author: Hunter, J. E., McLay, L. K., France, K. G., & Blampied, N. M. (2021). Sleep and stereotypy in children with autism: Effectiveness of function-based behavioral treatment. *Sleep Medicine*, *80*, 301-304. https://doi.org/10.1016/j.sleep.2021.01.062

Table 6.1. *Summary of Participant Characteristics at the Time of Pre-Treatment Assessment*

Participant	Age (Y-M)	Sex	Ethnicity	Diagnosis	VABS-II		Medication	Number of parents in household
					Receptive (Y-M)	Expressive (Y-M)		
Eddy	4-4	Male	NZE Māori	ASD	1-3	2-1	-	2
Tessa	5-1	Female	NZE	ASD	2-5	3-6	Melatonin	1
Bella	5-2	Female	NZE	ASD	2-2	3-6	-	1

Note. ASD: autism spectrum disorder; M: months; NZE: New Zealand European; VABS-II: Vineland Adaptive Behavior Scales 2nd Edition; Y: years

Measures and Materials

Stereotypy

For Tessa, stereotypy was recorded via parent-reported sleep diaries on which they recorded the type, frequency and duration of any stereotypy and their response. Eddy and Bella's parents did not record stereotypy because they were unable to observe and report it (e.g., were in another room, or asleep). For Eddy and Bella, it was measured by video observations; these data were limited to the amount of video recorded for each child (i.e., was not continuous). Data were available for 46%, 34% and 43% of nights for Eddy, and 45%, 30% and 43% of nights for Bella, across the baseline, treatment and STFU phases, respectively. The duration (seconds) of all instances of stereotypy for each child were coded from the point at which repetitive behaviour began, until the child ceased the behaviour (e.g., the child lay still, or transitioned to another activity). The total duration of stereotypy in minutes (rounded to the closest minute) was then calculated during SOL and NWs for Tessa and Bella, and during SOL for Eddy. If the total duration of stereotypy was < 30 seconds (on a single night, across all phases) then it was considered non-problematic and was not

graphed. Only stereotypy that was observable (e.g., repetitive motor movements) could be recorded because the VSG technology used did not support audio recordings.

Procedure

Design

Outcomes were assessed using a non-concurrent multiple baseline across participants design (Barlow et al., 2009; Cooper et al., 2020; Watson & Workman, 1981) with random allocation to three baseline lengths.

Baseline

Upon obtaining informed consent, parents of Eddy and Tessa began recording sleep diaries as part of the FBA process (Baseline 1: seven nights for Eddy, nine nights for Tessa). Each participant was then randomly allocated an additional baseline period (Baseline 2), of one, two or three weeks (consistent with the non-concurrent design). Both Eddy and Tessa randomly received a one-week baseline (Eddy's parents recorded six nights), yielding total baselines of 13 (Eddy) and 16 (Tessa) nights. Both baselines have been included as the data were available and help to establish the trend, level, and variability of baseline data for these children. Bella was randomly assigned a continuous baseline of three weeks (20 nights were recorded), during which FBA was completed.

Function-Based Case Conceptualisation and Treatment Planning

SOD, NWs, and sleep-interfering behaviours, including stereotypy and CCs, were present for all three children, except Tessa for whom CCs were not a problem. The sleep problems, precipitating and maintaining factors, hypothesised functions of the problem behaviour, and individualised treatments for Eddy, Tessa and Bella are summarised in Table 6.2 and discussed below.

Table 6.2. *Summary of Problem Behaviours, Precipitating/Maintaining Factors, Hypothesised Functions and Method of Intervention*

	Problem behaviours	Hypothesised precipitating/maintaining factors	Hypothesised functions	Intervention
Eddy	SOD	Parent attention; access to food; stereotypy	Attention Escape Tangible Automatic	Faded bedtime; altered bedtime routine (satiation); PI; social story; positive reinforcement
	CCs	Parent attention; access to food	Attention Escape Tangible	Altered bedtime routine (satiation); PI
	Stereotypy	Automatic sensory reinforcement	Automatic	Faded bedtime; altered bedtime routine (satiation)
	NWs	Parent attention	Attention	Faded bedtime; PI; Gro-clock™; social story; positive reinforcement
Tessa	SOD	Insufficient sleep pressure; access to preferred items; stereotypy; bedding on floor	Tangible Automatic Escape	Faded bedtime and set wake time; altered bedtime routine (satiation); removal of preferred items; consistent sleep onset in bed; picture card; social story; positive reinforcement
	Stereotypy	Access to preferred items; automatic sensory reinforcement	Tangible Automatic	Faded bedtime and set wake time; altered bedtime routine (satiation); removal of preferred items
	NWs	Insufficient sleep pressure; access to preferred items; stereotypy; bedding on floor	Tangible Automatic	Faded bedtime and set wake time; altered bedtime routine (satiation); removal of preferred items; consistent sleep onset in bed; picture card; social story; positive reinforcement
Bella	SOD	Parent attention; access to preferred items; stereotypy	Attention Tangible Automatic	Faded bedtime; altered bedtime routine (satiation); removal of preferred items; PI; picture card; social story; positive reinforcement

CCs	Access to preferred items; parent attention	Tangible Attention	Altered bedtime routine (satiation); removal of preferred items; PI
Stereotypy	Automatic sensory reinforcement	Automatic	Faded bedtime; altered bedtime routine (satiation)
NWs	Parent attention; protest behaviour; stereotypy	Attention Escape Automatic	Faded bedtime; PI; picture card; social story; positive reinforcement

Note: CCs: curtain calls; NWs: night wakings; PI: planned ignoring; SOD: sleep onset delay

Eddy. Eddy had a reliable bedtime routine, bedtime, and wake time. Presenting sleep problems included SOD, CCs, stereotypy, and NWs. After being bid goodnight, Eddy exhibited motor stereotypy in the form of repetitive rocking lying in a prone position in bed, occasionally accompanied by repetitive vocalising (non-word sounds). Eddy's parents ignored all occurrences of stereotypy. Eddy displayed frequent CCs, in which he often requested food, and needed to be returned to bed by a parent after being fed; thereafter he would re-engage in stereotypy immediately prior to falling asleep. Eddy typically woke 1-2 times during the night, at which point he would enter his parents' bedroom until they returned him to bed. Eddy's parents did not report stereotypy as a problem following NWs.

The FBA indicated that Eddy's sleep onset delay was likely to be primarily because of his sleep-interfering behaviours (CCs and stereotypy), which actively competed with the behavioural quietude necessary for sleep onset. CCs were hypothesised to serve multiple functions for Eddy, including positive (i.e., food and parent attention) and negative (i.e., escape from the demand to go to sleep) reinforcement. Stereotypy was thought to be automatically maintained because it occurred while he was alone. NWs were hypothesised to be positively reinforced through parental attention.

Tessa. Tessa had a consistent bedtime routine and bedtime. Her sleep problems were SOD, stereotypy, and NWs. After being bid goodnight, Tessa exhibited RMO (e.g., lining up toys, spinning car wheels) often accompanied by vocal stereotypy in the form of 'scripting' (i.e., restricted and repetitive vocalisation of scripts from TV programmes). Occurrences of stereotypy were ignored by her parent unless disruptive in volume, in which case a verbal instruction to go to sleep was given; parent instruction was only briefly (e.g., 5-10 min) effective in ceasing the behaviour. Tessa moved her bedding onto the floor each night where she would then settle to sleep. During the night, Tessa would wake and engage in long periods of RMO and scripting. Tessa was woken each morning at varying times.

The FBA indicated that Tessa's SOD and prolonged NWs were primarily due to engagement in stereotypy, which interfered with the initiation and re-initiation of sleep. Stereotypy appeared to be precipitated by access to preferred items (i.e., toys) in the bedroom, and was likely to be automatically maintained because it occurred in the absence of any social interaction. Stereotypy during SOL and NWs were hypothesised to serve the same function. Further, decreased sleep pressure owing to an inconsistent wake time likely contributed to difficulties initiating and maintaining sleep. The FBA indicated that Tessa shifted her bedding on to the floor because of a preference for a hard sleeping surface. Avoidance of lying in bed (i.e., negative reinforcement) may have contributed to Tessa's SOD and NWs (e.g., waking due to discomfort on the ground) and meant that her own bed was not established as a S^D for sleep.

Bella. Bella had a reliable bedtime routine, bed and wake time. Her sleep problems included SOD, CCs, stereotypy, and NWs. After being bid goodnight, Bella exhibited motor stereotypy in the form of repetitive rocking lying in a prone position in bed, which was ignored by her parent. She played with toys and displayed frequent CCs, in which she would call out or leave her bed, needing to be returned each time by her parent; thereafter she would

re-engage in stereotypy immediately prior to falling asleep. During the night, Bella typically woke once for long periods, during which she would call out or enter her sibling's room and needed to be returned to bed by her parent. This was often met with protest. She would engage in motor stereotypy prior to re-initiating sleep.

The FBA indicated that Bella's sleep onset was delayed primarily because of her sleep-interfering behaviours (CCs, play and stereotypy), which impeded sleep onset. CCs were hypothesised to be maintained via access to toys that provided reinforcement, and by parent attention. Stereotypy was likely maintained by automatic reinforcement because it occurred when Bella was alone. NWs were hypothesised to be maintained through positive (i.e., parent attention), negative (i.e., escape through protest behaviour), and automatic (i.e., stereotypy) reinforcement. Stereotypy during SOL and NWs appeared to share the same topography and function.

Intervention

For all parents, intervention goals included: (1) a reduction in sleep onset (i.e., to within 15-30 min); and the elimination of (2) sleep-interfering (stereotypic and CC) behaviours; and (3) NWs. All parents preferred their child falling asleep without stereotypy, considered stereotypy generally disruptive (e.g., from noise), and were concerned it would become increasingly inappropriate as the child aged. In addition, Tessa's parent wished for her to sleep in her bed during the night. Based on the FBA and case conceptualisation, multicomponent, individualised interventions were designed for each child (see Table 6.2 and below). Interventions ran for 35, 29 and 44 nights, for Eddy, Tessa, and Bella, respectively.

Sleep Restriction: The Faded Bedtime Procedure. All children spent extended periods in bed awake during which time other problem behaviours (e.g., stereotypy) occurred. A faded bedtime procedure (Piazza & Fisher, 1991a; 1991b; as described in Chapter 2) was

employed to enhance the value of sleep as a reinforcer (i.e., an EO) and ensure the child was sufficiently motivated to enter into and maintain sleep. The procedure was also used to improve stimulus control for sleep (i.e., establish the bedroom as a S^D for sleep; Bootzin, 1977; Piazza & Fisher, 1991a; 1991b). Further, the faded bedtime procedure was utilised to address multiple sleep problems (i.e., SOD, sleep-interfering behaviours, and NWs) for the child simultaneously (Piazza et al., 1997). Bedtimes for all participants were delayed until 30 min past the time of typical sleep onset. Once each child was reliably falling asleep within 15-30 min over three nights, bedtime was systematically faded earlier (i.e., by 15 min/night) until a desirable bedtime was reached (Piazza et al., 1997). Sleep restriction was also applied in the form of a set wake time (6.30 a.m.) for Tessa, regardless if she had woken during the night (Piazza & Fisher, 1991a; 199b).

Altered Bedtime Routine. To improve stimulus control for sleep, children were kept out of their bedroom (e.g., in the lounge) until the set bedtime each night (i.e., so the bedroom was reserved for sleeping). During this time, routines were altered so that the child's needs were fully addressed prior to being bid goodnight (e.g., toileting, hunger for Eddy). For Eddy and Bella, this included 1:1 time with parents (i.e., engaging in calming activities). All children were able to engage in stereotypy, with toys available to Tessa and Bella, up until the start of the bedtime routine. Encouraging pre-bedtime access to putative reinforcers maintaining the sleep problem (e.g., preferred items, stereotypy, social interaction) was used to satiate the child and reduce motivational state competition (e.g., the child seeking parental attention) after being bid goodnight (i.e., an AO).

Planned Ignoring. To eliminate CCs (Eddy and Bella) and NWs (Eddy and Bella), planned ignoring was used. This involved the immediate withdrawal of parent attention. Instead, parents responded to CCs or NWs by returning the child to bed quickly and quietly, without verbal interaction (children were also toileted if required).

Removal of Preferred Items. To reduce the opportunity for sleep-interfering behaviour, all toys were removed from Tessa and Bella's bedrooms and kept in the lounge. Bella's parents allowed her a choice of one of two soft toys (that were unlikely to prompt play) to take to bed with her each night.

Social Stories. A child-specific 'sleep' story, following the procedures outlined in Chapter 3, was made for each child. The ending depicted a reward for each child for appropriate behaviour.

Visual aids. Visual aids were incorporated to help to establish discriminative control for sleep-conducive behaviour. For Tessa and Bella, a laminated picture card (depicting a 'finished' symbol and moon, respectively) was placed on the back of the bedroom door when the child was bid goodnight, to indicate that it was time for sleep and that activities in the lounge had ended. For Eddy, a Gro-clock^{TM} was introduced to help to reduce NWs. These visual aids also helped parents reiterate that it was sleep time (e.g., by pointing to it and stating "sleep time") if they needed to respond to their child after bidding them goodnight.

Positive Reinforcement. Tangible and social reinforcement (i.e., praise) was used for all children each morning, contingent on the child remaining in their bed until the morning wake time. Parents also praised their child for falling asleep quickly and quietly (i.e., without stereotypy). Eddy was permitted to watch a short video clip of preferred content on his iPad, Bella received a lucky dip choice of small tangible rewards (e.g., a new hair clip), and Tessa was permitted to play with a preferred toy in the lounge before getting ready for school. This contingency was explained to each child verbally and was depicted in the social story.

Additional Individualised Interventions. In order to establish the bed as an S^D for sleep, with parental agreement Tessa's bed-base was removed from her bedroom and a thin

mattress with comfortable bedding was placed on the floor, so that she had a consistent and preferred setting for sleep until a more suitable bed could be obtained for her.

Maintenance and Follow-up

A maintenance phase (as described in Chapter 3) was established once parents felt their treatment goals had been met. STFU was collected at eight weeks and seven weeks post-treatment for Eddy and Bella, respectively. LTFU was collected at 12 weeks and 20 weeks for Eddy and Tessa, respectively. STFU data were unavailable for Tessa owing to parental illness, and LTFU data were unavailable for Bella because the family ceased contact with the researchers.

Interobserver Agreement, Treatment Fidelity, and Treatment Acceptability

IOA, treatment fidelity and treatment acceptability data were gathered following the method outlined in Chapter 3. IOA was collected across study phases for all participants except Tessa, for whom IOA data were only available for the treatment phase due to a technical failure. Treatment fidelity was calculated for treatment and follow-up phases for all children, except STFU and LTFU for Tessa and Bella, respectively.

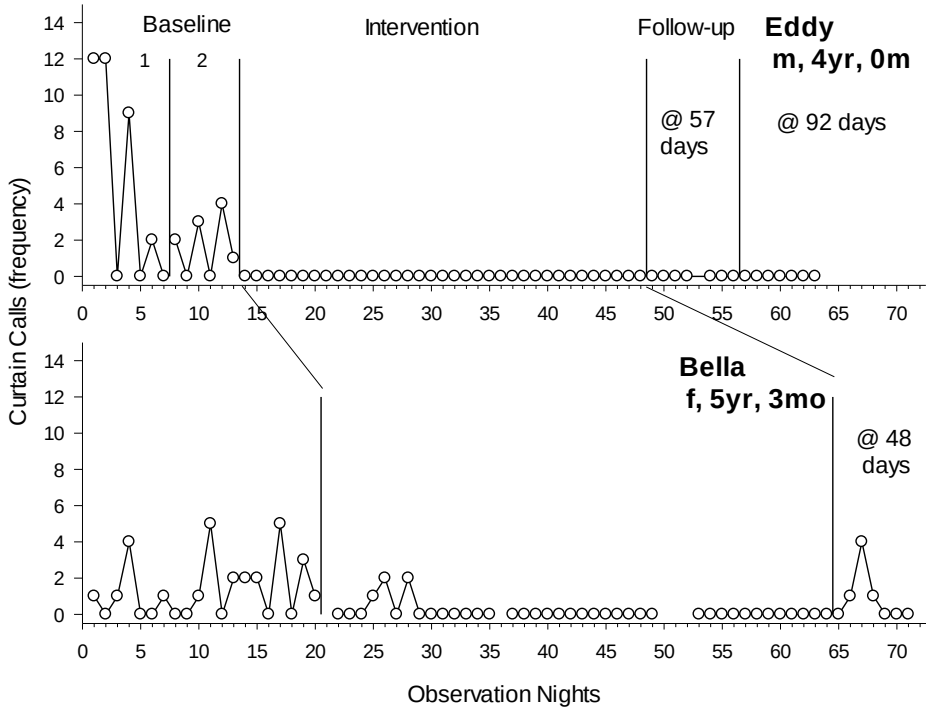
Results

Data were examined using visual analysis of the graphed data and PBM was used as an effect-size measure to supplement visual analysis, as described in Chapter 3. The frequency of CCs, duration of SOL (min), frequency and total duration (min) of NWs, and total duration (min) of stereotypy is shown in Figures 6.1-6.4, respectively. The PBM scores for the frequency of CCs, duration of SOL, frequency of NWs, duration of NWs and duration of stereotypy are presented in Tables 6.3-6.7, respectively.

Curtain Calls

The frequency of CCs for Eddy and Bella is shown in Figure 6.1, with the PBM scores presented in Table 6.3. Missing data points in Figure 6.1 represent periods of illness where no data were recorded. During baseline, the frequency of CCs for Eddy ranged between 0-12 (median: 2) and for Bella from 0-5 (median: 1). CCs were eliminated during treatment for both children. During follow-up, CCs remained eliminated for Eddy, and remained reduced for Bella with a moderate treatment effect (PBM = 86%).

Figure 6.1.



The Frequency of Curtain Calls for Eddy and Bella across Baseline, Intervention and Follow-up Phases

Table 6.3. *Percentage below the Median Scores for the Frequency of Curtain Calls for Eddy and Bella*

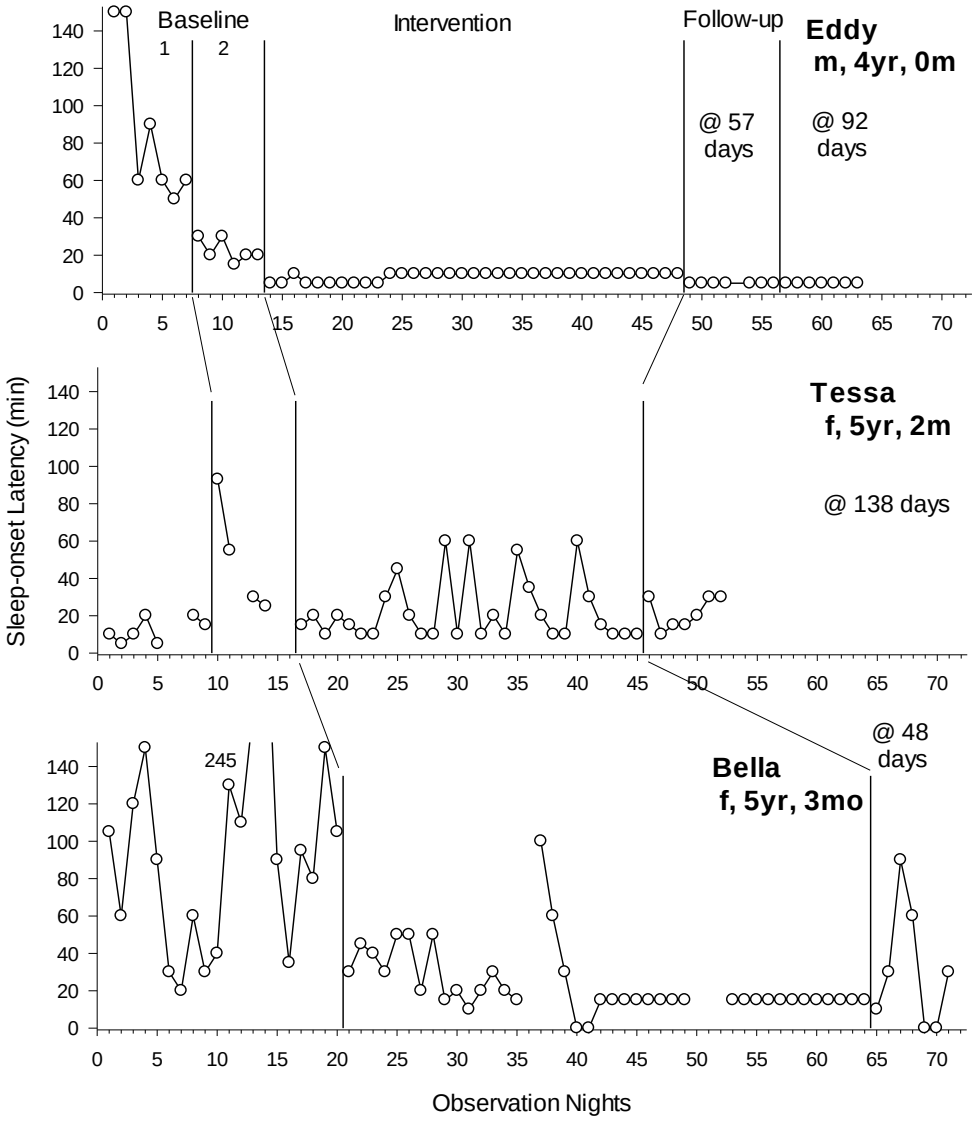
	Treatment	STFU	LTFU
Eddy	100%	100%	100%
Bella	95%	86%	-

Note. LTFU: long-term follow-up; STFU: short-term follow-up

Sleep Onset Latency

The duration of SOL for Eddy, Tessa and Bella is shown in Figure 6.2, with the PBM scores presented in Table 6.4. The number on the graph for Bella gives the duration of the off-scale data point. Missing data points represent periods of illness where no data were recorded. Initially in baseline, Eddy had a prolonged SOL, levels of which then decreased to approximately 60 min and then further reduced to a stable level of approximately 20 min/night. There was an immediate reduction to a sustained low level with treatment (PBM = 100%), which was maintained during follow-up (PBM = 100%). Tessa exhibited an increasing trend in SOL during baseline from an initial low-moderate level. During treatment SOL cycled through peaks of high latency (60 min), with no evidence of a treatment effect (PBM = 55%). At LTFU, SOD $\leq$ 15 min occurred on 3/7 nights, with no evidence of a treatment effect (PBM = 43%). Bella displayed huge variability in SOL in baseline (range: 20-245 min). There was an immediate reduction in variability at treatment outset, with a declining trend. After a spike in SOL mid-treatment following a period of illness, Bella then demonstrated a highly stable SOL of 15 min (PBM = 98%). During STFU, SOL increased in variability (3/7 nights $\leq$ 15 min), however, remained significantly reduced compared to baseline (PBM = 100%).

Figure 6.2.



Duration of Sleep Onset Latency for Eddy, Tessa and Bella across Baseline, Intervention and Follow-up Phases. The number on the graph for Bella gives the duration of the off-scale data point.

Table 6.4. *Percentage below the Median Scores for the Duration of Sleep Onset Latency for Eddy, Tessa, and Bella*

	Treatment	STFU	LTFU
Eddy	100%	100%	100%
Tessa	55%	-	43%
Bella	98%	100%	-

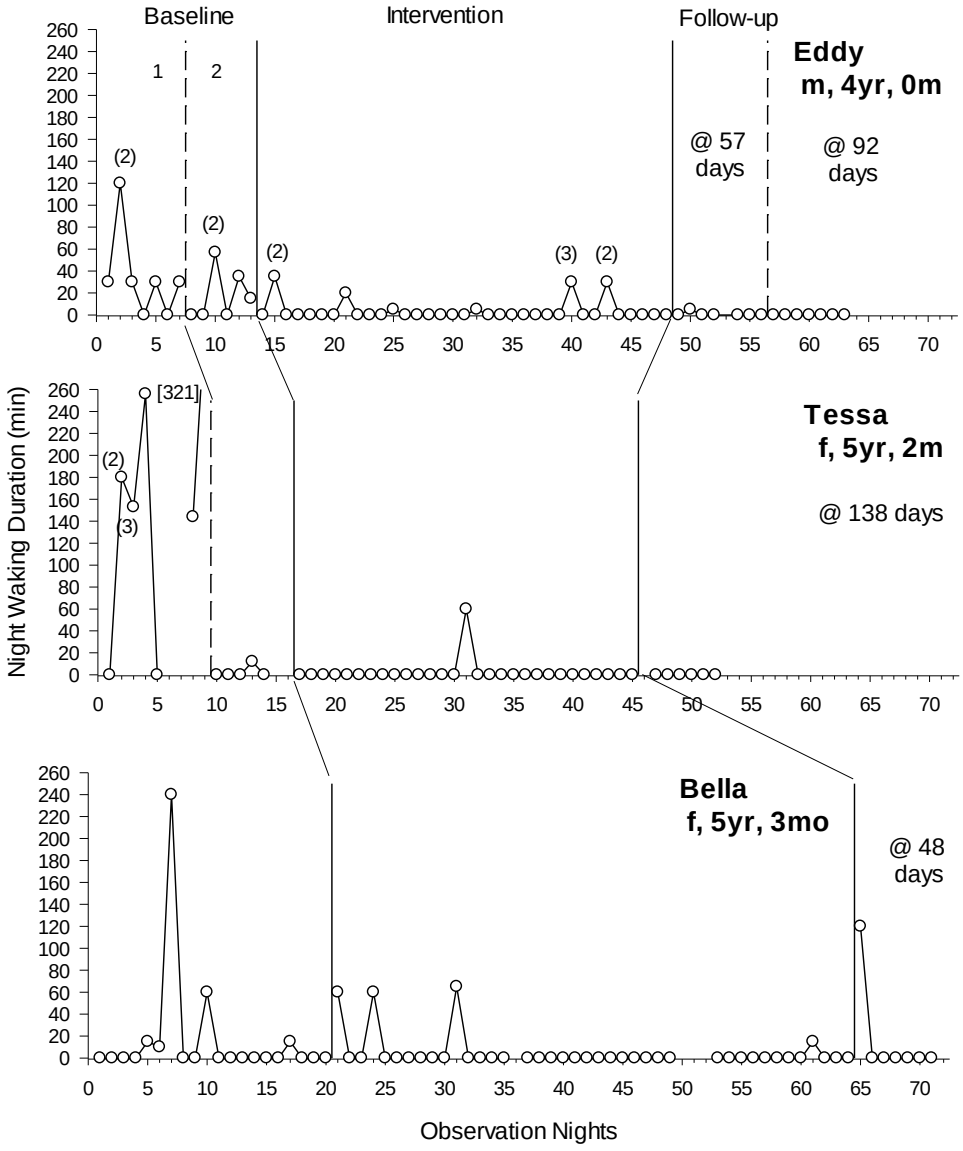
Note. LTFU: long-term follow-up; STFU: short-term follow-up

Night Wakings

The frequency and total duration of NWs for Eddy, Tessa and Bella is shown in Figure 6.3, with the PBM scores presented in Table 6.5. The numbers in rounded brackets on Figure 6.3 show the frequency of wakes (when greater than one) that contribute to the data point. The number in square brackets gives the duration of the off-scale data point. Missing data points represent periods of illness where no data were recorded. Except as noted, all children mostly woke once/night. During baseline, Eddy displayed a pattern of either not waking or waking at a stable level. During treatment, NWs occurred infrequently (6/35 nights) with a moderate treatment effect (PBM = 83%), and the duration was significantly reduced ($\leq$ 35 min; PBM = 91%) compared to baseline. This pattern continued in follow-up (PBM range: 86%-100%).

Tessa's NW durations were highly variable in baseline (range 0-321 min). This pattern abruptly changed towards the end of baseline to brief or no wakings and was maintained in treatment and at LTFU with a moderate-strong treatment effect. The cause of this is unclear, but parent-report suggests the introduction of VSG was a possibility. Bella woke only occasionally in baseline (5/20 nights) and only once for an excessively long duration (240 min) and generally did not wake during treatment and follow-up. PBM scores for the frequency and duration of Bella's NWs could not be calculated because the median was zero.

Figure 6.3.



Frequency and Total Duration of Night Wakings for Eddy, Tessa and Bella across Baseline, Intervention and Follow-up Phases. The numbers in rounded brackets show the frequency of wakes (when greater than one) that contribute to the data point. The number in square brackets gives the duration of the off-scale data point.

Table 6.5. *Percentage below the Median Scores for the Frequency and Duration of Night Wakings for Eddy and Tessa*

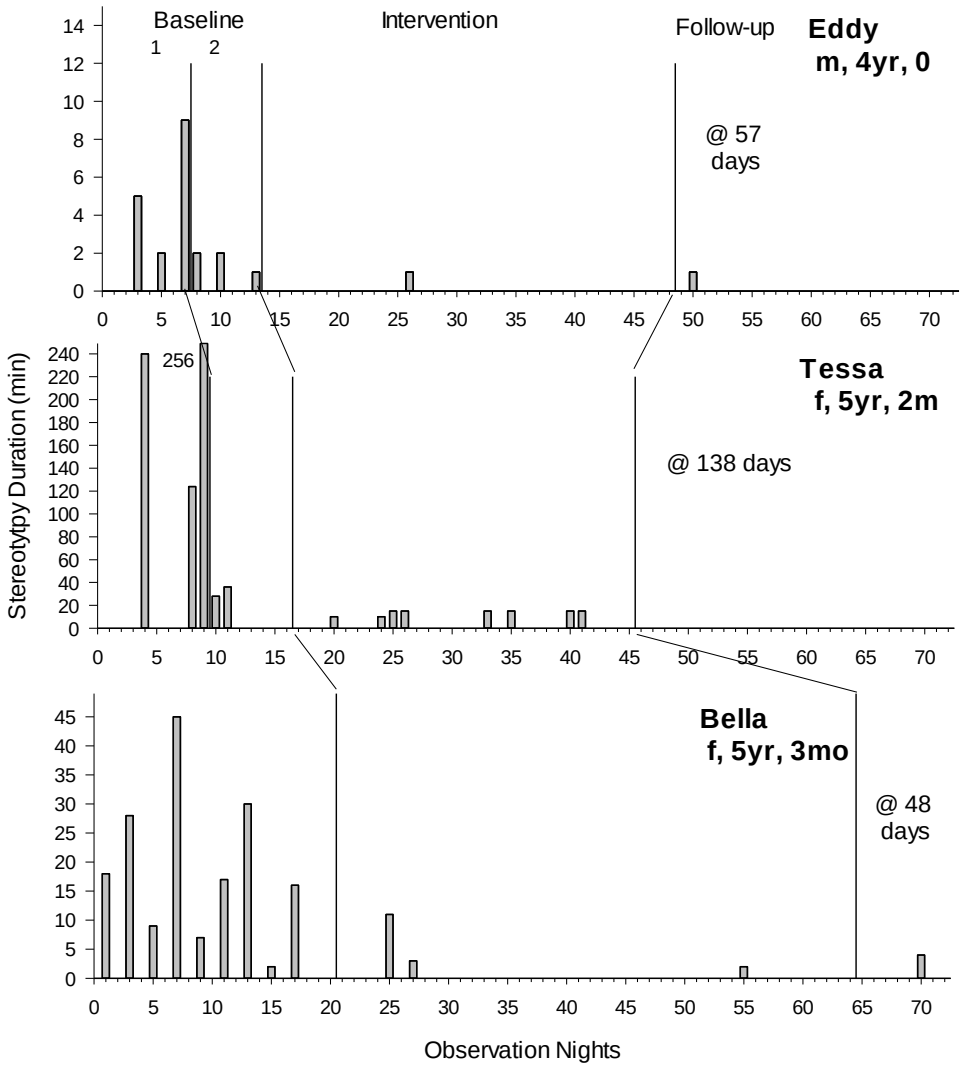
	Treatment		STFU		LTFU	
	Freq	Dur	Freq	Dur	Freq	Dur
Eddy	83%	91%	86%	100%	100%	100%
Tessa	97%	97%	-	-	86%	100%

Note. Dur: duration; Freq: frequency; LTFU: long-term follow-up; STFU: short-term follow-up

Stereotypy

The total duration of stereotypy for Eddy, Tessa and Bella is shown by bar graph (because data were not continuous) in Figure 6.4, with the PBM scores presented in Table 6.6. The number on the graph for Tessa gives the duration of the off-scale data point. The Y-axis scale has been corrected for each child so that changes are clearly visible. All children engaged in stereotypy of varying durations in baseline (range: 1-9, 0-256, and 2-45 min for Eddy, Tessa, and Bella, respectively). For Eddy and Bella, stereotypy reduced from the beginning of treatment and throughout STFU with a strong treatment effect (PBM = 100%). During treatment, Tessa continued to display stereotypy but at reduced levels (0-15 min) both absolutely and relative to baseline, and it was not observed in follow-up. PBM scores for the duration of Tessa's stereotypy could not be calculated because the median was zero.

Figure 6.4.



Total Duration of Stereotypy for Eddy, Tessa and Bella across Baseline, Intervention and Follow-up Phases. The number on the graph for Tessa gives the duration of the off-scale data point. The Y-axis scale has been corrected for each child so that changes are clearly visible.

Table 6.6. *Percentage below the Median Scores for the Duration of Stereotypy for Eddy and Bella*

	Treatment	STFU	LTFU
Eddy	100%	100%	-
Bella	100%	100%	-

Note. LTFU: long-term follow-up; STFU: short-term follow-up

Interobserver Agreement

IOA was collected for 39%, 31% and 35% of nights for Eddy, Tessa, and Bella, respectively and was respectively 97%, 79% and 86%. IOA for stereotypy for Eddy and Bella could not be calculated because their stereotypy was recorded only by VSG.

Treatment Fidelity

Treatment fidelity scores are presented in Table 6.7. Treatment fidelity during the intervention phase was calculated for 34% of sessions for Eddy, and 30% of sessions for Tessa and Bella (range: 94-96%). Treatment fidelity during STFU was calculated for 100% and 43% of sessions for Eddy and Bella, respectively (range: 43-88%). Treatment fidelity during LTFU was calculated for 100% of sessions for Eddy and Tessa, respectively (range: 50-87%). Treatment fidelity was unable to be calculated for STFU for Tessa, and for LTFU for Bella, owing to insufficient information. The overall treatment fidelity scores ranged from 68-94% across children indicating that in general, parents implemented intervention with moderate-high fidelity across phases. For Eddy, fidelity scores decreased during follow-up compared to intervention, whilst for Tessa and Bella fidelity was generally maintained.

Table 6.7. *Treatment Fidelity Scores across Phases*

	Intervention	STFU	LTFU	Overall score
Eddy	94%	43%	50%	68%
Tessa	95%	-	87%	92%
Bella	96%	88%	-	94%

Note. LTFU: long-term follow-up; STFU: short-term follow-up.

Children's Sleep Habits Questionnaire Scores

The CSHQ total pre- and post-treatment scores for all children are presented in Table 6.8. A clinically significant reduction occurred for Eddy, with his post-treatment score falling below the clinical cut-off (41). Tessa's pre- and post-treatment scores only reduced slightly (37 to 36) and were both below the clinical cut-off. Bella's score reduced post-treatment, however, it remained above the clinical cut-off.

Table 6.8. *Children's Sleep Habits Questionnaire Scores*

	Pre-treatment	Post-treatment
Eddy	52	38*
Tessa	37*	36*
Bella	60	51

Note. *Below clinical cut-off

Social Validity

Post-Treatment Interview

All parents felt the intervention improved their child's sleep. Eddy's parents attributed change to the implementation of the Gro-clock™ and consistency in their response to his sleep-related behaviour; specifically, that Eddy was able to wait in bed until the clock indicated morning wake time. They also noted that Eddy's circadian rhythms appeared to be more synchronised with socially acceptable times (i.e., he appeared tired at night and awake during the day). Tessa's parent felt Tessa learned to change her own sleep behaviour in response to the social story. Bella's parent felt the faded bedtime procedure and the bedtime pass were the most effective intervention components. All parents perceived positive secondary effects of intervention, including improvements in their own sleep quantity,

improvements in their child's daytime behaviour (e.g., their child seeming calmer, more tolerant, and/or reduced tantrums, aggression, and repetitive behaviours), and other family benefits including households being tidier and more relaxed. Bella's parent reported that her school attendance increased. Tessa's parent reported she suffered fewer physical complaints (e.g., ear infections). Disadvantages for one parent included lowered motivation to continue with the sleep programme during times of additional external stressors; however, this parent felt that the level of support throughout intervention enabled them to achieve their goals.

The Treatment Acceptability Rating Form - Revised

TARF-R scores are presented in Table 6.9. One parent completed the TARF-R per family. Overall, parents indicated a high level of treatment acceptability (range: 100-104 out of a possible 119). Parents rated treatment favourably in terms of the effectiveness of intervention, reasonableness, cost, and willingness, and viewed treatment as easy to understand. Parents rated the time and effort required to implement intervention moderately, and side effects of treatment was rated as moderate by one parent.

Table 6.9. *Treatment Acceptability Rating Form-Revised Scores*

Scale	Eddy	Tessa	Bella	Maximum Score
Effectiveness	18	18	21	21
Reasonableness	21	20	21	21
Willingness	17	16	18	21
Cost	14	13	14	14
Negative side-effects	17	21	15	21
Disruptive/time	17	16	11	21
Problem Severity*	12	2	8	14
Understanding of treatment*	6	6	7	7
Total Acceptability	**104**	**104**	**100**	**119**

Note. *Not included in total acceptability score

Discussion

This study evaluated the effects of individualised, function-based, behavioural interventions on sleep-interfering stereotypy and sleep problems in three children on the autism spectrum. The maintenance of treatment effects and parent acceptability of treatment procedures were also assessed. Overall, results showed a reduction in CCs for each of the two children for whom this was a problem, and in SOD and NWs for 2/3 children following the BSI. These effects were largely maintained at follow-up. For Tessa, a reduction in NWs occurred beginning during baseline 2, therefore change cannot be attributed to treatment. For all three children, FBA revealed putative sleep-interfering stereotypy as a target for treatment. Results showed that the total duration of stereotypy reduced during treatment and follow-up phases across all participants. This finding supports the research by Jin et al. (2013) and the results of Study 1 (Chapter 4) suggesting that BSI, including the faded bedtime procedure, may effectively reduce sleep-interfering stereotypy. Treatment fidelity and overall acceptance ratings in this present study were high. This finding is consistent with previous research highlighting the importance of parent choice and collaboration in the design and implementation of BSI (Jin et al., 2013; Kirkpatrick, Louw, et al., 2019).

This study included only three case presentations, and therefore there are limited replications on which to base conclusions. Results are therefore tentative and await further direct and systematic replication. One limitation of this study is that stereotypy data were not continuous, so the full impact of treatment on stereotypy was not able to be assessed. Engagement in stereotypy was also not monitored other than in the bedroom, so it is not known whether stereotypy was reduced in total, or rather displaced across settings (Lanovaz et al., 2013). In addition, the lack of audio recordings of vocal stereotypy meant it was not possible to determine whether treatment also had an effect on reducing sleep-interfering vocalisations. It is important for future research to include direct, comprehensive and

continuous measures of stereotypy to assess behaviour change (Cooper et al., 2020). There were also limitations with regards to not being able to collect data until baseline data were stable or rising for all sleep-related behaviours. This may limit certainty in the findings, given the possibility that change may be better attributed to naturally occurring patterns of behaviour, rather than to treatment itself (Kazdin, 1982; L. Cohen, Feinstein, et al., 2014). However, in applied research, it is not always feasible nor desirable to delay treatment for prolonged periods (e.g., owing to clinical need; Carter, 2013).

Caution notwithstanding, the results of this study present important implications for treatment research and for the translation of research into practice. The utility of FBA in informing sleep treatment was underscored by the unique set of factors contributing to sleep problems for each child, including but not limited to, automatically reinforced stereotypy. Results of this study provide tentative evidence of the effectiveness and practicality of manipulation of MOs in treating sleep-interfering stereotypy; however, the mechanism by which this was achieved remains unclear. One possibility is that the faded bedtime procedure manipulated the MOs for sleep by increasing homeostatic sleep pressure (i.e., via mild sleep deprivation), thereby enhancing the value of sleep as a reinforcer and ensuring a rapid sleep onset (Piazza & Fisher, 1991a). In turn, this may have improved stimulus control for behavioural quietude, such that it allowed children to experience falling asleep without stereotypy (McLay, France, Blampied, van Deurs, et al., 2021).

On the other hand, the time children in this study spent in bed awake was greatly reduced, which may have effectively removed the opportunity for children to engage in stereotypy (and other sleep-interfering behaviour), regardless of sleep changes. In the case of Bella, a reduction in stereotypy was found to be maintained despite SOL increasing during follow-up. This suggests that mechanisms other than decreased SOL may be responsible for a maintained reduction in stereotypy over time.

Children were also given pre-bedtime access to preferred items and activities, to increase the likelihood the child would be satiated with regards to putative reinforcement associated with the behaviour, and less likely to engage in sleep-interfering behaviour after being bid goodnight. Once in bed, access to the preferred items and activities was not available. Thus, manipulation of the MOs in treating sleep-interfering stereotypy in this study involved targeting two central, interrelated motivational processes within sleep problems, namely: (a) ensuring a child is sufficiently tired and ready for sleep, and (b) ensuring a child's motivation to fall asleep is not in conflict with motives to engage in sleep-interfering (including stereotypic) behaviour. Strategies that manipulate MOs for sleep such as the faded bedtime may be particularly useful in the treatment of automatically reinforced stereotypy, given the challenges with treating such behaviour as discussed in Chapter 5 (e.g., the sensory consequences are immediate and concomitant with the behaviour itself). Results also show that the interventions were acceptable to parents and implemented with fidelity. Manipulation of MOs warrants further attention as treatment for sleep-interfering stereotypy, and future research would benefit from independently examining the effects of established behavioural interventions for stereotypy (as identified in Chapter 5) applied within the sleep context.

Extant research suggests the core symptoms of autism may affect sleep (H. Adams, Matson, Cervantes, et al., 2014; Hollway & Aman, 2011; Hundley et al., 2016), yet little is known about the specific ways in which stereotypy may contribute to sleep disturbance. This study builds on the findings of Study 1 to provide insight into the individual nature of sleep-interfering stereotypy in children on the autism spectrum. Two separate forms of stereotypy were observed across children in this study; Eddy and Bella exhibited motor stereotypy immediately prior to sleep onset, whereas Tessa exhibited RMO whilst out of bed. For Eddy and Bella in particular, it is not clear whether stereotypy interfered with sleep (i.e., delayed sleep onset), served a self-settling function, occurred because the child was in bed awake, or

if these problems exacerbated each other. As previously noted in Chapter 5, different subtypes of stereotypy may be differentially associated with sleep problems (Hundley et al., 2016; MacDuffie et al., 2020). Determining the function of stereotypy in relation to sleep is an important and interesting avenue for further investigation and is examined further in the next chapter.

Chapter 7: Study 3[8]

The Effects of Sequentially Implemented Function-Based Behavioural Treatment Components on Sleep-Related Stereotypy and Sleep Problems

The impact that stereotypy has on sleep in children on the autism spectrum is currently unclear, including whether stereotypy contributes to sleep problems by delaying sleep onset or the resumption of sleep following NWs, if it occurs as a result of sleep disturbance (e.g., in virtue of a child lying in bed awake), or is a combination of these factors. One means to investigate this relationship is to directly target stereotypy that occurs in the sleep context, separate from other sleep-related problems, and determine whether reducing stereotypy alone decreases problems with sleep onset and/or maintenance. Such an outcome would suggest that stereotypy plays a contributing role in sleep disturbance (i.e., is sleep-interfering).

Further, targeting stereotypy in isolation may help to inform understanding of which interventions can effectively reduce stereotypy in the sleep context. BSI are primarily multimodal, consisting of multiple treatment components (i.e., antecedent- and consequence-based; Carnett et al., 2020; Cuomo et al., 2017). However, this limits our ability to determine the effectiveness of specific procedures in treating sleep problems (Cuomo et al., 2017; Pattison et al., 2020; Vriend et al., 2011) and sleep-related stereotypy. Staggering the introduction of treatment components enables the effects of strategies to be evaluated cumulatively. It also fits with minimally sufficient principles (Johnston & Sherman, 2017; Kazdin, 1984) by using only that which is necessary to change behaviour (van Deurs et al., 2021). The principle of minimal sufficiency underscores the importance of balancing

[8] An article including sections of this chapter pertaining to James' case information has been published in *Advances in Neurodevelopmental Disorders* with myself as the first author: Hunter, J. E., McLay, L. K., France, K. G., Swit, C. S., & Blampied, N. M. (2022). Parent perceptions of sleep-related stereotypy within sleep problems in children on the autism spectrum: Implications for behavioral treatment. *Advances in Neurodevelopmental Disorders*, https://doi.org/10.1007/s41252-022-00246-w

treatment efficacy with ease of delivery when selecting and implementing treatment components (Sanders et al., 2014). It is possible that some treatment components are superfluous within multimodal treatment, which is an important consideration given the time and energy required by parents to implement and maintain a behavioural sleep programme. Further, maximising the effectiveness of treatment within the minimum amount of therapy time needed is important to helping services meet demand.

This chapter reports Study 3, which examined the impact of sequentially implemented function-based behavioural intervention strategies on sleep-related stereotypy and sleep problems in three children on the autism spectrum. In this study, FBA was used to identify the factors contributing to and maintaining sleep problems, including sleep-related stereotypy, for each child. Intervention strategies were then selected in accordance with behavioural function, and with less/least restrictive (i.e., less time and intensity of demands on parents) principles, progressing from antecedent-based procedures (e.g., altering the environment/instructional context and MOs for sleep), which are generally considered less restrictive than extinction procedures, followed by consequence-based (e.g., extinction, reinforcement) strategies, as required (van Deurs et al., 2021). Treatment components were implemented sequentially and cumulatively. This study aimed to answer the following questions:

(1) Does the duration of SOL and frequency/duration of NWs decrease if sleep-related stereotypy targeted in intervention is reduced?
(2) Which behavioural treatment strategies are minimally sufficient to effectively reduce sleep-related stereotypy?
(3) Are treatment effects maintained at short- and long-term?
(4) Are the selected treatment approaches implemented with fidelity and acceptable to parents?

Method

Participants and Setting

Three families living in three different urban centres in NZ participated. Child participants included one girl and two boys aged 3-7 years, who met the inclusion criteria described in Chapter 3. All three children had communication difficulties (as defined in Chapter 3). In addition to ASD, Finn was diagnosed with a rare genetic neurodevelopmental disorder (RGND) called 22q11.2, also known as DiGeorge syndrome. Elsie also had a RGND though the diagnosis was unspecified owing to the rarity of her condition. Table 7.1 presents a summary of participant characteristics; all names are pseudonyms.

Table 7.1. *Summary of Participant Characteristics at the Time of Pre-Treatment Assessment*

Participant	Age (Y-M)	Sex	Ethnicity	Diagnosis	VABS-II		Medication	Number of parents in household
					Receptive (Y-M)	Expressive (Y-M)		
James	7-10	Male	Asian	ASD	2-2	1-8	Melatonin	2
Elsie	5-6	Female	South African European	ASD, RGND	1-4	2-1	-	2
Finn	3-6	Male	NZE	ASD, RGND (22q11.2)	1-2	2-3	-	2

Note. ASD: autism spectrum disorder; M: months; NZE: New Zealand European; RGND: rare genetic neurodevelopmental disorder; VABS-II: Vineland Adaptive Behavior Scales 2nd Edition; Y: years

Measures and Materials

Stereotypy

For James, stereotypy was recorded via parent-reported sleep diaries on which they recorded the type, frequency, and duration (min) of any stereotypy during NWs and their response. For Elsie and Finn, it was measured via video observations; however, no video data

were available for Finn owing to technical failures combined with his parents later choosing not to record video. As a result, stereotypy data were not available for Finn. For Elsie, the duration (seconds) of all instances of stereotypy were coded from the point at which repetitive behaviour began, until she ceased the behaviour (e.g., lay still, or transitioned to another activity). The total duration of stereotypy (during SOL and NWs, rounded to the closest minute) was then calculated; if the total duration was < 30 s on a single night then it was considered non-problematic, and was not graphed.

Procedure

Design

Outcomes were assessed using a single-case AB (baseline [A], intervention [B]) design. Baseline lengths were randomly allocated, and treatments were added cumulatively to assess the added benefit of each treatment component. For all children, stereotypy was targeted first because it was identified in the FBA as a primary problem behaviour and targeting stereotypy in isolation allowed the impact of treatment on stereotypy to be investigated (e.g., whether reducing stereotypy reduced SOD). Treatment effects were replicated across target sleep variables including stereotypy, SOD, CCs, and NWs.

Baseline

James was randomly allocated a three-week baseline, which was extended to 36 nights (baseline 1) to accommodate family needs. Prior to intervention, melatonin (1 mg) was eliminated in accordance with parents' goals, yielding an additional 15 nights of baseline data (baseline 2). Elsie had two separate baseline and treatment phases, in 2017 and 2018. In 2017, she was randomly allocated a baseline period of one week (9 nights were recorded); the family later withdrew from the study during the treatment phase. In 2018, Elsie's family returned to the study, and she was again randomly allocated a baseline period of one week (5

nights were recorded). Finn was randomly allocated a two-week baseline, which was extended to 50 nights (31 nights were recorded) to accommodate family needs and child illness.

Function-Based Case Conceptualisation and Treatment Planning

The problem behaviours, hypothesised precipitating/maintaining factors, behavioural functions, and individualised treatment plans for each child are summarised in Table 7.2 and described below. Evidence-based treatment components were selected according to their suitability to address the function of the behaviour and were implemented sequentially across treatment phases to examine the added benefit of each treatment component. Treatment proceeded to the next phase if reductions across target sleep variables were not evident within five days. Parents' intervention goals included: (1) a reduction in sleep onset latency (i.e., to within 15-30 min); and the elimination of (2) sleep-interfering (stereotypic and CC) behaviours; and (3) NWs. In addition, Elsie's parents wished to eliminate co-sleeping. Co-sleeping was not a parent-defined problem for James; however, his parents preferred to sleep independently if his sleep problems (particularly NWs) could be managed by other means. James' parents also wished to eliminate use of melatonin (1 mg).

Table 7.2. *Summary of Problem Behaviours, Precipitating/Maintaining Factors, Hypothesised Functions and Method of Intervention*

	Problem behaviours	Hypothesised precipitating/maintaining factors	Hypothesised functions	Intervention
James	SOD	Inconsistent S^D for sleep; insufficient sleep pressure; parent attention	Attention	Sleep restriction; Gro-clock™; social story; PP; PI; positive reinforcement
	NWs	Insufficient sleep pressure; stereotypy; parent attention	Attention; automatic	Sleep restriction; Gro-clock™; social story; PP; PI; positive reinforcement
	Vocal stereotypy	Auditory sensory feedback; parent attention	Automatic; attention	Matched stimulation (white noise); PP; PI
Elsie	SOD	Parent attention; stereotypy	Attention; automatic	Sleep sack; social story; PP; PI
	CCs	Parent attention	Attention	PP; PI
	NWs	Parent attention (co-sleeping); stereotypy; inconsistent S^D for sleep	Attention; automatic	Sleep sack; hug pillow; social story; PP; PI
	Motor stereotypy	Automatic sensory feedback; parent attention (co-sleeping)	Automatic Attention	Sleep sack; hug pillow; PP; PI
Finn	SOD	Insufficient sleep pressure; lack of stimulus control for sleep; access to preferred items; parent attention; stereotypy	Tangible; attention; automatic	Faded bedtime procedure; altered bedtime routine; finished box + reward; sleep restriction (set wake time, no naps); social story; PI*
	CCs	Access to preferred items; parent attention	Tangible; attention	Altered bedtime routine; finished box + reward; PI*
	Motor stereotypy	Automatic sensory feedback	Automatic	Indoor trampoline
	Vocal stereotypy	Automatic sensory feedback	Automatic	White noise

Note: CCs: curtain calls; PP: parental presence procedure; PI: planned ignoring; NWs: night wakings; S^D: discriminative stimuli; SOD: sleep onset delay; *planned strategy but not implemented.

James. James had a consistent bedtime routine including taking 1mg melatonin, and reliable bedtime that was intentionally delayed by his parents (10.00 p.m.) as a means of managing NWs (i.e., they reported an earlier bedtime resulted in a higher frequency of NWs). His sleep problems consisted of SOD and NWs. After being bid goodnight, James took 30-40 min to fall asleep, during which time a parent would intermittently lie with him. His NWs, although infrequent, were long (mean = 90 min, range = 30-180 min). James engaged in vocal stereotypy involving repetition of numbers, movie phrases and non-contextual laughter, which was predominantly problematic during his NWs. He also engaged in vocal stereotypy during SOD; however, his parents considered this to be relatively unproblematic because these vocalisations were generally low in frequency, intensity, and duration. A parent co-slept with James as a pre-emptive means of managing his NWs; when he woke, the parent responded by ignoring his vocalisations, feigning (modelling) sleep, or using short sentences to redirect James to return to sleep. Parent-report indicated none of these responses altered James' behaviour. In the morning, James woke on his own at inconsistent times, and frequently napped at school during the day.

The function-based case conceptualisation indicated that James' stereotypy and sleep problems were multiply determined. Antecedent factors that appeared to contribute to sleep problems included the intermittent presence of a parent during the sleep onset period (i.e., providing an inconsistent S^D for sleep onset) and insufficient physiological sleep pressure owing to inconsistent wake times and daytime naps. Stereotypic vocalisations during NWs appeared to actively compete with behavioural quietude and thereby prevent the re-initiation of sleep. The reinforcing consequences for vocal stereotypy were likely to be automatic because the vocalisations occurred if he was alone, or not. Further, the auditory sensory stimulation produced by the vocalisations were theorised to be reinforcing. Because vocal stereotypy often resulted in parent-attention, it was also possible that it was (partially)

reinforced by attention. Parent attention (social interactions whilst awake) was also likely to contribute to the maintenance of NWs, which were likely to be further complicated by James' reliance on a parent's presence for sleep (i.e., inappropriate stimulus control).

James' treatment (T) ran for 80 nights and consisted of four sub-phases with sequential addition of treatment elements: (T1) white noise to target vocal stereotypy (night 52 to 71); (T2) white noise, sleep restriction and visual aids (night 72 to 101); (T3) white noise, sleep restriction, visual aids and gradual reduction of parental presence (night 102 to 121); (T4) white noise, sleep restriction, visual aids and the full removal of parental presence (night 122 to 131). It was hypothesised that if vocal stereotypy interfered with James' ability to re-initiate sleep, then reducing stereotypy may subsequently reduce the duration (but not frequency) of NWs. White noise (i.e., matched stimulation, as described in Chapter 5) was selected to target the putative automatic auditory consequences produced by his vocalisations. It was hypothesised that the alternative auditory stimulation (i.e., white noise) would have a satiating effect on the stereotypy and thus reduce his motivation to engage in this behaviour (i.e., an AO). White noise was chosen because the consummatory response of listening to white noise was likely to be less sleep-disruptive than the self-produced vocalisations. White noise was also expected to be less stimulating than music, and, as discussed in Chapters 2 and 5, is a readily available strategy that is feasible for parent-implementation in the home-setting overnight and is generally well-tolerated by children.

T1 involved playing white noise (continuous rain sound) in James' bedroom through an application on a portable device. James' parents turned on the white noise once James was in bed and turned it off once James was awake in the morning. The volume was held consistent at 70 dB across treatment and follow-up phases; this volume helped to mask sounds whilst being comfortable for James (Knight & Johnson, 2014).

In T2, sleep restriction procedures were implemented to manipulate the MOs for sleep; specifically, to increase physiological sleep pressure and thereby strengthen James' motivation to initiate and maintain sleep during the night. Sleep restriction involved establishing a consistent morning wake time (7.30 a.m.) and the elimination of daytime naps. James' bedtime of 10.00 p.m. was continued. In addition, visual aids in the form of a Gro-clock™ and a social story were introduced to establish a discriminative stimulus for night-time (e.g., as a visual cue to return to sleep following a NW) and morning wake time. The social story illustrated James' sleep routine and expectations around sleep, with an emphasis on sleep-conducive behaviour (e.g., lying quietly in bed without vocal stereotypy). The ending depicted a reward for James for appropriate behaviour.

In T3, a parental presence procedure was implemented to improve stimulus control for sleep onset (i.e., during SOL), and to eliminate the possible attention-seeking function of vocal stereotypy and NWs. During the sleep onset period, a parent sat on a chair near James' bed without interacting with him, and during the night, following a NW, a parent slept in James' room but in a bed separate to him. The parent was instructed to ignore James' behaviour during SOL and NWs, where safe to do so.

In T4, parental presence was fully removed; rather than sitting on a chair during SOL the parent bid James goodnight and left the room, and during the night slept in their own bed, ignoring James' behaviour during any NWs where safe to do so.

Elsie. Elsie had a consistent bedtime routine and reliable bed and wake time. Her sleep problems were SOD, CCs, NWs, and unwanted co-sleeping. After being bid goodnight, Elsie stood on her bed and engaged in a standing side-to-side body-rock, often pushing off the wall with one hand. A parent would respond by lying Elsie back down in her bed; however, she typically stood and re-engaged in rocking as soon as the parent left the room. Elsie also intermittently left her bed and entered the lounge, upon which a parent would

return her to her bed. Elsie rocked until sleep onset occurred; that is, until she physically dropped into a prone position and slept. Elsie frequently woke 1-2 times during the night, during which she would stand and rock. A parent would respond by lying her down and leaving (i.e., returning to the parent's own bed), or by co-sleeping with Elsie in her bed for the remainder of the night. If the parent left, Elsie would often wake again, upon which co-sleeping typically occurred. Elsie's parents reported that co-sleeping assisted Elsie's return to sleep because the presence of the parent helped to physically restrict her motor movements such that she remained lying down.

The function-based case conceptualisation indicated that Elsie's sleep problems were maintained via social and non-social consequences. Her sleep onset was likely to be delayed because of her engagement in stereotypy and CCs. CCs appeared to be reinforced by access to parent attention, and stereotypy was hypothesised to be automatically maintained because it occurred when she was alone in her bedroom. As stereotypy often resulted in attention, however (including co-sleeping during NWs), it was also possible that stereotypy was (partially) socially maintained. Her NWs appeared to be perpetuated by engagement in stereotypy and by parent attention. NWs were further complicated by intermittent co-sleeping; the presence of a parent created an inconsistent and inappropriate S^D for the resumption of sleep. Based on parent report, it was also possible that the presence of a parent helped to restrict her motor movements allowing her to resume sleep, perhaps providing a form of sensory extinction by blocking her engagement in the behaviour.

Elsie's treatment ran for 28 (2017) and 21 (2018) nights before her parents withdrew from the study. Reasons cited for withdrawal included family commitments that made adherence to a behavioural programme and data collection difficult. In 2017, stereotypy was targeted first (i.e., in isolation) because it was identified by the FBA as a primary problem behaviour and because doing so enabled its impact on Elsie's sleep to be examined. A 'sleep

sack' (like a sleeping bag) was made to target the automatic function of her rocking by restricting her motor movements (thereby replacing the potential need for a parent to co-sleep with her during NWs). The sack was made by a qualified tailor and the author using cotton and Lycra material that meant it was stretchy and lightweight. It was loose enough that she was able to get in and out of it on her own. Elsie wore the sleep sack under her duvet when she got into bed each night. In addition, a 'hug pillow' (a pillow shaped like a person's torso with an outstretched arm) was introduced as a sleep item for her to hold inside her sleep sack. The sleep item was provided to create a new, consistent S^D for sleep and Elsie's parents were able to instruct her to cuddle it during the night if they needed to redirect her toward sleep. A social story was made that described sleep-conducive behaviour (e.g., lying quietly in bed) and use of the sleep sack and hug pillow.

Upon returning to the study in 2018, Elsie continued to use the sleep sack and sleep item (the hug pillow had been replaced with a soft toy by her parents) each night. Following her 2018 baseline, a parental presence procedure was implemented (in addition to the sleep sack and soft toy) to remove the contingency of parent attention (including co-sleeping) during the initial sleep onset period and NWs. During her initial sleep onset, a parent sat in a chair near Elsie's bed without interacting with her, until she fell asleep. The parent then left the room to sleep in their own bed overnight, returning to sit in the chair if Elsie woke, until she resumed sleep. If Elsie stood up to rock or left the bed, a parent returned her to bed and/or restored her sleep position in the sleep sack and directed her to hug her soft toy. On night seven, a second treatment phase (T2) involved the withdrawal of parental presence; after Elsie was bid goodnight the parent immediately left the room, ignoring Elsie's behaviour during the sleep onset period and NWs where safe to do so. A new version of the social story was made, which, in addition to the original story, emphasised her sleeping independently.

Finn. Finn had a consistent bedtime routine and bedtime of 7.30 p.m., however, he had substantial SOD. After being bid goodnight, he engaged in motor stereotypy (bouncing on his backside in bed) frequently accompanied by vocal stereotypy (squealing) and made numerous CCs requesting food and preferred items (e.g., books, toys). Motor stereotypy often occurred in the context of watching a preferred TV programme on a mobile phone and looking at a preferred book in bed. Immediately prior to falling asleep, Finn wrapped himself in a blanket and vigorously rolled side-to-side in a supine position until sleep onset occurred. He typically fell asleep between 12.00–2.00 a.m., and once asleep, slept through until he either woke on his own around 10.00 a.m. or was woken for preschool (two mornings per week) at 8.00 a.m. He frequently napped throughout the day. Finn's parents reported he appeared tired during the day and wide awake at night.

The function-based case conceptualisation indicated that Finn's sleep onset was delayed because of insufficient sleep pressure (owing to a delayed wake time and daytime naps) at night and his sleep phase was discordant with conventional day-night rhythms. Further, it was probable that his engagement in stereotypy and CCs actively interfered with behavioural quietude and the onset of sleep. Access to preferred items (i.e., phone, book) likely contributed to the occurrence of stereotypy (bouncing and squealing), which appeared to be automatically reinforced as it occurred when Finn was alone in his bedroom. CCs were likely to be reinforced by access to preferred items and parent attention. Finally, the extended periods of time that Finn spent in bed awake meant that there was a lack of stimulus control for sleep (e.g., his bed was not an established S^D for sleep).

Finn's treatment ran for 28 nights before the family withdrew during the treatment phase. Reason for withdrawal was not explicitly given; however, the family noted that several medical/developmental conditions across family members for which they were receiving support from multiple external services complicated their ability to adhere to the

requirements of the treatment programme. In T1, motor stereotypy was targeted in isolation through the introduction of a small indoor trampoline. Finn was encouraged to bounce on the trampoline for 20-30 min immediately prior to the commencement of his short bedtime routine. It was theorised that bouncing prior to bed on the trampoline would satiate Finn's desire to bounce once in bed (i.e., acting as an AO), thereby helping to reduce sleep-related stereotypic behaviour. Because of the severity of SOD (e.g., up to 7 hours), it was deemed clinically necessary to introduce additional strategies rapidly if reducing stereotypy had no effect on other sleep variables, so T1 was brief.

On night six, T2 included the faded bedtime procedure and altered bedtime routine to improve stimulus control for sleep; Finn was kept out of his bedroom until 10.00 p.m. This time was agreed upon in collaboration with Finn's parents as a balance between moving closer to his natural sleep onset time whilst being feasible for his parents to keep him up and out of his bedroom (e.g., before the parents' own bedtime). During this time, Finn's bedtime routine was altered so that he received access to items that provided putative reinforcement for sleep-interfering behaviours prior to bed (i.e., acting as an AO); this included being given a light snack, 1:1 time with parents, and access to preferred items including his phone and book. He was freely able to engage in stereotypy and continued to use the trampoline during this time.

Prior to going to bed, Finn packed his book and phone into a 'finished box'; he received a small reward (a preferred edible item) for compliance with this step. To increase physiological sleep pressure, sleep restriction was implemented using a set wake time (7.45 a.m.) and the elimination of daytime naps. A social story was made that depicted his bedtime routine (including use of the trampoline and finished box), sleep-conducive behaviour (e.g., lying quietly in bed with a 'calm body') and expectations around his sleep. The story ended with Finn receiving access to his finished box in the lounge upon waking in the morning.

On night 13, T3 involved the addition of white noise to target vocal stereotypy (squealing). As described for James above, white noise was selected to target the putative automatic auditory consequences produced by Finn's vocalisations. It was hypothesised that the alternative auditory stimulation (i.e., white noise) would reduce his motivation to engage in vocal stereotypy (i.e., an AO). Finally, it was intended that planned ignoring would be implemented as a fourth treatment phase to remove the contingency of parent attention and place CCs on extinction; however, the family withdrew before this phase was implemented.

Maintenance and Follow-up

For James, a maintenance phase (as described in Chapter 3) was established once his parents felt their treatment goals had been met, and STFU and LTFU data were collected for seven nights at 24 and 73 days post-treatment, respectively. No follow-up data were collected for Elsie or Finn because the families withdrew from the study.

Data Analysis

As detailed in Chapter 3, data were examined using visual analysis of the graphed data with assessment of change in level, trend and variability across phases and cases. PBM was used as an effect-size measure of single-case data to supplement visual analysis. The PBM scores for James, Elsie and Finn are presented in Table 7.3. Additional intervention phases are indicated by phase-change lines labelled Treatment 1 (T1) Treatment 2 (T2) and so forth (see Figures 7.1-7.3).

Results

James

The dependent variables for James were the total duration of vocal stereotypy during NWs, duration of SOD, and frequency and duration of NWs (see Figure 7.1). The number 2 on the graph indicates that James woke twice; otherwise, he woke only once/night. During

baseline 1 and 2, the duration of vocal stereotypy ranged from 15-180 min (mean = 87 min). During T1 (white noise) there was no evidence of a treatment effect, nor throughout the other treatment phases. Although he continued to vocalise during NWs, the occurrence of stereotypy reduced in accordance with reductions in NWs, as described below. PBM scores for the duration of vocal stereotypy could not be calculated because the median was zero.

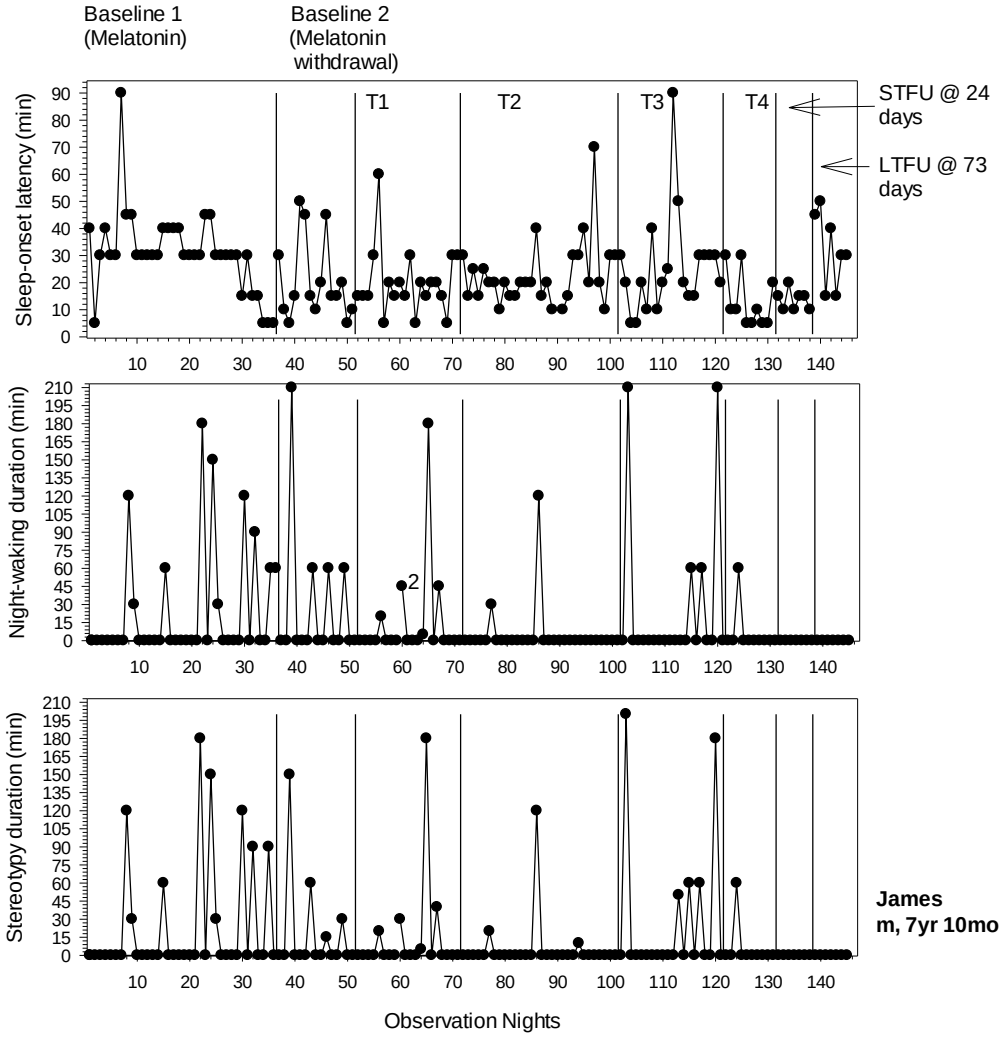
During baseline 1, SOD was 30-40 min on average. After melatonin was eliminated, SOD ranged from 5-45 min (mean = 21 min). In comparison to baseline 2, there was no evidence of a treatment effect in T1, T2 nor T3 (PBM = 15%, 14% and 20%, respectively). In T4, SOD reduced with a moderate-sized treatment effect (PBM = 70%), with all nights $\leq$ 30 min and 7/10 nights $\leq$ 15 min. Although a treatment effect was no longer evident at STFU (PBM = 43%), visual analysis shows that the target goal (SOL 15-30 min) was still met, with SOL $\leq$ 20 min each night and 6/7 nights $\leq$ 15 min. At LTFU, a treatment effect was not evident (PBM = 0%).

During baseline 1 and 2 James woke infrequently (invariably once) but for extended periods of 30-120 min. There was no evidence of a treatment effect during T1 and on one occasion James woke twice. During T2, NWs reduced to near zero with one wake on 2/29 nights (duration 30 and 120 min). During T3, James woke once on four nights (duration of either 60 or 120 min), and in T4 he woke once for 60 min. No NWs occurred during follow-up. PBM scores for the frequency and duration of NWs could not be calculated because the median was zero.

In sum, white noise in isolation had no effect on vocal stereotypy nor other sleep variables, but a combination of white noise, sleep restriction and visual aids appears to have reduced (at least partially) the frequency of NWs. A further reduction in NWs and a reduction in SOD was observed when the parent was fully removed from the bedroom, supporting the hypothesis that SOD and NWs were at least partially maintained by parent attention.

Improvements may have also occurred owing to improved stimulus control for sleep. Although the frequency of NWs reduced, the duration of NWs did not. When James woke, he tended to remain awake for at least an hour, and he vocalised during this time. The absence of NWs particularly during follow-up concomitantly resulted in the elimination of vocal stereotypy; that is, it appears vocal stereotypy reduced owing to a lack of opportunity to perform the behaviour, because he was no longer waking at night.

Figure 7.1.



Sleep Outcomes for James: Duration of Sleep Onset Latency, Night Wakings and Vocal Stereotypy across Baseline, Intervention and Follow-up Phases. The number 2 on the graph indicates that James woke twice; otherwise, he woke once/night. T1 = white noise; T2 = + sleep restriction, Gro-clock™, social story, positive reinforcement; T3 = + parental presence; T4 = + planned ignoring

Elsie

The dependent variables for Elsie were the duration of stereotypy and SOD, frequency of CCs, frequency and duration of NWs, and co-sleeping (see Figure 7.2). The open circles on the graph represent nights without co-sleeping. The numbers 2, 3 and 4 show the number of wakes when greater than one. Missing data points represent periods where data were not recorded. In her 2017 baseline, the total duration of stereotypy ranged from 0-20 min (mean = 11 min). Stereotypy continued to occur at slightly increased levels during the first week of treatment (range 8-35 min, mean = 19 min), however, from night 11 the total duration of stereotypy reduced with a moderate-sized treatment effect (PBM = 77%), with only three nights where it was observed to occur at reduced levels (duration: 1, 6 and 8 min). In 2018, no stereotypy was evident during baseline or treatment phases.

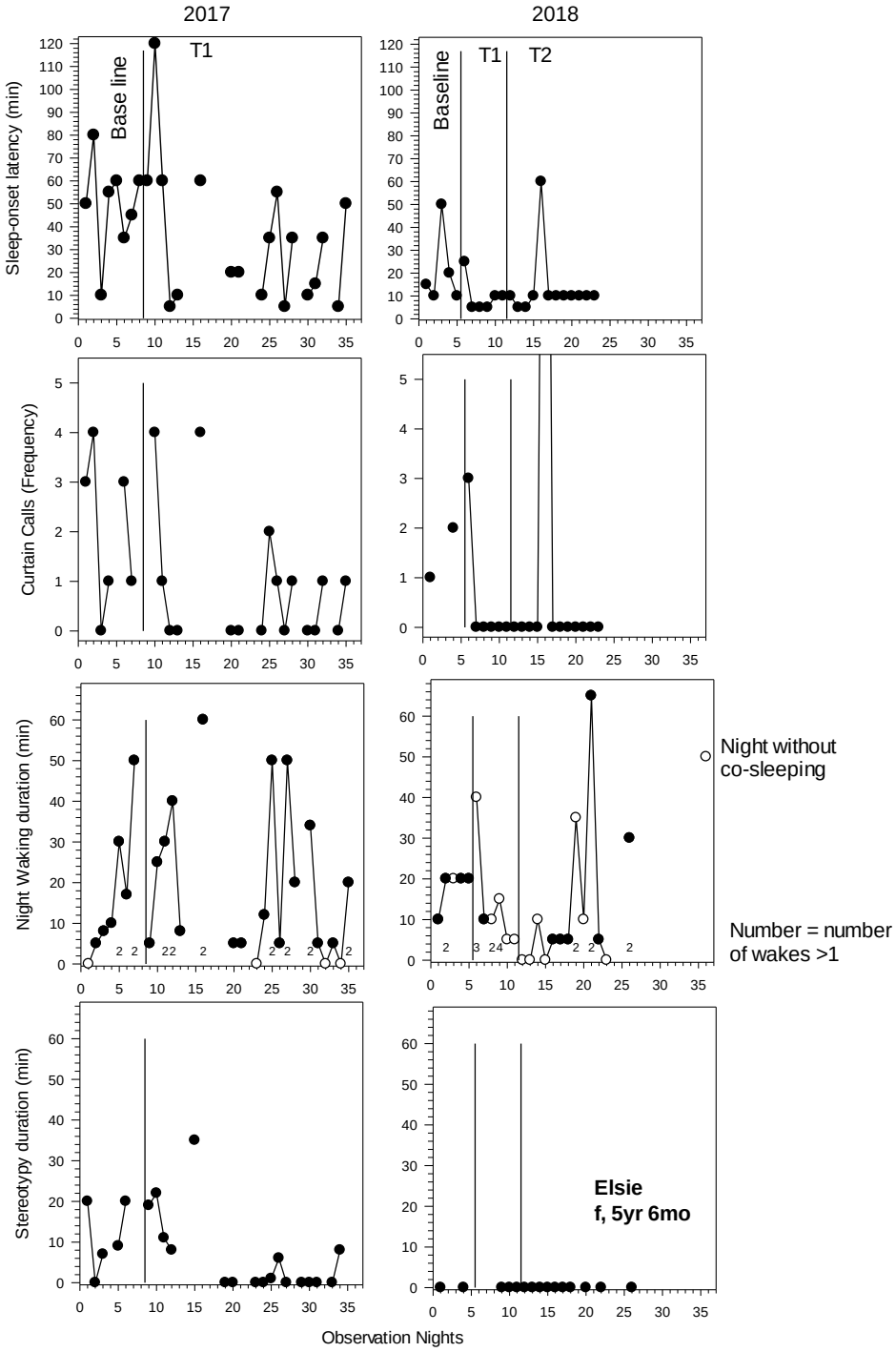
During her 2017 baseline, SOD ranged from 10-80 min (mean = 49 min); during treatment SOD reduced with a moderate-sized treatment effect (PBM = 72%). During her 2018 baseline, SOD was prolonged on one occasion (50 min), with 4/5 nights $\leq$ 20 min. SOD remained reduced during treatment (PBM = 92%) with only two nights > 10 min (duration 25 and 60 min). During her 2017 baseline, the frequency of CCs ranged between 0-4 (median = 2). During treatment, the frequency of CCs reduced with a moderate-sized treatment effect (PBM = 82%), particularly toward the end of treatment (range 0-1). Interpretation of the frequency of CC data in 2018 is limited by baseline data being missing, however, with the exception of two nights (frequency =3 and 10 [note 10 is off scale]) CCs were eliminated during treatment (PBM = 92%).

During baseline in 2017, the frequency of NWs ranged from 0-2 (median = 1) and the duration of NWs ranged from 5-50 min (mean = 20 min). There was no evidence of a treatment effect for the frequency (PBM = 15%) nor the duration (PBM = 50%) of NWs. Unwanted co-sleeping occurred reliably in response to NWs (note if two NWs occurred, co-

sleeping occurred in response to the second NW); co-sleeping was not targeted during intervention in 2017. During baseline in 2018, the frequency of NWs ranged from 1-2 (median = 1) and the duration of NWs ranged from 10-20 min (mean = 18 min). There was no evidence of a treatment effect on the frequency of NWs (PBM = 31%), however, the duration of NWs reduced with a moderate-sized treatment effect (PBM = 77%). During her 2018 baseline unwanted co-sleeping occurred reliably in response to NWs, except for one NW in which co-sleeping did not occur (note, if two NWs occurred, co-sleeping occurred in response to the second NW). Although co-sleeping continued to occur throughout treatment, its frequency appeared to be reduced, as on 8/15 nights (53%) when NWs occurred co-sleeping did not occur, including on nights when multiple NWs occurred.

In sum, in 2017, the combination of the sleep sack, hug pillow and social story appears to have helped to reduce SOD and the frequency of CCs; a further reduction was evident when the contingency of parent attention was removed in 2018. The sleep sack, hug pillow and social story had no effect on the frequency nor duration of NWs in 2017, however, the duration of stereotypy reduced. In 2018, the parental presence procedure followed by planned ignoring appears to have reduced the duration, but not frequency, of NWs. It is noteworthy that co-sleeping continued to occur intermittently and thus the contingency of parent attention during NWs was not fully removed. No stereotypy was observed in 2018.

Figure 7.2.



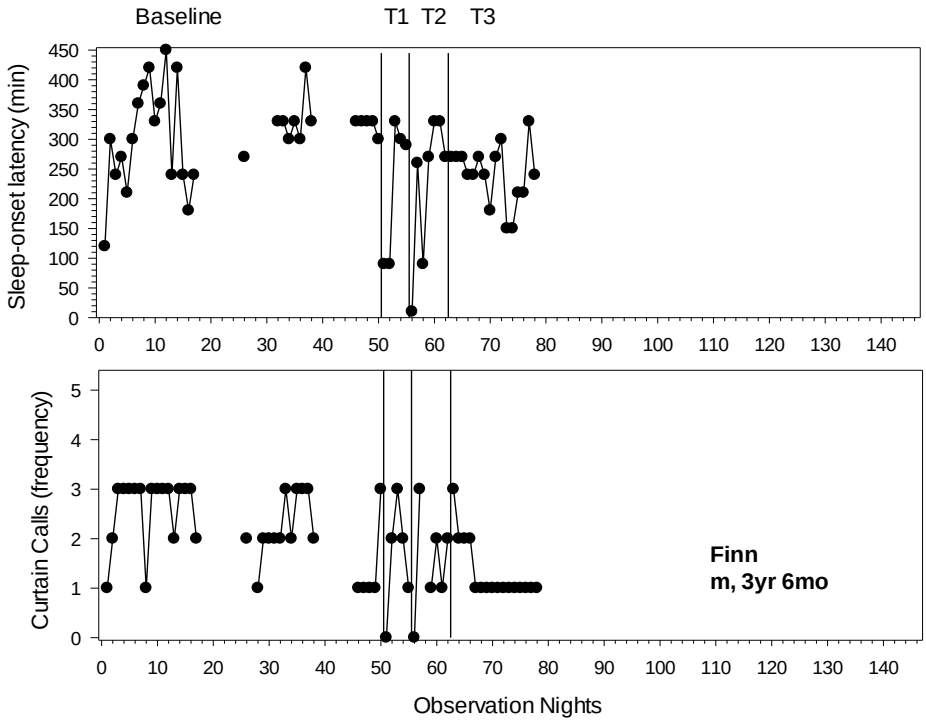
Sleep Outcomes for Elsie: Duration of Sleep Onset Latency, Frequency of Curtain Calls, Duration of Night Wakings and Total Duration of Stereotypy across Baseline and Intervention Phases in 2017 and 2018. The open circles on the graph represent nights without co-sleeping. The numbers 2, 3 and 4 show the number of wakes when greater than one. 2017: sleep sack, hug pillow, social story; 2018: T1 = + parental presence; T2 = + planned ignoring

Finn

The dependent variables for Finn were the duration of SOD and frequency of CCs (see Figure 7.3). Missing data points represent periods where data were not recorded. Stereotypy was also targeted in T1 but there was insufficient data by which to determine treatment effects on stereotypy. During baseline, SOD ranged from 120-450 min (mean = 310 min). SOD reduced with a moderate-sized effect during T1 (PBM = 80%) and T2 (PBM = 71%) and was further reduced with high effectiveness in T3 (range 150-330 min, mean = 240 min, PBM = 94%). During baseline, the frequency of CCs ranged from 1-3 (median = 3); CCs reduced with a moderate-sized treatment effect during T1 (PBM = 80%) and T2 (PBM = 83%) and were further reduced with high effectiveness in T3 (PBM = 94%).

In sum, the use of the trampoline appears to have reduced the longer durations of SOD and the frequency of CCs (i.e., frequency = 0-1 compared to 2-3). The addition of the faded bedtime procedure, altered bedtime routine and finished box, sleep restriction and social story do not appear to have further benefitted his sleep onset, although variability in the frequency of CCs stabilised to occur once/night at the end of intervention. Further improvements were observed, however, following the addition of white noise. It is noteworthy that at the time of the family's withdrawal from the study Finn continued to take approximately four hours on average to fall asleep, and thus his SOD remained a clinically significant problem.

Figure 7.3.



Sleep Outcomes for Finn: Duration of Sleep Onset Latency and Frequency of Curtain Calls across Baseline and Intervention Phases. T1 = pre-bedtime satiation (trampoline); T2 = + faded bedtime, pre-bedtime satiation (food, parent attention, access to preferred items), finished box and reward, sleep restriction, social story; T3 = + white noise

Table 7.3. *Percentage below the Median Scores for James, Elsie and Finn*

	DV	Treatment PBM				STFU PBM	LTFU PBM
James	SOD	**T1**	**T2**	**T3**	**T4**		
		15%	14%	20%	70%	43%	0%
Elsie		**2017**		**2018**			
	SOD	72%		92%		-	-
	CCs	82%		92%		-	-
	Freq NWs	15%		31%		-	-
	Dur NWs	50%		77%		-	-
	Stereotypy	77%		-		-	-
Finn		**T1**	**T2**	**T3**			
	SOD	80%	71%	94%		-	-
	CCs	80%	83%	94%		-	-

High effectiveness (>90%)	Moderate effectiveness (70-90%)	Ineffective (<70%)

Note. CCs: curtain calls; Dur: duration; DV: dependent variable; Freq: frequency; LTFU: long-term follow-up; NWs: night-wakings; PBM: percentage below the median; SOD: sleep onset delay; STFU: short-term follow-up; T: treatment phase

Children's Sleep Habits Questionnaire Scores

Following treatment, Elsie's score reduced from 64 to 46, whilst James' score remained the same (45). Finn's score increased from 45 to 50 for pre- and post-treatment, respectively. None of the children's scores fell below the clinical cut-off (41).

Interobserver Agreement

For James, IOA was collected for 20%, 24% and 29% of the baseline, treatment and STFU phases respectively, and was 83%, 87% and 100%, respectively. For Elsie (2018), IOA was collected for 40% and 24% of the baseline and treatment phases respectively and was respectively 80% and 93%. IOA was unable to be calculated for her 2017 data because there was insufficient parent-reported data to compare with VSG. IOA was not able to be calculated for Finn because of insufficient VSG data for comparison.

Treatment Fidelity

Treatment fidelity during the intervention phase was calculated for 30% of sessions for James, Elsie, and Finn, and was respectively 92%, 48% and 56%. Treatment fidelity during follow-up was calculated for 40% of sessions for James and was 81% (STFU) and 87% (LTFU). Treatment fidelity scores could not be calculated for follow-up for Elsie or Finn because the families did not complete this phase. The overall score for James (91%) suggested that his parents implemented intervention with high fidelity across phases. Fidelity scores for Elsie (48%) and Finn (56%) were lower, suggesting that their parents may have found it difficult to adhere to the behavioural programme during the intervention phase.

Social Validity

Post-Treatment Interview

James' parents reported high satisfaction with the outcomes of treatment and with the treatment process overall. Specifically, they were pleased with the pace of intervention such

that they felt it was tailored to their goals and capacity for change. According to James' parents, "small changes" resulting from multiple strategies, particularly the social story and white noise, effectively improved his sleep. Although James continued to vocalise, they felt he did so more quietly and less frequently when white noise was used. They reported that he tended to wake in the morning in a better mood and reported that maternal sleep quality improved when James began sleeping independently. They did not report any negative side effects of intervention.

Post-treatment interviews were not held for Elsie nor Finn; however, a brief discussion was held with Finn's parents regarding their involvement in the study. His parents reported that he enjoyed the trampoline, and that he lay more quietly with less bouncing and squealing in bed following its use. Further, they felt that the structure of the altered bedtime routine, including packing away items that he previously took to bed, reduced his disruptive (CC) behaviour, and created a calmer family environment. Finn's parents commented that although they were motivated to improve Finn's sleep, the requirements for on-going data collection and adherence to treatment strategies in the context of other family priorities meant that continued involvement in the study was not sustainable.

The Treatment Acceptability Rating Form - Revised

TARF-R scores for James' parents are presented in Table 7.4. Overall, James' parents indicated a high level of treatment acceptability (114 and 117/119). James' parents perceived his sleep problems to be severe. They indicated that they found treatment to be effective and reasonable, with an absence of negative side effects, including minimal disruption and time. Data were not available for the other two families because they did not complete the final phases of the study.

Table 7.4. *Treatment Acceptability Rating Form - Revised Scores*

Scale	Parent 1	Parent 2	Maximum score
Effectiveness	19	19	21
Reasonableness	21	21	21
Willingness	19	21	21
Cost	14	14	14
Negative side-effects	21	21	21
Disruption/time	20	21	21
Problem severity*	12	14	14
Understanding of treatment*	7	6	7
Total acceptability	**114**	**117**	**119**

Note. *Not included in total acceptability score

Discussion

This study evaluated the effects of function-based behavioural interventions on sleep-related stereotypy and sleep problems in three children on the autism spectrum. Stereotypy was targeted in T1 for all three children, with data available for two children. Results showed that the duration of James' vocal stereotypy remained unchanged during intervention but was eliminated along with NWs during follow-up. The duration of Elsie's stereotypy reduced following treatment in 2017 and was absent during baseline and intervention in 2018. The reason for its absence in 2018 is unclear; it is possible that improvements occurred because of continued use of the sleep sack and hug pillow over this time, and/or that change occurred naturally (e.g., as she matured) or in response to extraneous variables outside of treatment.

The preliminary results of this study also showed that SOD reduced in all three children, and that CCs reduced for the two children for whom these were a problem. This included a reduction in SOD and CCs in T1 for Finn, during which his stereotypy was targeted without his sleep problems being targeted per se. NWs reduced in frequency but not duration for James, whilst for Elsie, NWs reduced in duration but not frequency. The maintenance of treatment effects and parent perceptions of treatment were examined in one family; except for

SOD in LTFU, treatment effects were largely maintained, and parent acceptability was high. Results must be interpreted with caution however, given that there were methodological problems (e.g., missing data) and participant attrition.

A central aim of this study was to investigate whether the duration of SOD and NWs decreased if sleep-related stereotypy was reduced. Vocal stereotypy for James and Finn was targeted with white noise (matched stimulation). For Finn, white noise appears to have improved SOD and occurrence of CCs, although the effects on his vocal stereotypy were not able to be determined. For James, white noise alone had no effect on his vocal stereotypy nor other sleep variables. This is unsurprising given the case conceptualisation indicated that his sleep problems were multiply determined, and therefore required multiple function-based treatment components to address the maintaining factors. However, given the evidence that matched stimulation can effectively reduce vocal stereotypy in individuals on the autism spectrum (D. Wang et al., 2020; DiGennaro Reed et al., 2012; Lanovaz et al., 2011; Saylor et al., 2012), the lack of effect on stereotypy is more surprising. There are several reasons why white noise may have been ineffective, including that satiation did not occur, or that vocal stereotypy was reinforced by consequences other than, or in addition to, auditory stimulation (e.g., the tactile sensation of throat vibrations, or the content of vocalisations relating to an enjoyable memory).

This finding underscores the challenges in treating behaviour for which the specific reinforcers may be impossible to identify and manipulate (Shore et al., 1997). Further, studies suggest that preferred forms of matched stimulation are more effective than non-preferred stimulation (D. Wang et al., 2020; Lanovaz et al., 2012). For example, Saylor et al. (2012) compared the effects of three types of noncontingent auditory stimulation (white noise, music and recordings of children's own vocal stereotypy) on vocal stereotypy in two children on the autism spectrum. Results showed that music, which was the most preferred treatment selected

by both children, reduced vocalisations most effectively for each child. Future research could help to determine whether matched but also preferred auditory stimulation may help to decrease sleep-related stereotypy in children on the autism spectrum. In line with the findings of Chapter 4, white noise appears to have been well tolerated by children in this study and was perceived by James' parents as beneficial (e.g., they reported that it reduced the volume of his vocalisations).

Finn and Elsie both engaged in motor stereotypy. For Finn, bouncing on a trampoline prior to bed (i.e., an AO) was used to reduce motor stereotypy. Although the impact of the trampoline on his motor stereotypy was not able to be determined, SOD and CCs reduced following its introduction. It is possible, but unable to be verified, that bouncing on the trampoline reduced his motor stereotypy, enabling him to fall asleep more efficiently (in line with anecdotal parent report that he lay more calmly in bed following its use). It is also possible that physical exercise using the trampoline functioned as an AO for stereotypy (Rapp & Lanovaz, 2016). Alternatively, his sleep may have been improved via other mechanisms; for example, physical exertion from bouncing on the trampoline may have increased physiological sleep pressure (and thus motivation to fall asleep) without directly affecting stereotypy.

Elsie's stereotypy was targeted with a sleep sack in attempt to restrict her motor movements in bed; these items appeared to reduce the duration of stereotypy (and SOD and CCs), whilst the frequency and duration of NWs remained unchanged. This suggests that stereotypy may not have had a sleep-interfering effect during NWs. The relationship between stereotypy and initial sleep onset for Elsie is less clear, for example, whether the sleep sack helped to reduce sleep-interfering (stereotypic and CC) behaviour and thus enabled a more efficient sleep onset, or whether SOD reduced and in return removed the opportunity for her to perform sleep-interfering behaviour. It is also possible that improved sleep onset enabled

Elsie to experience falling asleep without stereotypy, which may have helped to reduce the duration of her rocking during NWs (e.g., via improved stimulus control for behavioural quietude).

It is noteworthy that for James, as well as Eddy, Tessa, and Bella in Study 2 (Chapter 6), stereotypy was reduced (or eliminated) commensurate with the reduction (or elimination) of sleep problems; thus, treating sleep problems may be an effective means by which to indirectly treat sleep-related stereotypy (i.e., by removing opportunity for children to perform stereotypic behaviour). This point is considered more thoroughly in Chapter 11.

Overall, the impact that stereotypy had on sleep for Finn and James was unable to be determined following treatment. It is probable that the relationship between sleep problems and stereotypy is reciprocal; for example, for James, NWs precipitated his vocal stereotypy (i.e., provided opportunity for him to perform the behaviour), and in return, vocal stereotypy may have perpetuated his NWs. For Elsie, the finding that NWs did not change when the duration of stereotypy reduced raises the possibility that stereotypy may have served other functions in relation to sleep for her. For example, as she typically rocked until sleep onset occurred, it is possible she used repetitive rocking to self-settle. It is noteworthy that Finn also engaged in body rolling immediately prior to sleep onset, which differed in topography from bouncing and squealing and potentially may have differed in function. Thus, stereotypy may differentially affect sleep across and within children; this specific line of inquiry is investigated in the next chapter.

Results tentatively showed improvement in sleep across children in this study. For Elsie and Finn, improvements in SOD and CCs (and the duration of stereotypy for Elsie) were observed following the use of a sleep sack, hug pillow and social story (Elsie) and trampoline (Finn), without the use of extinction procedures. It is important to note, however, that IOA for Finn was unable to be calculated, and fidelity was generally low; thus, improvements in

sleep may not be directly related to treatment. Importantly, for Elsie and James, further improvement was observed when parent attention to problem behaviour was removed (i.e., modified extinction, in addition to antecedent-based procedures), supporting the hypothesis that sleep problems were (at least partially) socially maintained, in this case underscoring the need for multimodal interventions that targeted both antecedent and consequence variables.

Sequential implementation of treatment strategies warrants further investigation in future research because it may be a feasible way to reduce parent burden by using only what is necessary to change behaviour (van Deurs et al., 2021). This is an important consideration, given that some parents in this study found adherence to a behavioural programme and the requirements of on-going data collection arduous (in the context of other priorities). The length of data collection (when prolonged) may also affect fidelity in the long-term, as in Finn's case his extended baseline may have affected his parents' ability to sustain data collection during treatment (e.g., by reaching a capacity limit). Of note, some families in this study experienced several complexities (e.g., varying medical/developmental diagnoses across family members) that may have affected their ability to complete treatment. This underscores the importance of assessing for the presence of complexities and stressors on families during comprehensive assessment and supporting parents to prioritise family needs. A family's priorities may also change during a behavioural sleep programme, given the length of time (e.g., months) that families may be provided with individualised support.

This study included only three case presentations, which limits the extent of replication and generalisability of results. There were also limitations with how stereotypy was recorded across children. For James, vocal stereotypy was recorded via parent-report; while IOA for other behaviour was high (range 83%-100% across phases), IOA was not able to be calculated for vocal stereotypy owing to the inability of the available VSG to capture audio recordings. For Finn and Elsie, stereotypy was recorded with VSG (owing to parents being

unable to report on the behaviour because they were in a different room). Sole reliance on VSG recordings was problematic, however, because in Finn's case a combination of technical failures and his parents later choosing not to record video meant there was insufficient data to determine treatment effects on stereotypy. In addition, IOA for Finn was not able to be calculated. This underscores the importance of using a range (e.g., parent-report and instrumental measures [e.g., VSG]) of measures to assess sleep outcomes, as well as the importance of maintaining close contact with families to overcome challenges relating to data collection as they arise.

Chapter 8: Study 4

Parent Perceptions of Sleep-Related Stereotypy within Sleep Problems in Children on the Autism Spectrum: Implications for Behavioural Treatment

This chapter presents Study 4, which examines parent perceptions of stereotypy in relation to sleep problems in children on the autism spectrum. This qualitative study used thematic analysis to analyse clinical assessment reports obtained from 21 parents of 15 children on the autism spectrum, inclusive of participant children in Studies 1-3. This study aimed to improve our understanding of the types of sleep-related stereotypy exhibited by children on the autism spectrum, as well as how parents manage such behaviour. In addition, this study explored parent perspectives of the ways in which children's stereotypy potentially affected their sleep, with the central aim of better understanding the role of stereotypy in relation to sleep problems.

A qualitative framework was chosen for this study because it was the most suitable method through which to reveal complexities of stereotypic behaviour, which could not be captured through a quantitative framework alone. Qualitative methods provide a means of exploring the phenomenon of sleep-related stereotypy, including characteristics of and parent experiences with children's behaviour, which can provide valuable insight into the complex nature of stereotypy in relation to sleep disturbance in children on the autism spectrum (Belotto, 2018; Kirkpatrick, Gilroy, et al., 2019). Thereby, this chapter aimed to extend and complement the quantitative findings of Studies 1-3 by providing a qualitative perspective of the nature of sleep-related stereotypy in children on the autism spectrum.

This chapter consists of a manuscript that has been published in the journal *Advances in Neurodevelopmental Disorders* with myself as first author[9]. James' case study (Chapter 7,

[9] Hunter, J. E., McLay, L. K., France, K. G., Swit, C. S., & Blampied, N. M. (2022). Parent perceptions of sleep-related stereotypy within sleep problems in children on the autism spectrum: Implications for behavioral treatment. *Advances in Neurodevelopmental Disorders*, https://doi.org/10.1007/s41252-022-00246-w

Study 3) was included in the full manuscript because his information helped to expand on the outcomes of the thematic analysis, by further illustrating the potential role of vocal stereotypy within sleep difficulties in a child on the autism spectrum, including examination of the effects of targeted treatment on vocal stereotypy. His case information has been removed from this chapter (including the Discussion) to reduce duplication of previously presented information.

Introduction

Sleep problems in children on the autism spectrum are ubiquitous, affecting up to 80% of children across all cognitive levels (Abel et al., 2018; S. Cohen, Conduit, et al., 2014). The most frequently reported sleep difficulties are associated with sleep onset and maintenance, and include SOD, frequent and prolonged NWs, poor sleep efficiency and reduced TST (Cortesi et al., 2010; Johnson et al., 2018; Singh & Zimmerman, 2015). These problems are often accompanied by bedtime resistance, daytime sleepiness, disruptive behaviours (e.g., calling out to parents and caregivers [hereafter referred to as parents], screaming or crying, playing with toys or objects) and unwanted co-sleeping (Cortesi et al., 2010; Kirkpatrick, Gilroy, et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021; Richdale & Schreck, 2009). Sleep problems in children on the autism spectrum do not tend to remit without treatment, often persisting throughout adolescence (Goldman et al., 2012; Mazurek et al., 2019; Sivertsen et al., 2012).

Inadequate sleep has significant detrimental effects on children's daytime functioning and wellbeing, including increased internalising and externalising behavior (e.g., anxiety, aggression) and reduced cognitive and adaptive functioning, and impaired physical health (e.g., obesity, epilepsy; Accardo & Malow, 2015; Bangerter et al., 2020; Delahaye et al., 2014; M. Taylor et al., 2012; S. Cohen et al., 2018; Zuckerman et al., 2014). Furthermore, sleep disturbance may exacerbate autism symptom severity, with higher rates of stereotypy

and social impairment found in children on the autism spectrum who are also poor sleepers (Abel et al., 2018; Gabriels et al., 2005; H. Adams, Matson, Cervantes, et al., 2014; Hundley et al., 2016; Park et al., 2012; Schreck et al., 2004; Veatch et al., 2017). Parents of children with sleep problems are also at heightened risk of sleep deprivation, resulting in elevated levels of stress, anxiety, depression, fatigue, relationship discord and familial dysfunction (Chu & Richdale, 2009; Johnson et al., 2018; Levin & Scher, 2016; Martin et al., 2019; Meltzer, 2011). Thus, there are many reasons to seek timely and effective treatment of sleep problems to remediate the widespread effects of sleep deprivation (Goldman et al., 2012; Singh & Zimmerman, 2015).

The aetiology of sleep disturbance in autism is unknown, however it is widely agreed that the causes are multifactorial, involving biological, psychological, behavioural, social, medical, and environmental factors, either alone or in combination (Cortesi et al., 2010; Mazzone et al., 2018; Richdale & Schreck, 2009; S. Cohen, Conduit, et al., 2014). Inclusive within these contributing factors, sleep problems often have a behavioural basis. Sleep is a bio-behavioural state that reinforces the behaviour of falling asleep. Falling asleep can be understood as the end point of an operant chain, which begins with bed-preparatory behaviours (e.g., putting on pyjamas, brushing teeth) and ends with behavioural quietude (i.e., lying quietly in bed, with low levels of behavioral, cognitive, and emotional arousal; Blampied & Bootzin, 2013; Blampied & France, 1993). Behavioural quietude must be maintained for a sufficient period in order for the wake-sleep transition to occur (Blampied, 2013a; Blampied & France, 1993).

Sleep problems can develop and be maintained when children engage in problem behaviour (e.g., calling out, leaving the bed, stereotypy) that disrupts the behavioural chain of falling asleep (Blampied, 2013a). Parent responses (e.g., providing attention) to child behaviour may also inadvertently reinforce these behaviours (Beresford et al., 2016;

Blampied, 2013a; Blampied & France, 1993; Richdale & Schreck, 2009). Further, the core characteristics of autism may interfere with the behavioural chain of falling asleep, including that children may experience difficulty transitioning between tasks and interpreting social expectations related to going to bed and to sleep (Mazzone et al., 2018; Reynolds & Malow, 2011; Richdale & Schreck, 2009).

Extant research suggests that stereotypy (e.g., repetitive vocalisations or motor movements) can accompany sleep problems (e.g., SOD, NWs) in children on the autism spectrum (Jin et al., 2013; Hunter et al., 2021; Malow, Marzec, et al., 2006; McLay, France, Blampied, et al., 2019; Richdale & Schreck, 2009; Weiskop et al., 2005). For example, in a recent case analysis by McLay, France, Blampied, van Deurs, et al. (2021) of 41 children and adolescents on the autism spectrum with sleep problems, 27% of participants exhibited stereotypy in the sleep context. Stereotypic behaviours in children on the autism spectrum are heterogeneous, but can broadly be classified as either motor stereotypy, consisting of motor movements (e.g., hand-flapping, body-rocking), RMO (e.g., mouthing or spinning objects), or vocal stereotypy, involving non-contextual vocalisations (i.e., without a clear communicative purpose, e.g., recurring words or non-word sounds; Ahearn et al., 2007; Akers et al., 2020; Cunningham & Schreibman, 2008; D. Wang et al., 2020; DiGennaro Reed et al., 2012). All forms of stereotypy share a response topography of repetition, invariance and atypicality (Didden et al., 2012; Rapp & Vollmer 2005).

Stereotypy is operant behaviour that frequently functions to produce automatic reinforcement; that is, the behaviour is intrinsically reinforced via non-social sensory consequences (Ahearn et al., 2007; DiGennaro Reed et al., 2012; Rapp & Lanovaz, 2016; Rapp & Vollmer, 2005). There is also evidence to suggest that stereotypy can be socially-mediated, through contingencies of positive (e.g., social attention) and negative (e.g., escape from demands) reinforcement (Durand & Carr, 1987; Kennedy et al., 2000; Roantree &

Kennedy, 2006; Wilke et al., 2012). Without intervention, stereotypy tends to persist throughout the lifespan, and may increase in frequency and intensity over time (Akers et al., 2020; Bodfish et al., 2000; D. Wang et al., 2020).

Stereotypy can be problematic when the intensity of and/or persistence of the behaviour impedes an individual's adaptive functioning; for example, stereotypy can significantly interfere with socialisation and learning in children on the autism spectrum (Akers et al., 2020; D. Wang et al., 2020; Lydon et al., 2016). Stereotypy can also disrupt family functioning and is associated with severe parental stress (Boyd et al., 2012). As such, stereotypy is a common target for intervention, particularly in the educational setting (Akers et al., 2020; D. Wang et al., 2020). There is also evidence to suggest that stereotypy may serve as a coping mechanism for individuals on the autism spectrum, possibly helping to alleviate distress and to regulate thoughts and feelings (Gabriels et al., 2005; Gabriels et al., 2013; McCarty & Brumback, 2021). Individuals on the autism spectrum may report experiencing stereotypy as comforting, calming and enjoyable (McCarty & Brumback, 2021).

In comparison to the literature on daytime stereotypy, very little is known about sleep-related stereotypy (Hunter et al., 2021). The reasons why children on the autism spectrum engage in stereotypy in the sleep context are currently unclear and may be multifaceted across and within individuals (Cunningham & Schreibman, 2008; Kennedy et al., 2000). For example, automatic reinforcement generated through stereotypy may be positive (i.e., producing a sensory consequence) and/or negative (i.e., reducing or eliminating a sensory experience, such as physical discomfort or pain [an instance of escape/avoidance]; Leader & Mannion, 2016). It is possible that sleep-related stereotypy may serve a self-settling purpose, such as rhythmic movements (RM; e.g., body-rocking, head-banging) commonly observed in infants and young children during the sleep onset period (Haywood & Hill, 2012; Hoban, 2003). As for RM and daytime stereotypy, however, sleep-related stereotypy may be

considered problematic when persisting in the behaviour incurs a clinical consequence including interference with adaptive functioning (Didden et al., 2012; Gwyther et al., 2017). Specifically, in the sleep context, after a child is bid goodnight, stereotypy may become sleep-interfering if it inhibits a child's ability to establish and maintain the behavioural quietude necessary to initiate and/or re-initiate sleep (i.e., if the child wakes and engages in stereotypy; Jin et al., 2013; Hunter et al., 2021; McLay, France, Blampied, et al., 2019).

The relationship between autism and sleep problems is understood to be complex and cyclical; for example, greater autism symptom severity may predict greater severity of sleep problems, and vice versa (H. Adams, Matson, Cervantes, et al., 2014; Hollway & Aman, 2011; Hundley et al., 2016; S. Cohen, Conduit, et al., 2014; Schreck et al., 2004). However, the mechanisms of this relationship are currently unclear; most of the research examining this relationship has been cross-sectional and correlational in design, thereby limiting our understanding of causality and directionality (H. Adams, Matson, Cervantes, et al., 2014; Schreck & Richdale, 2020). This includes the impact that stereotypy may have on sleep in children on the autism spectrum, which is not well understood (Hunter et al., 2021). The nature of this relationship requires further investigation, including whether stereotypy primarily interferes with sleep onset, whether it may serve a self-settling function for some children, or whether in fact it may have no impact on sleep (i.e., occur simply by virtue of the child being awake).

Further, sleep-related stereotypic behaviours are not well characterised, including the patterns of behaviour specific to individuals (Gabriels et al., 2005). For example, it is not clear whether stereotypy differs in topography or function from that which children engage in throughout the day. Better understanding of the types of stereotypy that children on the autism spectrum exhibit at night, and how these behaviours may affect sleep, can provide an avenue for targeted treatment that can generate important primary and secondary benefits for

children and parents (Boyd et al., 2012; Delahaye et al., 2014; Hunter et al., 2020; Hundley et al., 2016; Mazurek et al., 2019; McLay, France, Blampied, Hunter, et al., 2021; S. Cohen, Conduit, et al., 2014).

Typically, it is parents who can detect and respond to children's sleep problems and stereotypy. Thus, parents can provide valuable insight into stereotypy that occurs in the context of sleep, particularly where self-report is not possible (e.g., owing to younger age and/or limited verbal ability). The present study involved qualitative analysis of semi-structured clinical interviews with the parents of children on the autism spectrum who were assessed for a behavioural sleep intervention. This study examined parent descriptions of stereotypy in relation to sleep disturbance, including types and topography of stereotypic behaviour, occurrence within sleep problems, the impact of stereotypy on sleep onset, and parent response to stereotypic behaviour. Parent reported information was then examined with the aim of enhancing our understanding of: (a) the types and topography of stereotypy exhibited by children on the autism spectrum at night; (b) the potential role of stereotypy within sleep difficulties (e.g., whether stereotypy interferes with sleep); and (c) how parents manage stereotypy in the sleep context.

Methods

Participants

The participants were parents of a child or adolescent (henceforth referred to as 'child') on the autism spectrum, who was referred or self-referred to a specialist clinic to receive behavioural sleep intervention services. Children who received services were part of a wider study designed to investigate the effectiveness of function-based, parent-implemented interventions for children on the autism spectrum. To be eligible to receive services, children met the following inclusion criteria: (a) aged between 3-17 years; (b) a formal diagnosis of

autism (APA, 2013) confirmed by a pediatrician; (c) parent reported sleep problems including SOD, NWs, bedtime resistance, and/or unwanted co-sleeping, later confirmed by formal assessment measures (e.g., the CSHQ and analysis of VSG). CSHQ total scores were available for 10 children in this study and are included as Supplementary Information (S1; see Appendix K); 8/10 children exceeded the clinical cut-off score (41). This study reports findings from a subset of children who engaged in at least one form of stereotypy (e.g., repetitive vocalisations) during sleep onset and/or NWs. Children were excluded from the study if there was contraindication for a behavioural intervention (e.g., if they had a medical condition that affected their sleep).

Parent participants included 21 parents of 15 children on the autism spectrum (see child details below). In six families both parents participated in the interview, in five families, one parent from a two-parent household participated in the interview, and four children had a solo parent. Parent's ethnicity included European (*n* = 14), NZ European/Māori (*n* = 4), Asian (*n* = 2) and South African (*n* =1). Two parents in the same household spoke English as a second language. In 11 families one or both parents were employed, and in four families the solo caregiver was unemployed or retired.

Child participants were 11 boys and four girls, aged between 3-15 years (median = 5 years). Only one participant was an adolescent (15 years), all other children were $\leq$ 8 years, and 11 of the 15 children were $\leq$ 6 years. Co-occurring conditions, as diagnosed by a pediatrician (*n* = 5) included RGND (e.g., chromosomal deletions; *n* = 4), ID (*n* = 4), global developmental delay (*n* = 1) and ADHD (*n* = 1). Verbal ability ranged from non-verbal (*n* = 3), language-delayed (i.e., below chronological age, e.g., limited to single words/short sentences; *n* = 11), and no language difficulties (*n* = 2). Four children took melatonin at the time of the interview, the remainder were unmedicated. The types of child sleep problems reported were NWs (*n* = 11, 73%), SOD (*n* = 10, 67%), unwanted co-sleeping (*n* = 7, 47%)

and bedtime resistance (n = 2, 13%). Eleven children (73%) were reported to have two or more sleep problems.

Procedure

Settings and Personnel

A clinical interview was conducted by the first author (12 families), or another research clinician who was an intern psychologist (three families) as part of a comprehensive intake assessment. The first author, a trained psychologist and doctoral student, had a background in sleep and autism research, and was experienced in clinical interviewing. The interview was conducted either in person or via videoconferencing and was a pre-cursor to the development of an individualised, parent-implemented, behavioural sleep intervention.

Semi-Structured Interviews

Following a screening interview and informed consent processes, parents participated in a 1-1.5 hour-long, open-ended, semi-structured interview. The purpose of the intake interview was to comprehensively assess each child's sleep problems, in the context of broader child and family factors. The interview began with a discussion of the child's presenting sleep problems, including types of sleep problems (e.g., SOD, NWs), typical bedtime routine (e.g., time put to bed, steps in the sequence leading up to bedtime), history of sleep disturbance (e.g., onset of the sleep problem, chronicity) and impact (i.e., on child, parents, wider family) of sleep disturbance. Parents were also asked to describe children's behaviour after they were bid goodnight, including stereotypy. Parents were asked to describe the topography of their child's night-and day-time stereotypy, setting (e.g., physical location, timing within sleep problems), consequences of the stereotypic behaviour (e.g., parent response), their perception of the impact of their response on stereotypy, and parent's view of the role of their child's stereotypy in relation to sleep (e.g., what impact, if any, stereotypy

had on sleep onset). A list of the questions asked about stereotypy are included as Supplementary Information (S2; see Appendix L). If a parent reported more than one type of night-time stereotypic behaviour, then questions were asked in relation to each type (e.g., what impact [if any] each of the behaviours had on sleep). Parents also spoke about stereotypy in response to questions non-specific to stereotypy (e.g., "tell me what your child does when they wake during the night?"). A full developmental history and relevant child (e.g., interests, schooling) and family information (e.g., living situation including sleeping arrangements, employment, parental mental health and wellbeing, sources of support) was also obtained. Finally, parents were asked about their goals for their child's sleep.

Data Analysis

This research was conducted within a theoretical thematic analysis framework (Braun & Clarke, 2006), driven by the researcher's theoretical and analytical interest in stereotypy in relation to sleep disturbance (i.e., the first author focussed on the feature of stereotypy when coding the data). Themes were identified through a semantic approach, involving a progression from the organisation of parent description to interpretation (Braun & Clarke, 2006). This framework enabled exploration of the phenomenon of stereotypy in relation to sleep disturbance in children on the autism spectrum; drawing on parents' perspectives of their child's behaviour via an interpretive paradigm enabled unique insight into the characteristics of, and parent experiences with, sleep-related stereotypy (Belotto, 2018).

Clinical assessment reports for each child (n = 15) were analysed by the first author, under the supervision of the second and fourth author, following the guidelines of Braun and Clarke (2006). Thematic analysis involved initially reading and re-reading the reports to ensure familiarisation with the parent accounts. All information pertaining to stereotypy was then manually coded, to generate an initial list of codes, and verbatim statements were extracted. Next, the initial codes were organised into broader themes and sub-themes and

reviewed for relevance to the research questions. The themes and codes were then refined until definitive codes and themes were established (Braun & Clarke, 2006). The second and fourth author checked the definitive codes and themes for consistency and coherence.

Reliability

Interrater reliability was assessed for the thematic analysis (Elliot, 2018; O'Connor & Joffe, 2020). A research assistant, who was part of the wider sleep research team with experience conducting sleep treatment research, independently coded the reported information for eight (53%) randomly selected, de-identified participants. Interrater reliability was calculated using the formula [Agreement/ (Agreement + Disagreement)] x 100%. Data pertaining to stereotypy were recorded as an agreement if the same definitive code was applied by both the first author and the research assistant. First, the research assistant independently coded the report information for two participants (i.e., to establish satisfactory reliability); the research assistant and first author compared the two sets of coded information and calculated the level of agreement (75% and 80%; Belotto, 2018; O'Connor & Joffe, 2020). Instances of disagreement were discussed and resolved using a process of negotiated agreement; in this case, all disagreements were reconciled through one person deferring to the other, and 100% agreement was reached across both participants (Belotto, 2018; Elliot, 2018).

This process was then repeated for the other six participants; initial agreement was 90%, 79%, 71%, 60%, 78% and 75%. Following discussion, the wording of three codes was amended to resolve ambiguity (e.g., whether stereotypy was a 'predominant' behaviour) and improved consensus in interpretation (Elliot, 2018); these codes were then re-applied across all data sets to ensure consistency, and 100% agreement was reached. A full list of definitive codes and themes is available from the corresponding author upon request.

Results

The findings of the qualitative analysis of the 15 interview reports are presented below. Five themes including: (1) type and topography of stereotypy; (2) timing of stereotypy; (3) stereotypy as sleep-interfering; (4) stereotypy as sleep-conducive; and (5) parent responses to stereotypy, were identified. Two sub-themes, (a) co-sleeping; and (b) escalation, were identified within the 'parent responses to stereotypy' and 'stereotypy as sleep-interfering' themes, respectively.

Types and Topography of Stereotypy

Parents described a diverse range of stereotypic behaviours exhibited by children on the autism spectrum, in the sleep context. Akin to daytime stereotypy, night-time stereotypy could be grouped into motor behaviours, vocal behaviours, or RMO. Vocal stereotypy (*n* = 11 children, 73%) encompassed non-word sounds (i.e., throat clearing, humming, croaking, groaning, "babbling", non-contextual "belly/hysterical" laughing and squealing), repetition of words (i.e., numbers and letters), and scripting (i.e., television phrases, song lyrics). Many parents described their child's vocalisations as "talking to him/herself". Vocalisations were often related to children's preferences (e.g., *Thomas the Tank Engine*: "you're a very useful engine"). Motor behaviours (n = 10, 67%) included repetitive bouncing (on bottom), hand-flapping, body-rocking (on stomach or standing), flicking legs, twiddling fingers, head-banging, and body-rolling (side to side). One parent described their child's stereotypy as "hip thrashing" on his stomach, while another parent reported "she rocks before she falls asleep, and sucks her thumb, so rocks in a way that doesn't interfere with her thumb-sucking…sort of pushes off the bed with the other arm". RMO (n = 4, 27%) was the least common form, and included lining up toys and spinning toy car wheels, twiddling items (e.g., a pen), sniffing a scarf, and manipulating magazine pages: "she will fiddle with pages, and sort of nibble the centre". Frequently, parents reported that children (n = 8, 53%) engaged in

multiple forms of stereotypy, with vocal and motor stereotypy being the most common combination (e.g., simultaneous bouncing and squealing).

Timing of Stereotypy

The timing of when children's stereotypy occurred was identified as a theme. Six children (40%) engaged in stereotypy during both sleep onset and NWs. Four children (27%) engaged in stereotypy during sleep onset only and NWs were not a parent-reported problem. Five children (33%) exhibited stereotypy during NWs only; in three of these children sleep onset was managed with melatonin, and for the other two sleep onset was not a parent-reported problem. Overall, parents were aware of children engaging in stereotypy both at the start of and/or during the night if the child woke.

Stereotypy as Sleep-Interfering

Many parents indicated that stereotypic behaviours resulted in the child having difficulty initiating and/or re-initiating sleep. For nine children (60%), stereotypic behaviours were reported to be sleep-interfering; eight of these children (89%) engaged in vocal stereotypy, accompanied in some cases by motor stereotypy (n = 3) and RMO (n = 1). The ninth child engaged in RMO; it was unclear to her parents if she also vocalised. Sleep-interfering stereotypy was often referred to as "play", with parents indicating the child was in a stimulated state (e.g., "wired", "vocally active" or "bouncing off the walls") and became more wide-awake (e.g., "winding/ramping up") when engaging in stereotypy.

Interestingly, a sub-theme was identified, where vocalisations escalated. Specifically, in three children vocalisations were reported to begin quietly and infrequently, but then to intensify as time went on, particularly in volume. According to one parent "he starts by whispering numbers and letters to himself". The child would then "get louder, and start shouting, and get more excited". In two children when vocalisations escalated, motor

stereotypy also occurred (e.g., the child began bouncing, whilst shouting/laughing). Parents termed nights when children vocalised as "bad" or "unlucky" nights, and vocal stereotypy was strongly viewed as sleep-competing; "the more he babbles, the harder it is for him to get to sleep". A parent stated "it's as though (the child) wants to go to sleep, but he's fighting his body and brain that is keeping him more awake". Overall, it appeared that in a majority of children, stereotypy, particularly vocal stereotypy, made the initiation of sleep difficult.

Stereotypy as Sleep-Conducive

A contrasting theme was stereotypy as sleep-conducive; including parent report that stereotypy was beneficial to the child (e.g., soothing), and/or the stereotypy occurred during the wake-sleep transition. Stereotypic behaviours in six children (40%), all motor, were classed as sleep-conducive. One parent described her child's continuous bouncing as a "wind-down" and stated "it helps him prepare for sleep". Body-rolling in another child was described as "he holds a blanket over him and does his death roll, like a crocodile, he thrashes from side to side". Another child engaged in a side-to-side rock whilst standing on the bed, pushing off the wall with one hand, until she physically 'dropped' into a sleep state: "she has to work very hard to fall asleep". Interestingly, in five of these children (83%), sleep-conducive motor stereotypy only occurred at night. This contrasted with vocal stereotypy and RMO, which were frequently reported to also occur throughout the day. One exception was the child who engaged in a standing-rock during the wake-sleep transition, who also rocked (swayed side-to-side) whilst standing throughout the day, albeit at a reduced intensity.

Notably, two children (13%; with severe SOD) were reported to exhibit both sleep-interfering and sleep-conducive stereotypy. In these children, sleep-interfering stereotypy (bouncing, squealing, and hand-flapping for one child; RMO with magazines for the other) occurred extensively both throughout the day and the sleep onset period. Sleep-conducive stereotypy (body-rolling, and body-rocking, respectively) occurred immediately prior to

falling asleep, specific to the wake-sleep transition. Overall, parent report suggests that children's repetitive behaviours may serve different roles in relation to sleep disturbance, across and within children.

Parent Responses to Stereotypy

Parent responses to stereotypy varied and included no response/ignoring the behaviour (i.e., purposefully withholding attention), giving a verbal instruction (e.g., to cease the behaviour and/or to go to sleep), and co-sleeping (in the child or parent bed). Parents tended to ignore vocal stereotypy when it occurred at low volume, but to intervene with a verbal instruction (e.g., "it's time for sleep") when vocalisations escalated: "if he is being loud, then one of us will go in and have stern words with him". Seven families co-slept; among parents who co-slept, five (71%) reported doing so as a reactive response to NWs, while two families co-slept from the beginning of the night.

Interestingly, a sub-theme was identified regarding parent's intentional use of co-sleeping to manage stereotypy. Parents recounted using their body to try to physically restrict children's motor stereotypy: "(parent) will use our body, an arm and a leg, to physically help to hold (child) down to try to calm his body enough to help him to get off to sleep". Parents saw their presence as helpful in reducing motor movements: "so long as I lie with him, he doesn't bounce" and "if I don't co-sleep with her, she'll stand up and rock". Parents also co-slept in response to vocal stereotypy, particularly if less intensive attempts (i.e., ignoring, verbal instructions) to interrupt vocalisations were unsuccessful. Parent report indicated that children tended to vocalise despite parental presence. One parent explained that their child was unlikely to resettle when brought into the parent bed, instead going into "his own world" continuing to "babble and chat to himself". Overall, parent responses to children's behaviour varied across and within families. Although coded, not enough data were available to

generate a theme regarding the impact of parent response on the child's behaviour (i.e., whether behaviour increased, decreased, or was unaltered), thus, it remains unclear.

Discussion

This study drew upon the perspectives of parents of children on the autism spectrum with sleep problems, to increase our understanding of the types and topography of sleep-related stereotypic behaviour, how parents are managing stereotypic behaviours in the sleep context, and the potential role of stereotypy in relation to sleep disturbance. Parents reported their children exhibiting a wide range of stereotypic behaviours at night, broadly including motor and vocal behaviours, and to a lesser extent RMO. This finding is unsurprising, as it is widely accepted that stereotypy presents heterogeneously across and within individuals (Cunningham & Screibman, 2008; Rapp & Vollmer, 2005). However, it does underscore extant research (e.g., Jin et al., 2013; Hunter et al., 2021; McLay, France, Blampied, et al., 2019; McLay, France, Blampied, van Deurs, et al., 2021) suggesting children on the autism spectrum can engage in a wide variety of repetitive behaviours both at the start of and/or during the night. Future research is needed to better understand the types of stereotypy children engage in at night, including characterising the topography, severity (e.g., frequency, duration), prevalence and patterns (e.g., temporal occurrence) of behaviour, which ultimately will help practitioners to align treatment to the individual needs of children on the autism spectrum. The relationship between day- and night-time stereotypy also requires further investigation, including the extent to which behaviours may differ, and whether treating daytime stereotypy reduces the occurrence of stereotypy at night, and vice versa.

Parent responses to children's sleep-related stereotypy varied in intensity, from no response to co-sleeping. That some parents employ co-sleeping (i.e., a response that was undesired by the parents) as an intentional attempt to manage sleep problems including stereotypy, particularly motor stereotypy, suggests parents may struggle to overcome these

behaviours alone, and require specialist support. Although the impact of parent response on behaviour was not able to be established, this also raises important questions regarding the behavioural function of children's stereotypy. Notably, given that children are typically alone (i.e., in their bedroom) when they engage in stereotypy, it is likely that stereotypy frequently functions to produce automatic reinforcement. That parents tend to ignore stereotypy may be a learned product of their response having had little to no impact on automatically reinforced behaviour. The extent to which stereotypy serves other or additional functions in the sleep context, however, requires further investigation. In many cases in this study, children's stereotypy eventually resulted in parental attention, including co-sleeping; the possibility that stereotypy is socially-mediated, even partially, cannot be ruled out.

It is also feasible that escalations in children's behaviour are reinforced by parents consistently responding to stereotypy when displayed at heightened levels. Further, stereotypy may operate as a form of escape (i.e., obtaining negative reinforcement), for example from the demand to go to sleep or by mitigating physical discomfort. Careful consideration of the possible motivating consequences for stereotypy is important, to inform function-based treatment (Cunningham & Schreibman, 2008; Jin et al., 2013; McLay, France, Blampied, et al., 2019). Specifically, as was reported by parents and further illustrated in this case study, if behaviour is multiply determined, it may be necessary to eliminate external motivators for behaviour (e.g., parent attention), as well as addressing underlying automatic sensory consequences (Cunningham & Schreibman 2008; Gwyther et al., 2017; Jin et al., 2013; McLay, France, Blampied, et al., 2019).

The distinction that parents made between sleep-interfering and sleep-conducive stereotypy was an interesting finding, which underscores the importance in utilising FBA to inform individualised case conceptualisations and treatment plans and holds pragmatic value for treatment. In a majority of children, stereotypy, particularly vocalisations, were

conceptualised as sleep-interfering; however, this was not reported to be the case for all children. This raises questions about whether stereotypy may differentially affect sleep onset across and within children and is an important consideration in clinical assessment and treatment, and an interesting avenue for further research. Sleep-interfering stereotypy might be considered a form of stimulation or "play" that directly competes with behavioural quietude, and thus inhibits the onset of sleep. In such instances, the underlying issue may be motivational, in that the reinforcer of stereotypy is more salient, immediate, and desirable than the delayed reinforcer of sleep (Blampied & France, 1993; Blampied & Bootzin, 2013; Rapp et al., 2017).

Accordingly, treatment focus would be on the reduction of sleep-interfering behaviour to allow opportunity for behavioral quietude to occur, and strategies that increase motivation for sleep (e.g., through sleep restriction or faded bedtime procedures; Hunter et al., 2021; Jin et al., 2013; McLay, France, Blampied, van Deurs, et al., 2021; Piazza & Fisher, 1991a). Stereotypy can be difficult to treat owing to the challenges inherent in interrupting automatic reinforcement contingencies that are not accessible to external control; in the case of vocal stereotypy for example, the reinforcer is concomitant with the behaviour itself and highly immediate (Akers et al., 2020; Boyd et al., 2012; Lanovaz & Sladeczek, 2012; Rapp & Vollmer, 2005). Procedures that increase motivation to initiate and maintain sleep, in effect removing the opportunity for engagement in sleep-interfering behaviours, may be a useful means to treat stereotypic behaviour that is otherwise hard to manipulate (Hunter et al., 2021).

In contrast to sleep-interfering stereotypy, it is possible that sleep-conducive stereotypy represents a learned type of self-settling behaviour in some children, perhaps similar to RM in infants and young children (Haywood & Hill, 2012; Hoban, 2003). If sleep-conducive stereotypy is a learned variety of self-settling, a greater treatment emphasis may

need to be placed on teaching replacement skills (e.g., relaxation leading to behavioural quietude) so the child is taught more adaptive, efficient means to initiate sleep. The distinction between sleep-interfering and sleep-conducive stereotypy requires further investigation, including whether such a distinction exists, and how varying types of stereotypy fit within these categories (e.g., if vocalisations are primarily interfering).

Further, the mechanisms underlying the stereotypy and sleep relationship warrant further attention. For example, motor stereotypy may serve to modulate anxiety in some children on the autism spectrum (Russell et al., 2019); anxiety is prevalent in individuals on the autism spectrum (D. Adams et al., 2019) and may contribute to sleep disturbance (Mazurek & Petroski, 2015; Richdale & Baglin, 2015). It is noteworthy that when considering whether sleep-related stereotypy is problematic, it is the relationship of the behaviour to sleep onset that is important (Gwyther et al., 2017); sleep-conducive stereotypy may still warrant targeted treatment if a child's ability to settle and/or re-settle to sleep efficiently is impaired through perseverance with repetitive behaviour.

The degree of overlap between sleep-related motor stereotypy as identified in this study and rhythmic movement disorder (RMD; a form of parasomnia) is unclear (Veatch et al., 2015). There may be a high degree of correspondence, given RMD is defined by stereotyped and rhythmic movements often involving large muscle groups (e.g., body-rocking, head-rolling), that occur in association with sleep (often during the wake-sleep transition), and which can impair sleep by delaying sleep onset (Gwyther et al., 2017; Haywood & Hill, 2012; Veatch et al., 2015). As a parasomnia, rhythmic movements in RMD can also occur during sustained sleep, and disrupt sleep quality (Gwyther et al., 2017; Haywood & Hill, 2012; Hoban, 2003). Diagnosis of RMD is typically based on parent report (Gwyther et al., 2017).

The degree of overlap between stereotypic and RM behaviours requires further investigation; for example, it is unclear whether the child in this study who engaged in a standing rock until she 'dropped' into sleep would constitute an extreme variety of RMD. Notably, there is insufficient information regarding RMD in populations outside of children with typical development (Gwyther et al., 2017) to understand its overlay with autism-related stereotypy. Interestingly, persistent RMD (i.e., beyond five years of age) has been associated with developmental disorders, particularly ADHD, but also autism (Gwyther et al., 2017; Hoban, 2003; Johnson et al., 2009; Stepanova et al., 2005).

Conversely, motor stereotypy, although strongly associated with autism, is not exclusive to autism, and occurs in children with typical development, particularly during early childhood (Goldman, Wang, et al., 2009; Singer, 2009). This raises important questions regarding how repetitive behaviours are classified and understood in autism, for instance, whether RMD and sleep-related motor stereotypy are a similar or entirely different phenomenon, of shared or distinct aetiology. It also highlights the complexity and multiplicity of sleep problems that children on the autism spectrum can experience; for example, sleep-related repetitive behaviours may be linked to insomnia (e.g., SOD/NWs), parasomnia (e.g., RMD), or both. For example, in a study by Weiskop et al. (2005), one participant child on the autism spectrum exhibited stereotypic rocking during NWs. Weiskop et al. (2005) reported that it could not be determined whether the night-time rocking was a form of automatically reinforced stereotypy or occurred whilst the child was asleep and was thus a form of parasomnia.

Other sleep-related movement disorders are also relevant to autism, such as restless leg syndrome involving repetitive movement of the legs, or larger muscle groups (e.g., body-rocking) involving the legs, during the wake-sleep transition (Kanney et al., 2020; Veatch et al., 2015). Restless leg syndrome is often associated with periodic limb movements in sleep,

involving repetitive movements of extremities during sleep (Veatch et al., 2015). Understanding the distinction between repetitive movements in relation to sleep disturbance is important to knowing how to treat such behaviour (Veatch et al., 2015; Weiskop et al., 2005). It is necessary to determine whether a child is awake and there are operant qualities to repetitive behaviour when evaluating sleep problems for behavioural treatment.

The findings of this research underscore the importance of determining parent's views in relation to the assessment and treatment of children's sleep problems. Parents are in a unique position to report on child behaviour that occurs within the home environment; qualitative methods provide a means of exploring parent perceptions of child behaviour, which can add insight and understanding of the complex nature of sleep problems in children on the autism spectrum (Kirkpatrick, Gilroy, et al., 2019). Further, parents' sleep-related cognitions are an important consideration in treatment planning, not least because they inform parent's own response to child behaviour, which may be unintentionally reinforcing sleep problems (e.g., lying with a child in response to NWs; Hastings, 2002; Levin & Scher, 2016), and may inform parents' help-seeking behaviour (McLay et al., 2020). For example, parents who view their child's behaviour as beneficial to the child (e.g., stereotypy as soothing) may be less likely to seek or be willing to engage with treatment aimed at reducing the behaviour.

The sleep context also poses unique challenges for the treatment of sleep-related stereotypy, including that parents must act (where possible) as primary interventionists within their own home (Hanley et al., 2014; Jin et al., 2013). It is important, therefore, that sleep problems are assessed and addressed in collaboration with parents (Jin et al., 2013; Moes & Frea, 2002; Pattison et al., 2020), and that treatment includes parent education, training, and up-skilling, to help parents to manage sleep-related behaviours within their own home (Boyd et al., 2012; Haywood & Hill, 2012; Lanovaz et al., 2016; Specht et al., 2017).

Limitations and Future Research

Several limitations may limit confidence in the overall reliability of results. First, this study focussed on children who were referred for sleep intervention services and whose parents' reported engagement in sleep-related stereotypy. Results are therefore representative of a subset of children on the autism spectrum experiencing clinically significant sleep disturbance and stereotypy and may not accurately reflect the wider population. Second, stereotypy data were obtained using semi-structured interviewing within the context of a clinical assessment. This enabled the researchers to ensure consistency in how core information was elicited across parents, whilst allowing parents to share individualised experiences with depth (Belotto, 2018). However, given the purpose of the interview (i.e., to clinically assess children's sleep problems), the types of questions asked may have influenced the types of responses provided by parents (e.g., questions about stereotypy were asked in relation to known sleep problems). Further, thematic analysis was conducted by coding clinical assessment reports; thus, prior to coding, data were limited to that which were clinically relevant, and may have excluded information pertinent to this topic. Finally, the same author primarily conducted the interviews and led the data analysis; whilst steps were taken to mitigate risk of bias (i.e., assessing interrater reliability), the results may not be entirely free of interactions and influences of the author's own values and biases. Thus, the findings of this study are tentative, and are presented for the purpose of stimulating future research.

Future research could clarify and strengthen the results of this study by examining parent perspectives of sleep-related stereotypy in children on the autism spectrum using a broader interview framework to transcribe, code and analyse parents' verbatim responses. This is likely to generate fuller, unconstrained data sets for analysis. The incorporation of instrumented sleep measures (e.g., VSG) into assessment would help to triangulate parent-

report and broaden insight into children's stereotypic sleep-related behaviour, including behaviour which parents may be unaware of and therefore unable to report on. For example, it is possible that children engaged in a wider range of stereotypy, with differing implications for sleep, than what was identified in this research. Ideally, this should include assessment of vocalisations, as a limitation of the case study was the inability of VSG to capture audio, which meant IOA of vocal stereotypy was not possible.

It is important that future research specifically investigate whether a reduction in sleep-related stereotypy decreases SOD and/or the duration of NWs. Based on the findings of this analysis, it may be hypothesised that reducing sleep-related stereotypy (particularly vocal stereotypy) decreases SOD and/or the duration of NWs, which would suggest that stereotypy interferes with sleep onset. Finally, future research should explore whether children on the autism spectrum without sleep problems engage in stereotypy in the sleep context, and how this may differ to sleep-interfering stereotypic behaviour. It may be hypothesised that the severity (e.g., frequency, duration), prevalence and patterns (e.g., temporal occurrence) of behaviour differs in accordance with varying severity of sleep problems (e.g., no sleep problems, mild-moderate, and severe sleep disturbance). For example, in children who sleep well, stereotypy may be limited to the initial sleep onset period (in the absence of NWs) and occur briefly, such that perseverance with repetitive behaviour does not disrupt sleep. Such research would enhance our understanding of the sleep-stereotypy nexus; for example, it is possible that sleep-related stereotypy commonly occurs in children on the autism spectrum, but may not be considered problematic, detected and/or reported by parents.

Chapter 9

Systematic Review of the Collateral Effects of Behavioural Sleep Interventions in Children and Adolescents on the Autism Spectrum

A focus of the preceding quantitative Studies 1-3 was to examine the effectiveness of function-based behavioural interventions to treat sleep problems, and stereotypy in the context of sleep disturbance, in children on the autism spectrum. Aside from primarily evaluating the magnitude and durability of change in children's sleep following a BSI, it is important to assess whether there are wider treatment benefits experienced by children themselves. As discussed in Chapter 1, sleep problems are robustly associated with detrimental effects for children on the autism spectrum, including increased autism symptom severity (including increased stereotypy and social impairment) and increased internalising and externalising challenges (e.g., anxiety, aggression; H. Adams, Matson, Cervantes, et al., 2014; S. Cohen, Conduit, et al., 2014; Schreck et al., 2004; Veatch et al., 2017). Treating sleep problems therefore carries the potential to improve functional outcomes for children on the autism spectrum (H. Adams, Matson, Cervantes, et al., 2014; Hollway & Aman, 2011; Hundley et al., 2016).

Such 'secondary' outcomes are termed collateral benefits, defined as change in behaviour that was not directly targeted during intervention (Ledbetter-Cho et al., 2017), and are an important aspect of the generality of interventions (Cooper et al., 2020; Heyvaert et al., 2014). Therefore, the focus of this chapter and the next are to examine the collateral effects of BSI. This chapter consists of a manuscript of a systematic review of empirical research examining the collateral effects of BSI in children on the autism spectrum, which has been

published in *Research in Autism Spectrum Disorders* with myself as the first author[10]. Chapter 10 examines collateral effects in participant (Studies 1-3) children and parents.

Introduction

Autism is characterised by impairments in social interaction and communication, and the presence of restricted and repetitive behaviours (APA, 2013). Children on the autism spectrum also experience a multitude of co-occurring challenges, including emotional and psychiatric disturbances, externalising problems (D. Adams et al., 2019; Ledbetter-Cho et al., 2017; Mazzone et al., 2018; Postorino et al., 2017) and difficulties with sleep. Sleep problems affect up to 80% of children on the autism spectrum (Johnson et al., 2018; Richdale & Schreck, 2009; S. Cohen, Conduit, et al., 2014), and commonly include problems with sleep onset and maintenance. Examples are bedtime resistance, SOD, frequent and/or prolonged NWs, unwanted co-sleeping (CS), EMW and reduced TST (Abel et al., 2018; Cortesi et al., 2010; Krakowiak et al., 2008; Malow, Marzec, et al., 2006).

The aetiology of sleep problems is likely to be the result of a complex interaction between a range of factors, including biological, psychological, social, behavioural, medical, and environmental variables (Cortesi et al., 2010; Mazzone et al., 2018; Richdale & Schreck, 2009; Singh & Zimmerman, 2015). No single treatment approach can effectively reduce all types of sleep problems in children on the autism spectrum (Cuomo et al., 2017). Instead, the type of intervention chosen should be informed by the variables underpinning sleep disturbance (McLay, France, Blampied, van Deurs, et al., 2021). Behavioural interventions (i.e., interventions based on the principles of ABA) are recommended as the first line of treatment for sleep disturbance in children on the autism spectrum (K. Turner & Johnson,

[10] Hunter, J. E., McLay, L. K., France, K. G., & Blampied, N. M. (2020). Systematic review of the collateral effects of behavioral sleep interventions in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders, 79,* 101677. https://doi.org/10.1016/j.rasd.2020.101677

2013; Meltzer & Mindell, 2014; Rigney et al., 2018). Multiple reviews and a recent meta-analysis have shown that behavioural interventions, including parent training, sleep hygiene (i.e., practices promoting healthy sleep), extinction (i.e., the full removal of putative reinforcers for sleep disrupting behaviour), graduated extinction, faded bedtime and reinforcement (for sleep-facilitating behaviours), can significantly reduce sleep problems in children on the autism spectrum (Carnett et al., 2019; K. Turner & Johnson, 2013; Kirkpatrick, Louw, et al., 2019; Meltzer & Mindell, 2014; Rigney et al., 2018).

Without effective treatment, chronic sleep problems can have a widespread detrimental impact on children and their families (Austin et al., 2013; Johnson et al., 2018; Mazzone et al., 2018; S. Cohen, Conduit, 2014). Sleep plays an essential role in children's health and wellbeing, brain maturation, memory consolidation, and cognition (S. Cohen, Conduit, et al., 2014; Sannar et al., 2018). Conversely, sleep deprivation has been associated with a variety of deleterious effects in children on the autism spectrum, including anxious and depressive symptoms, hyperactivity, impulsivity, inattention, irritability, hostility, cognitive deficiencies, non-compliance, aggression (Goldman et al., 2011: Malow, Marzec, et al., 2006; Mayes & Calhoun, 2009; Mazurek & Sohl, 2016; Park et al., 2012; S. Cohen, Conduit, et al., 2014), self-injurious behaviours and disrupted eating habits (Hundley et al., 2016).

Furthermore, a number of studies have demonstrated that poor sleep is linked to autism symptom severity (Gabriels et al., 2005; Hundley et al., 2016; Schreck et al., 2004; S. Cohen, Conduit, et al., 2014). This specifically includes the core symptoms of autism, with higher rates of stereotypic behaviours and social impairment found in children who are poor sleepers (Abel et al., 2018; H. Adams, Matson, Cervantes, et al., 2014; Hoffman et al., 2005; Schreck et al., 2004; Veatch et al., 2017; Zachor & Ben-Itzchak, 2016). Thus, insufficient quantity and quality of sleep may further exacerbate challenges that many children on the autism spectrum experience (Johnson et al., 2018; Richdale & Schreck, 2009; Sikora et al.,

2012; Mazzone et al., 2018). In addition to the negative child impacts, parents of children on the autism spectrum who have sleep problems are found to experience significantly higher levels of stress, anxiety, depression, and fatigue than parents of children without sleep problems (Chu & Richdale, 2009; Johnson et al., 2018; K. Turner & Johnson, 2013; Wiggs & France, 2000). Thus, while evidence suggests that the negative consequences of sleep are extensive, the full range and magnitude of the impact of sleep problems is not yet known (Hundley et al., 2016; Johnson et al., 2018).

The mechanisms by which poor sleep affects autism symptom severity and daytime behaviour are not well understood (Mazurek & Sohl, 2016; Mazzone et al., 2018; Schreck et al., 2004; S. Cohen et al., 2018; Sannar et al., 2018). Most research examining the relationship has been cross-sectional, limiting our ability to determine the directionality of this relationship (Mazurek et al., 2019; S. Cohen, Conduit, et al., 2014; Wiggs & Stores, 1999). Based on extant research, it is thought that the relationship is bidirectional (Gregory & Sadeh, 2012; H. Adams, Matson, Cervantes, et al., 2014; Hollway & Aman, 2011; Krakowiak et al., 2008). For example, anxiety, underpinned by hyperarousal, is both a consequence of and a risk factor for sleep disturbance in children both with typical development and on the autism spectrum (Gregory & Sadeh, 2012; Hollway & Aman, 2011; Mazurek & Petroski, 2015).

Furthermore, specific behaviours are associated with particular types of sleep problems. For example, stereotypic behaviours are significantly correlated with sleep fragmentation (Goldman, Surdyka, et al., 2009), SOD (Tudor et al., 2012), sleep-disordered breathing (Hoffman et al., 2005), nightmares, night terrors, confusional arousals, screaming during the night (Schreck et al., 2004) and short sleep duration (Sannar et al., 2018; Schreck et al., 2004; Tudor et al., 2012; Veatch et al., 2017). More research is needed to better understand the complex interplay between sleep problems and these common characteristics

of autism (Hoffman et al., 2005; Hollway & Aman, 2011; Mazurek & Sohl, 2016; Mazzone et al., 2018; S. Cohen et al., 2018).

Behavioural interventions for challenging behaviour in children, including those on the autism spectrum, are generally focussed on a specific class of behaviours (e.g., aggression), and success is primarily evaluated in terms of the magnitude and durability of any change observed in the target behaviour(s). There is, however, often an expectation accompanying such interventions that there will be wider treatment benefits experienced by the child themselves, or by siblings, parents, or other caregivers of the child. Such outcomes are termed collateral benefits of treatment, defined as a change in behaviour that was not directly targeted during intervention (Ledbetter-Cho et al., 2017). Collateral treatment outcomes are highly desirable as they contribute to individual and collective wellbeing and represent an important aspect of the generality of behavioural treatments (Cooper et al., 2020).

A review by Ledbetter-Cho et al. (2017) of 46 behavioural intervention studies for children on the autism spectrum found that effective behavioural interventions were associated with 14 collateral benefits across several domains of functioning, including social communication, stereotypic and challenging behaviours. Interventions mediated by parents or teachers were particularly likely to produce collateral treatment effects (Ledbetter-Cho et al., 2017). These authors suggested collateral change in untargeted behaviours may be a common but under-researched outcome of behavioural interventions in children on the autism spectrum, but questions remain about the occurrence of collateral benefits resulting from interventions for sleep problems. Given the evidence of a relationship between sleep problems and autism symptom severity and daytime behaviour problems (Schreck et al., 2004; S. Cohen, Conduit, et al., 2014), and the collateral benefit of behavioural interventions for behaviours other than sleep (Ledbetter-Cho et al., 2017), it is plausible that effective

treatment of sleep problems in children on the autism spectrum may generate collateral improvement in areas of daytime functioning and wellbeing (Abel et al., 2018; Malow, McGrew, et al., 2006; Malow et al., 2014; Mazurek et al., 2019; Phung et al., 2019; Tudor et al., 2012; Wiggs & France, 2000).

The purpose of the current review is to systematically identify and evaluate studies that have assessed collateral effects of behaviourally-based sleep interventions for children and/or adolescents on the autism spectrum. To match the scope of Ledbetter-Cho et al (2017), this review also focussed on collateral effects for the children who were the target of the interventions, excluding possible benefits accruing to other family members or more widely. The findings should enhance our understanding of the relationship between sleep problems and autism, inform the focus of future research, and encourage practitioners to intervene effectively with sleep problems in children on the autism spectrum.

Methods

This systematic review and the preparation of this manuscript was undertaken in accordance with the PRISMA guidelines pertaining to systematic literature reviews (Moher et al., 2009), with the exception that this review was not registered.

Inclusion Criteria

Studies were included if they: (a) included at least one participant with ASD (or Pervasive Developmental Disorder, PDD); (b) included child and adolescent participants aged $\leq$ 18 years; (c) treated sleep problems using interventions that were based on the principles of ABA (e.g., stimulus control, graduated extinction), including cognitive behavioural therapy; (d) included sleep treatment outcomes (e.g., SOD, NWs or CS) as a dependent variable; (e) assessed at least one outcome other than sleep at pre- and post-treatment; (f) were published in a peer-reviewed journal; and (g) were published in English. Studies were excluded from

the review if they did not directly treat sleep problems, or if the treatment was not based on the principles of ABA (e.g., medical or pharmacological approaches). Outcomes related to the treatment itself (e.g., treatment acceptance or social validity) were not deemed to be collateral effects. In addition, measures of collateral impact on parents or families (e.g., parental stress) were also not considered within the review. Due to a lack of extant research no restriction was placed on study design nor date of publication. Eligible articles may have also included children with co-occurring diagnoses in addition to ASD and children with and without ID.

Search Strategy and Study Selection

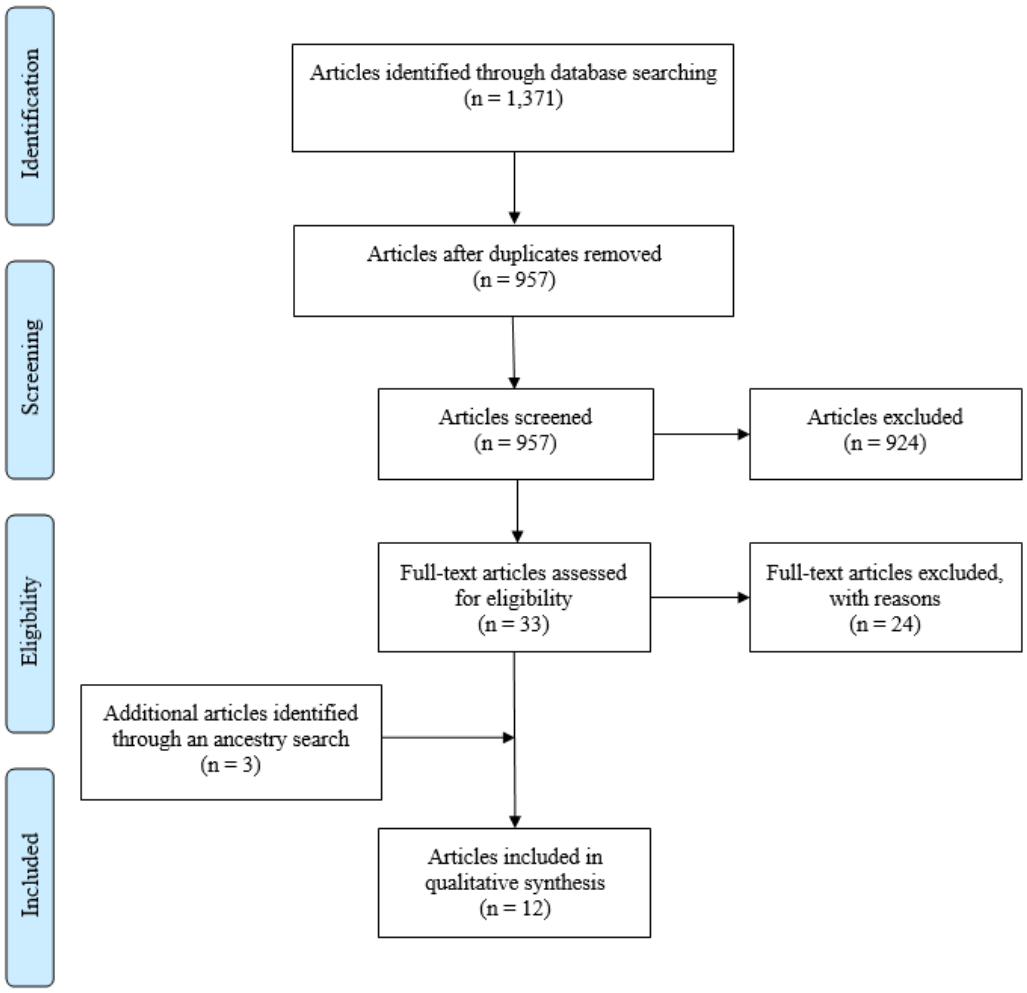
Figure 9.1 summarises the systematic search procedures for this review. A search of the electronic databases Education Resources Information Center (ERIC), PsycArticles, PsycINFO and the Psychology and Behavioural Sciences Collection was undertaken by the first author in October 2018 and updated in April 2020. Initially, the terms "ASD", "autism spectrum disorders", "autistic", "Asperger" or "pervasive developmental disorder" were combined with "sleep", "sleep problems", "sleep disturbance", "sleep difficulties" or "insomnia". If more than 1,000 results were returned a third search term "treatment", "intervention", "therapy", "management" or "training" was added. The initial search (2018 and 2020 combined) returned 1,371 articles. After the removal of duplicates, 957 articles were identified for screening.

The title and abstract of each article were read to assess whether behavioural interventions were used to treat sleep problems in children and/or adolescents on the autism spectrum. This resulted in the exclusion of 924 articles, and 33 full text articles were retrieved for further screening. Thereafter, the method section of the 33 articles was read to evaluate whether the study also included at least one collateral outcome measure concurrent with treatment. Measures were considered collateral if they assessed any outcome variable

that was not directly targeted by a behavioural sleep intervention (e.g., a measure of anxiety). In comparison, primary outcome measures were those reported as directly assessing the outcome of the sleep intervention (e.g., a measure of sleep latency). Based on this criterion, a further 24 articles were excluded. An ancestry search identified a further three articles for inclusion, thus a total of 12 articles were included in the final review, published between 1998 and 2020.

A research assistant independently replicated the search and screening process. Interrater reliability was calculated by dividing the total number of agreements by the total number of agreements plus disagreements and multiplying by 100 (Alresheed et al., 2018). Initial interrater reliability was 73%. The three articles that caused disagreement were then discussed until a consensus was reached about the inclusion or exclusion of each article. Following this discussion, agreement was 100%, with a total of 12 articles representing 10 research studies (for two studies the intervention and collateral results were presented across two articles) identified for data extraction.

Figure 9.1.



PRISMA Flow Diagram (Moher et al., 2009)

Data Extraction

Each study was summarised according to: (a) participant characteristics (gender, age, diagnoses, and sleep problems) and dependent variables; (b) research design and any follow-up assessment; (c) sleep intervention (agent, type, and dosage); (d) child sleep measures, intervention results, and social validity; (e) collateral outcome measures and results. Data extraction was completed by the first author and checked by the second and third authors to ensure the accuracy of the summaries and to validate the evaluation of methodological rigor.

Quality Assessment

The methodological rigor of the included studies was appraised, specifically for the intervention directed at the target behaviour(s), according to the evaluative method for determining evidence-based practices (EBP) in ASD (Reichow et al., 2008). This method offers a standardised means for rating the empirical evidence for ASD interventions for both group and single-case research design (SCRD). Individual studies were rated for research *rigor* which provided the basis for the evaluation of each study's research *strength* and the *EBP* of the body of research as a whole (see Reichow et al. 2008). These decisions were based on the steps detailed below.

Rigor of Research Reports

Depending on whether the study had a group design or SRCD, rubrics (guidelines) were applied to gauge the rigor of each study related to primary and secondary quality indicators (Reichow et al., 2008). Primary indicators were operationally defined according to a trichotomous scale (high quality, acceptable quality, or unacceptable quality) and secondary quality indicators were operationally defined according to a dichotomous scale (evidence or no evidence). Primary indicators common to both group and SCRD included the study reporting: (a) participant characteristics; (b) dependent measures; and (c) independent variables. Common secondary indicators included the study reporting: (a) interobserver agreement; (b) blind raters; (c) procedural fidelity; (d) generalisation and maintenance; and (e) social validity (Reichow et al., 2008).

Primary indicators specific to group research included the study reporting: (a) comparison condition; (b) link between research question and data analysis; and (c) use of statistical tests. Secondary indicators included reporting: (a) random assignment; (b) attrition; and (c) effect size (ES; Reichow et al., 2008).

Primary indicators distinct to SCRD included the study reporting: (a) a baseline condition; (b) visual analysis; and (c) experimental control. There was also one distinct SCRD secondary indicator, namely, calculation of kappa (a measure of IOA; Reichow et al., 2008).

Strength of the Research Report

Based on the number of primary and secondary indicators met, studies were classified as either *strong* (i.e., concrete evidence of high quality), *adequate* (i.e., strong evidence in most but not all areas) or *weak* (i.e., many missing elements and/or fatal flaws; Reichow et al., 2008). The strength rating for each study is provided in Table 1.

Classification of Treatment as Evidence Based Practice

The number of studies classified as meeting criteria for strong or adequate research rigor, in conjunction with the treatment outcomes reported in each study, determined whether the intervention practices met Reichow et al.'s (2008) criteria to be classed as either *established* or *promising* EBP.

One modification was made to Reichow et al.'s (2008) criteria; namely, the dependent variable primary indicator was altered to assess whether data gathered in each study was based on parent (or self-) report (e.g., sleep diaries or questionnaires), of if there were instrumented measures of sleep, such actigraphy or VSG (Sadeh, 2011). Sole reliance on self-report measures may be problematic for a range of reasons, including that parents cannot accurately report on aspects of sleep they are unable to detect; for instance, a child's quiet wakings (Jan et al., 2008; Rigney et al., 2018; Sadeh, 1994; Wiggs & Stores, 2004). The need for instrumented sleep measurement is well recognised (Carnett et al., 2019; Hodge et al., 2012; Knight & Johnson, 2014; Spruyt & Curfs, 2015), and studies were only awarded the highest quality rating for the dependent variable indicator if instrumented measures were

reported. The quality rating did not require that diary data have reliability assessed against any instrumented data concurrently obtained.

To assess the reliability of the research rigor ratings, IOA was assessed. One third of studies included in the review (group design $n = 3$, SCRD $n = 1$) were randomly selected and independently appraised by a trained research assistant. Agreement for rigor of the group and SCRD studies was 95% and 83%, respectively. Following discussion, a consensus was reached across all studies. Interrater evaluation was not performed for strength and EBP ratings owing to the straightforward numerical method involved in applying Reichow et al.'s (2008) criteria to these ratings.

Results

Table 9.1 summarises the studies' participants and sleep problems; research design; interventions; sleep measures and social validity; collateral effect measures and research rigor. All effects reported were either the result of visual analysis in SCRD studies, or for group studies were statistically significant at $p < 0.05$.

Table 9.1. *Summary of Studies that Treated Sleep Problems in Children and Adolescents on the Autism Spectrum and Included a Measure of Collateral Effects*

Authors	Participants and Sleep Problems	Research Design	Interventions	Sleep Measures and Social Validity	Collateral Effect Measures	Research Rigor
1. Austin et al. (2013)	7M, 2-6y (*M* = 4y) with ASD *Sleep problems:* BR (SOD), NWs, EMW, CS, irregular sleep-wake cycle	Pre/post *F/UP:* 1 mo post-T (Sleep Disturbance Index)	*Setting:* Home *Agent:* Parents *T:* Manualized parent group training - sleep hygiene, behavioural (e.g., grad. extinct.), sensory (e.g., darkened room) & communication (e.g., visual aids) & positive behav. support *Dosage:* 3x workshops 2 wks apart, then individual support & monitoring. Total dur. 15 wks	*Parent SD (rec. 2 wks pre- & post-T)*: < SOD & dur. of NWs. CS < for 1 par. *CSHQ:* < total score, but means remained above clinical cut-off post-T *Sleep Disturbance Index:* < between pre- & post-T scores. Gains not maintained at F/UP *CATS:* Satisfaction levels rated mod - high	*DBC-P:* < total behav. problem score	Weak
2. Delemere & Dounavi (2018)	4M/2F, 2-7y (*M* = 4.6y) with ASD *Sleep problems:* SOD, EMW, NWs, TST	Concurrent multiple-BL across par. *F/UP:* NR	*Setting:* Home *Agent:* Parents *T:* Individual parent training. Random allocation to bedtime fading or positive routines (*n* = 3) *Dosage*: Unclear	*Parent SD: Bedtime fading:* < SOD & > TST for all par. *Positive routines:* < SOD for all par., & > TST for 2/3 par. Dur. of NWs < for 1 par. *TAI and TEI-SF:* High acceptability	*Freq. of daytime CB (rec. by observer BL & T.): Bedtime fading*: freq. of CB < for all par. *Positive routines:* freq. of CB < for 1 par.	Adequate

3. Loring et al. (2016): Sleep data 3. Loring et al. (2018): Secondary outcome data	18 (M/F unclear), 11-18y (*M* = 14.8y) with ASD *Sleep problems:* SOD & WASO	Pre/post pilot study *F/UP (n* = 9)**:** 3 mo post-T (ph. call)	*Setting:* Home *Agent:* Parents & adol. *T:* Individual parent/adol. sleep ed. *Session 1:* Sleep ed., sleep hygiene, individual sleep plan & visual aids *Session 2:* Review 1 + relaxation & distraction *Dosage:* 2x 1 wk apart, then 2x ph calls 1 wk apart. Total dur. approx. 6 wks	*Actigraphy + parent/adol. SD (10-14 days pre- & post-T):* < SOD & > SE *ASHS:* > total score & 4/5 subscales parent & self-report *ASWS:* < total & all subscale scores parent & self-report *M-ESS:* < self-report total score *F/UP:* (verbal) gains maintained (parent/adol. report) *End of Education Survey:* High satisfaction overall	*RBS-R:* n.s. *ABC:* < hyperactivity & lethargy *C-CPT-II:* < % of Commissions Errors (measure of impulsivity/inattention) *BASC-2*: < 3/4 composite scores: Externalising Problems, Internalising Problems, & the Behavioural Symptoms Index. < in Internalising Problems subscales anxiety, depression, & somatisation, & Behavioural Symptoms subscale atypicality *F/UP:* (verbal) gains maintained (parent/adol. report)	Weak
4. Malow et al. (2014)	64M/16F, 2-10y (*M* = 5.8y) with ASD *Sleep problems*: SOD	Randomised 2-arm, pre/post *F/UP:* NR	*Setting:* Home *Agent:* Parents *T:* Manualized parent sleep ed. in individual (*n* = 41) or group (*n* = 39) format (size 2-4 parent) + 2 ph. calls Sleep ed.: Sleep hygiene, strategies to manage BR, NWs & CS (e.g., grad. extinct., visual	T comparison n.s. Individual vs. group arms combined. *Actigraphy (rec. 7-14 days pre- & 4 wks post-T) + parent SD (rec. 21 days pre- & 7-14 days 4 wks post- T):* < SOD & > SE	T comparison n.s. Individual vs. group arms combined. *RBS-R:* < stereotyped & restricted subscales & total score *CBCL:* < anxious/depressed, attention & withdrawn subscales	Strong

			aids, bedtime pass + Rx.), HW + 2x review ph. calls *Dosage*: *Group:* 2x 2-hr sessions 1 wk apart over 2 wks. *Individual:* 1-hr session. 4 wks between T & post-T measures.	*CSHQ:* < all insomnia-related subscales & total score *FISH:* > total score *End of Education Session Survey*: High satisfaction overall	*PedsQL:* > total score	
5. McCrae et al (2020)	12M/5F, 6-12y (*M* = 8.8y) with ASD *Sleep problems:* SOD & WASO	Pre/post pilot study *F/UP (n = 13)*: 1 mo post-T (all measures)	*Setting:* Home *Agent:* Parents & children *T:* Manualized individual parent/child CBT-CI adapted for ASD Sleep hygiene, stimulus control, parent management, fading CS, circadian ed., CBT (e.g., relaxation, managing worries), maintenance plan. *Dosage:* 8x 50-mins, 1 wk apart	*Actigraphy+ parent/child SD (rec. 2 wks at BL, post-T & F/UP):* < SOD & total wake time, > SE (*actigraphy & SD)* & > TST, < bed/wake time variability (*SD*) *F/UP:* > TST (*actigraphy*), all other gains maintained. *Treatment Satisfaction Questionnaire:* High satisfaction overall	*ABC:* < irritability, lethargy, stereotypy & hyperactivity *F/UP:* < inappropriate speech. All other gains maintained except hyperactivity	Weak
6. Moon et al. (2011)	2M/1F, 8-9y (*M* = 8.6y) with ASD *Sleep problems*: SOD	Case-series *F/UP:* 12 wks post-T (all measures)	*Setting:* Home *Agent:* Parents *T:* Manualized behav.T: FBRC + Rx. Handbook + exercises & weekly ph. call. *Dosage:* T dur. 7-9 wks	*Actigraphy + parent SD (rec. 1 wk at BL, post-T & F/UP):* < SOD for all par. *CSHQ:* < SOD for 2/3 par.	*CBCL:* < total scores (internalising & externalising) for all par. Gains maintained at F/UP	Weak

				F/UP: Maintained for all par., but higher SOD variability at F/up vs. BL for all par. *Parent Satisfaction Questionnaire:* Mod. levels of satisfaction		
7. Moss et al., (2014)	26 (M/F not reported), 8-17y (*M* = 11.7y) with DD (15 with ASD) *Sleep problems:* Parent-reported sleep disturb. (e.g.y, SOD, NWs, TST)	RCT *F/UP:* 8 wks post-T (all measures)	*Setting:* Home *Agent:* Parents *T:* Manualized parent group training. Random allocation to Tor WLC (*n* = 13) Ed. group workshops (5-7 per group) then individual T- sleep ed., sleep hygiene, communication, sensory (e.g., temperature) & behav. strategies (e.g., grad. extinct.), relaxation, Rx. On-going ph. calls for 3+ mo *Dosage:* 2x 3-hr workshops. 10 wks between pre- & post-T measures	*CSHQ:* < mean sleep disturb. scores for both groups (pre-, post-T & F/UP). Change sig. greater for T group *Semi-structured interview & CATS:* High satisfaction overall	*DBC-P:* n.s. in total scores nor subscales over time, nor between groups	Weak
8. Papadopoulos et al. (2019)	54M/7F, 5-13y (*M* = 10y) with ASD-ADHD *Sleep problems:* behav. sleep disorder or	RCT pilot study *F/UP:* 3 & 6 mo post-randomisatio	*Setting:* Home *Agent:* Parents *T:* Brief behav. sleep program. Random allocation to T group (*n* = 28) or UC (*n* = 33) Sleep ed., sleep hygiene & individual behav. strategies	*CSHQ:* < sleep problems at 3 mo for T group vs. UC, not maintained at 6 mo	*SDQ:* > emotional functioning for T group vs. UC at 3 mo (parent- & teacher-report), not maintained at 6 mo *ADHD Rating Scale IV:* n.s. (parent- & teacher-report)	Adequate

	night-time anxiety	n (all measures)	(e.g., bedtime fading, parental presence) *Dosage:* 2x consultations + ph. calls 2 wks apart		*PedsQL:* > psychosocial quality of life for treatment group vs. UC at 6 mo	
9. Reed et al. (2009)	16M/4F, 3-10y (*M* = 5.8y) with ASD *Sleep problems*: SOD, NWs, EMW & CS	Pre/post pilot study *F/UP:* NR	*Setting:* Home *Agent:* Parents *T:* 5 small group (3-5 per group) parent ed. workshops *Session 1*: Sleep hygiene, visual aids & grad. extinct. *Session 2:* < NWs & EMW, bedtime pass & Rx. *Session 3:* Addressed individual sleep concerns + HW & within session reviews *Dosage:* 3x 2-hr workshops over 3 wks	*Actigraphy + parent SD (n = 12; rec. 1 wk at BL & 1 mo post-T):* < SOD & time in bed *CSHQ:* < total scores & BR, SOD, sleep dur. & sleep anxiety subscales *FISH:* improved bedtime routine, > relaxing/ < stimulating activities, > sleep hygiene & < parent items *End of workshop parent survey:* Mod.- high satisfaction overall	*RBS-R:* > restricted behav. scale *PCQ:* < sleep disturb., hyperactivity & self-stimulatory behav.	Weak
10. Wiggs & Stores (1998): Sleep data 10. Wiggs & Stores (1999): Behav. data	18M/12F, *M* age 8.2-10.7y, with severe learning disabilities & CB (4 with ASD)	RCT *F/UP:* 1 & 3 mo post-randomisation (all measures)	*Setting:* Home *Agent:* Parents *T:* Individual behav. sleep program. Par.'s schools randomly allocated to T group (*n* = 15) or control (*n* = 15) Extinct., grad. extinct., stimulus control + Rx. Weekly ph. calls	*Actigraphy (rec. 3x nights pre-, 1 & 2 mo post-T):* n.s. between groups. > sleep period & < magnitude, freq. & pattern of movement during sleep over time for both groups	ABC admin. at BL (parent & teacher-report), 1 mo post-T (parent-report) & 3 mo post-T (parent- & teacher-report): *ABC (General Daytime Behav.): Parents:* n.s. between groups, but < over time for both groups' irritability, lethargy & hyperactivity	Adequate

	Sleep problems: SOD, NWs, CS & EMW	*Dosage:* 1x consultation + ph. calls 1 wk apart. 12 wks between pre- and final post-T measures.	*Composite sleep index:* < sleep problems for T group at 1 mo, maintained at 3 mo	*Teacher:* n.s. between groups, but < over time for both groups' irritability & hyperactivity *ABC (CB):* n.s. between groups. Aggression, noncompliance & temper tantrums < over time for both groups (teacher-report)

Note. <: decrease; >: increase; ABC: aberrant behaviour checklist; ADHD: attention deficit hyperactivity disorder; ASD: autism spectrum disorders; ASHS: adolescent sleep hygiene scale; ASWS: adolescent sleep wake scale; BASC-2: behavioural assessment system for children, second edition- parent rating scale; behave.: behaviour/behavioural; BL: baseline; BR: bedtime resistance; CATS: caregiver acceptance of treatment survey; CB: challenging behaviour; CBCL: child behaviour checklist; CBT: cognitive behavioural therapy; CBT-CI: cognitive behavioural treatment for childhood insomnia; C-CPT-II: Conners continuous performance test II; CS: co-sleeping; CSHQ: child sleep habits questionnaire; DBC-P: developmental behaviour checklist – parent version; DD: developmental delay; EMW: early morning wakings; F: female; FBRC: faded bedtime with response cost; FISH: family inventory of sleep habits; F/UP: follow-up; HW: homework; M: male; *M*: mean; M-ESS: modified Epworth sleepiness scale; NR: not recorded; NWs: night wakings; PCQ: parental concerns questionnaire; PedsQL: pediatric quality of life inventory; RBS-R: repetitive behaviour scale–revised; RCT: randomised control trial; Rx: reinforcement; SD: sleep diary; SDQ: strengths and difficulties questionnaire; SE: sleep efficiency; T: treatment; TAI: therapy attitude inventory; TEI-SF: treatment evaluation inventory – short form; TST: total sleep time; SOD: sleep onset delay; UC: usual care; WASO: wake after sleep onset; WLC: waitlist control; admin.: administered; adol: adolescents; approx.: approximately; behav: behaviour/al; diff: difference; disturb: disturbance; dur: duration; ed: education; extinct: extinction; freq.: frequency; grad: graduated; hr: hour/s; improv: improvement; mins: minutes; mo: months; mod: moderate; *n:* sample size; n.s.: not statistically significant; par: participant/s; ph: phone; post-T: post-treatment; rec: recorded; sig.: significant; vs.: compared with; wk/s: week/s; y: years

Participants and Sleep Problems

Prior to attrition, the total number of participants across the 10 studies was 306. The average age of participants was eight years old (range = 2–18 years) and 80% of participants were male. In seven[1-6,9] of the studies participants had a diagnosis of ASD (inclusive of PDD) only, with co-occurring diagnoses (e.g., learning disabilities or ADHD) or mixed samples (e.g., Down or Angelman syndrome) in the remaining studies[7,8,10]. All participants displayed some form of sleep disturbance, mainly SOD and NWs, followed by EMW, CS and reduced TST. Other sleep problems included irregular or delayed sleep/wake cycles, first wake after sleep onset and night-time anxiety.

Study Design and Follow-up

Four[4,7,8,10] of the 10 studies employed randomised control trials (RCT; sample size ranged from 26 to 80 participants, M = 49). Four[1,3,5,9] used a pre- post- group treatment design with no control (sample size ranged from seven to 20 participants, M = 16), and the remaining two were SCRD. Of these, one[2] was a multiple baseline across participants design (n = 6) and one[6] was a case-series design (n = 3). Seven studies[1,3,5-8,10] gathered follow-up data from one month to six months post-randomisation, only one of which was a SCRD study[6] with a 12-week follow-up.

Intervention Characteristics

All interventions were home-based interventions in which parents acted as primary intervention agents. All studies included ABA intervention components including sleep hygiene modifications, graduated extinction, and positive reinforcement. Five studies[1,4-7] utilised a treatment manual. Treatment duration ranged from two to 15 weeks. Intervention dosage for treatment groups typically included two to three training sessions or workshops

usually one week apart, followed by individual monitoring and support through phone calls. In one study[2] the duration of treatment was unclear.

Sleep Measures, Treatment Results and Social Validity

The most common dependent sleep measures were parent-reported sleep diaries and the CSHQ, used in seven[1-6,9] and six studies[1,4,6-9,] respectively. Most studies incorporated at least two measures of sleep, but three studies[2,7,8] included only one sleep measure (parent-reported sleep diaries[2] and the CSHQ[7,8]). Of the seven studies that included sleep diaries, five studies also utilised actigraphy[3-6,9]. One further study[10] employed actigraphy without accompanying sleep diaries. All 10 studies reported the targeted interventions had a positive effect on improving at least some aspect of sleep-related behaviour for children and adolescents on the autism spectrum. These positive effects were evident regardless of study design.

The most frequent reported improvement was a reduction in SOD, as measured by sleep diaries and actigraphy in five studies[3-6,9], and sleep diaries alone in two studies[1,2]. Six studies[1,4,6-9] reported an improvement in the total score and/or insomnia-related subscale scores on the CSHQ. Interestingly, one study[10] reported improvements in sleep (increased sleep period and decreased movement during sleep) for both the treatment and control group. Improvement in sleep outcomes was reported to be generally maintained in five[3,5-7,10] of the seven studies employing follow-up measures, while in two studies[1,8] intervention gains were found to have deteriorated at follow-up. Social validity was assessed in all but two studies[8,10] and consumer satisfaction was largely reported to be favourable.

Collateral Effect Measures and Results

A wide variety of secondary dependent measures were used to assess collateral effects; the range per study was one to four measures. Most studies incorporated a general

measure (i.e., the measure assessed a wide range of symptoms and behaviour) of problem behaviour or of internalising and externalising symptoms. Specifically these were the Aberrant Behaviour Checklist (ABC; Aman & Singh, 1986) in three studies[3,5,10], the Behavioral Assessment System for Children, Second Edition (BASC-2; Community-University Partnership for the Study of Children, Youth, and Families, 2011) in one study[3], the CBCL in two studies[4,6], the Developmental Behaviour Checklist: Primary Carer Version (DBC-P; Einfeld & Tonge, 2002) in two studies[1,7], the Parental Concerns Questionnaire (PCQ; McGrew et al., 2007) in one study[9], and the Strengths and Difficulties Questionnaire (SDQ: Goodman, 2001) in one study[8].

Five studies investigated the effect of a behavioural sleep intervention on specific symptoms and behaviours. Of these, three studies[3,4,9] investigated the effect on stereotyped behaviours using the RBS-R, and two studies[3,8] explored the effect on symptoms of ADHD (inattention and/or hyperactivity/impulsivity) using the Conners' Continuous Performance Test II (C-CPT-II; Conners et al., 2004) and the ADHD Rating Scale IV (DuPaul et al., 1998), respectively. Improvement in psychosocial quality of life was assessed in two studies[4,8] using the Pediatric Quality of Life Inventory (PedsQL; Varni et al., 2007). Frequency of challenging behaviour was recorded via time-sampling in one study[2]. All measures were parent-report except for one SCRD study[2] in which time-sampling was recorded by an independent observer. Only two studies[8,10] incorporated teacher-report to triangulate parent-report on measures of behaviour using the SDQ[8], the ADHD Rating Scale IV[8] and the ABC[10].

Eight of the 10 studies reported positive collateral effects following the behavioural sleep intervention[1-6,8,9]. These effects were noted across several domains of functioning, including improvements in stereotypic (restricted and repetitive) behaviours[4,5,9] and in internalising and/or externalising symptomatology[1-6,8]. Specifically, gains were noted in

stereotyped[4,5,9] and restricted[4,9] behaviour, in the frequency of daytime challenging behaviour[2], emotional functioning[8], and in symptoms of anxiety[3,4], depression[3,4], withdrawal[4], irritability[5], lethargy[3,5], somatisation[3], inappropriate speech[5] and atypical behaviour[3]. Improvements were also found in areas of inattention[3,4] and hyperactivity[3,5,9]. Lastly, benefits were found in relation to children's quality of life[4,8]. While several benefits were observed across studies, it is important to note that not all studies found improvements using the same measures. For example, the RBS-R[3,4,9] and DCB-P[1,7] yielded different results in different studies. Furthermore, three studies[3,7,8] found no collateral treatment effects on at least one measure, and one study[10] demonstrated post-intervention change in both the treatment and control group.

In addition to assessing immediate collateral benefits, six studies[3,5-8,10] assessed the maintenance of these effects. Three studies[3,5,6] reported that collateral gains were generally maintained; one study[8] reported that gains were maintained on one measure and not maintained on another, and one study[10] reported maintenance of gains for both the treatment and control group. The final study[7] found no collateral treatment effects.

Rigor of Research Reports

Only one study[4] met criteria to be classified as *strong*. Three studies[2,8,10] were classified as having *adequate* strength, and six studies[1,3,5-7,9] were classified as *weak*. With regard to common primary indicators, all 10 studies received high quality ratings for participant characteristics and independent variables (e.g., treatment information was defined in replicable detail) except for two studies[3,7] which did not specify gender and one study[10] which summarised but did not specify their treatment program in detail. All 10 studies also included appropriate measures as dependent variables that were clearly linked to their

research focus. Six studies[3-6,9,10] included instrumented sleep measures (actigraphy) to triangulate parent-report, while the other four[1,2,7,8] relied solely on other self-report measures.

Four group design studies[1,3,5,9] scored poorly as they lacked a comparison control group. With respect to statistical power, most authors did not mention power considerations at all, or acknowledged the possibility that their study was under-powered due to small sample size. One SCRD[6] failed to demonstrate experimental control, due to the absence of replication (e.g., case-series design) or visual analysis (e.g., non-continuous data points). Many secondary indicators were absent across the 10 studies. Only three studies[2,5,10] conducted IOA and only two studies[5,8] incorporated blind raters. Treatment or procedural fidelity was assessed in five studies[2-5,8]. Maintenance of sleep and collateral treatment effects was assessed in seven studies[1,3,5-8,10] but one study[1] assessed the maintenance of sleep treatment effects only. Social validity was assessed in a majority (n = 8) of studies[1-7,9].

Only four studies[3,5,7,8] reviewed reported an ES (either a standardised mean difference or partial Eta-squared; Lakens, 2013). The lack of ES reporting in group design studies is a serious omission. It has long been recommended that researchers report a measure of effect of treatment as well as reporting any *p* value resulting from statistical testing (Wilkinson & Task Force on Statistical Inference, 1999; Wassertein & Lazar, 2016). Reporting ES for outcomes, including secondary measures, would be beneficial, because this would enhance understanding of the magnitude of any collateral benefits obtained. ES estimation and reporting is important because it sheds light on the clinical rather than just the statistical significance of a study's findings, and it has long been emphasised that clinical significance is what matters in applied research (Blampied, 2013b; Jacobson & Truax, 1991; Kline, 2013).

As part of determining clinical significance, Jacobson and Truax (1991) also introduced the concept of *reliable change*, defined as change in any measure of therapy

outcome that is larger than that expected due to measurement error alone. The absolute amount of change required in a measure (e.g., the CSHQ) can be stated as the Reliable Change Index (RCI) and is based on the standard error of measurement of the measure. To conclude that clinically significant change has occurred requires, first, that the amount of change observed exceed the RCI (on a case-by-case basis), and second, that the post-therapy measure has crossed some stated clinical threshold (Jacobson & Truax, 1991). It is clearly desirable, therefore, for research into both primary and secondary outcomes of therapies to report the RCI for the outcome measures and specify what criteria were used for determining clinically significant change. Methods for estimating ES for SCRD studies are much less well-developed, although a number are available and researchers should select one best suited to their data and report the chosen ES (Parker et al., 2011).

Overall, the empirical evidence generated by the 10 studies, in respect to both research rigor and observed outcomes (see Table 9.1), met criteria for the treatments evaluated to be classified as *promising* EBP (Reichow et al., 2008). It is important to note, however, that this conclusion was made with reference to the methodological rigor of the behavioural sleep interventions; empirical evidence regarding collateral treatment effects needs to be treated with caution.

Discussion

The purpose of this review was to identify and evaluate research into the collateral effects of behavioural sleep interventions in children and adolescents on the autism spectrum. All 10 treatment studies demonstrated some positive effects of behavioural sleep interventions across all participants. Eight of the 10 studies also reported collateral change in at least one untargeted area across participants. This included change in stereotyped behaviours, problematic daytime behaviour and other internalising and externalising

symptoms, including anxiety and hyperactivity. Benefits were also found for child quality of life.

Extant literature has shown an association between sleep problems and autism symptom severity including stereotyped behaviours and social impairment, as well as internalising and externalising difficulties such as anxious and depressive symptoms, inattention, hyperactivity, challenging behaviours such as aggression and non-compliance, and wellbeing. Little is known, however, regarding the mechanisms behind the sleep and autism relationship, including directionality and reciprocity of effects (Hollway & Aman, 2011; Mazurek & Sohl, 2016; Mazzone et al., 2018; S. Cohen, Conduit, et al., 2014). While previous research has focussed on the negative impact of poor sleep, the current review demonstrates that treatment of sleep problems may positively affect specific areas of daytime functioning and wellbeing.

The current review extends the findings of Ledbetter-Cho et al. (2017) who examined collateral benefits that resulted from behavioural interventions in children on the autism spectrum, without considering sleep interventions specifically. Akin to the findings of Ledbetter-Cho et al. (2017), behavioural sleep interventions appear to produce untargeted change in autism symptom severity (stereotyped behaviour) and challenging behaviours. In addition, Ledbetter-Cho et al. (2017) found collateral treatment effects may be more likely to occur when behavioural interventions are embedded in naturalistic settings. In this review, behavioural sleep interventions were conducted in the family home with parents as mediators in all treatment studies. While this does not contradict Ledbetter-Cho et al. (2017) it cannot be regarded as particularly supportive either, since there were no comparative exceptions to treatment in the home.

The results of this review need to be treated with caution until greater certainty of evidence has accumulated, as only one of the 10 studies met criteria to be classed as having

strong methodological rigor (i.e., concrete evidence of high quality) and only three studies were of *adequate* strength. The most common methodological limitations across treatment studies were poor experimental control, particularly in considering collateral effects, and a lack of IOA, blind raters, instrumented sleep measures and a lack of reporting of ES (Ledbetter-Cho et al., 2017; Reichow et al., 2008). When considering the results of studies with *strong* or *adequate* research rigor only, collateral effects remained evident across several domains of functioning. Specifically, for stereotyped and restricted behaviours, frequency of daytime challenging behaviour, symptoms of anxiety/depression, attention, withdrawal, emotional functioning, and child quality of life. One study also found changes in lethargy, irritability, hyperactivity, aggression, non-compliance, and temper tantrums for both the treatment and the control group.

Implications

Caution notwithstanding, the results of the current review present important implications for treatment research and for translation of research into practice. Improving children's sleep (and that of their family members) is a worthy goal in its own right, while the impact that poor sleep has on the daytime functioning of children on the autism spectrum makes it an important consideration for all treatment planning. Practitioners working to treat daytime challenges in children on the autism spectrum may wish to also prioritise the assessment and treatment of sleep disturbance when planning an intervention, since it may be underpinning the daytime behaviour. Furthermore, the potential efficacy of behavioural sleep interventions to positively affect the wider functioning of children and adolescents on the autism spectrum deserves consistent consideration because it is clearly an important aspect of the generality of the interventions. Efforts to assess treatment outcomes in addition to sleep should be routinely incorporated into research and clinical practice. To highlight one specific example, research studies often include measures such as the GARS as part of assessment to

corroborate diagnosis of autism in research participants. Repetition of such measures post-intervention would be a feasible way to assess change in symptoms of autism and would help to highlight the potential value of behavioural interventions in producing untargeted treatment benefits.

Limitations and Future Research

A limitation of this review is the potential that relevant studies were not identified for evaluation. The inclusion requirement that studies were published in a peer-reviewed journal may have meant that pertinent research was overlooked (e.g., unpublished theses). There are also several limitations inherent in the research reviewed. Firstly, the variety of dependent measures utilised and the reporting of results in total and subscale format made comparison of results difficult, owing to differing terminology across scales. The degree to which scales overlapped in terms of assessment was unclear. For example, collateral effects were reported for both problematic daytime behaviour and externalising symptoms, following the terminology of separate measures, but it is possible that these results were measuring the same construct. This consideration may also apply to a range of internalising symptoms. The use of a total score to assess collateral effects in some studies made it impossible to know which specific domains may have been altered by the behavioural sleep intervention.

In addition, this review was hindered by the limited range of collateral treatment effects identified in the literature. For example, no studies assessed effects on social functioning[11]. But poor sleep has been associated with social impairment in children on the autism spectrum (Schreck et al., 2004; Veatch et al., 2017) and improvement in social difficulties has resulted from behavioural interventions targeting other daytime behaviours

[11] Since the publication of the present review, a study by McLay, France, Blampied, Hunter, et al. (2021) has examined whether changes in social functioning in children on the autism spectrum occur following a BSI. The study by McLay, France, Blampied, Hunter, et al. (2021) is discussed in the next chapter.

(Ledbetter-Cho et al., 2017). The dependent measures of collateral effects were also limited by those chosen by the authors of the included studies. Half the studies utilised only one, usually general, measure of child behaviour and development (e.g., the CBCL or DBC-P). Such measures lack depth relating to specific autism symptoms or behaviours, so collateral effects relating specifically to autism remained unmeasured. Similar observations have been made about correlational studies examining sleep disturbance and autism symptomatology. For example, Veatch et al. (2017) noted the impact of short sleep duration on autism symptom severity was more apparent when symptoms were assessed using the Autism Diagnostic Interview – Revised (ADI-R; Lord et al., 1994) compared to the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1989), possibly because short sleep duration was most strongly associated with social impairment and rigidity which is assessed more thoroughly using the ADI-R than on the ADOS.

In addition, only one study (Delemere & Dounavi, 2018) incorporated direct observational measures of secondary treatment effects. Direct observational methods such as time-sampling and frequency counts are crucial to the assessment of behaviour change (Cooper et al., 2020). In future research a wider range of collateral outcomes, as well as the specific components that contribute to such effects, requires further investigation utilising a broader range of dependent measures.

The review findings also highlight questions regarding the nature of the relationship between sleep and autism, including the directionality and reciprocity of this relationship (S. Cohen et al., 2018; Sannar et al., 2018) and whether collateral improvements in daytime functioning (e.g., reduction of anxiety) have a bidirectional impact on improving sleep. The mechanisms underpinning collateral improvement are also not clear. For example, it is not known why, despite some aspect of sleep improvement occurring across all participants, not

all studies found collateral effects concurrent with treatment, and collateral effect outcomes varied across treatment studies.

It is also not known from this review which specific changes in sleep may be linked to collateral treatment effects. Akin to collateral effect measures, many authors chose to utilise general parent-report measures of sleep (e.g., the CSHQ), and often results were reported for total scores making it difficult to determine the specific nature of sleep changes. It is interesting to note that of the eight studies that employed observational measures of sleep (i.e., sleep diaries and/or actigraphy), seven reported a reduction in SOD along with at least one positive collateral effect. The two studies that utilised only one secondary outcome measure (the CSHQ in both cases) reported no collateral effects or mixed findings. These findings are not indicative of causative relationships since improvements in sleep may have occurred that went unmeasured. Instead, they raise important questions regarding the nature of the relationship between sleep and daytime functioning in children on the autism spectrum. For example, the literature suggests that particular types of sleep problems are associated with particular types of impairment; for example, short sleep duration has been specifically correlated with severe social impairment and with stereotypic behaviours (Schreck et al., 2004; Tudor et al., 2012; Veatch et al., 2017). Further research is needed to investigate whether targeting treatment to certain types of sleep problems leads to distinctive daytime improvements (Mazurek et al., 2019).

It is also possible that mechanisms other than improved sleep affected daytime functioning; for example, parents may learn to generalise behavioural management skills from the sleep context to daytime behaviours. Interestingly, one study in the current review noted objective changes in sleep as well as collateral improvements for both the treatment and the control group. It is possible that mechanisms for improved sleep, other than the behavioural sleep intervention, may lead to collateral change (e.g., responsivity to

assessment). Future analysis is needed to investigate how it is that poor sleep affects daytime functioning (S. Cohen et al., 2018) and vice-versa, and how the amelioration of sleep problems may result in secondary benefits in the immediate and longer-term. There may also be mediating factors that affect the likelihood of collateral treatment outcomes, such as the cognitive and communicative ability of participants (Gabriels et al., 2005; Ledbetter-Cho et al., 2017), the setting in which behavioural interventions are applied, and the capacity and competence of parents in delivering the treatments (Ledbetter-Cho et al., 2017). Research needs to be conducted to better identify the specific types of collateral effects that result from behavioural sleep treatments, for whom they are evident, and under which conditions.

A review of the literature on the collateral impact of all available treatments for sleep problems (e.g., medicinal, surgical) is also required as the scope of the current review was limited to behaviourally-based sleep treatments. Furthermore, the scope of the current review was limited to collateral effects that occurred for children and adolescents. As sleep problems also have a detrimental impact on parents/caregivers, siblings (Johnson et al., 2018; Spruyt & Curfs, 2015) and the wider family (Wiggs & France, 2000), further research should investigate the effect of improved sleep on parent/caregiver and family outcomes. Nor should possible school and educational collateral outcomes be ignored. Future research should also explore whether collateral effects that result from a behavioural intervention are typically beneficial in nature or whether negative (i.e., undesirable) effects also occur.

Finally, future research should aim to enhance methodological rigor by ensuring sufficient experimental control (e.g., use of comparison controls or replication of treatment effects), ensuring studies are adequately powered, reporting effect sizes, using reliability and fidelity checks, assessing maintenance of treatment effects, and using blinded data collection. However, it is noteworthy that the sleep context poses unique challenges as interventions are usually conducted in the family home and with parents as direct therapy agents (Carnett et al.,

2019; Kirkpatrick, Louw, et al., 2019). Consequently, it may not be possible for interventions to be blinded, and collection of IOA and procedural validity data can be difficult (Carnett et al., 2019; Rigney et al., 2108). Solutions may include using instrumented measures (e.g., VSG) to yield data for IOA and to reduce bias in reporting where blinding is not possible (Carnett et al., 2019; Rigney et al., 2018).

Conclusion

Overall, these findings suggest that the treatment of sleep problems in children and adolescents on the autism spectrum using ABA-based behavioural interventions may produce collateral benefits in other areas of functioning, such as improvement in internalising symptoms, challenging behaviours, autism symptom severity and child quality of life. However, the ability to draw definitive conclusions about collateral treatment effects in the review was limited by the quality and quantity of published results available for evaluation. More research yielding high quality independent treatment studies is needed to extend and replicate treatment effects, particularly research that focusses experimental control on collateral effects of treatment. In addition, the mechanisms for change underlying these effects are not known. The findings of the current review add to what is known regarding the relationship between sleep and autism and highlight important implications for practice and avenues for future research. Researchers and clinicians alike should consistently consider the role poor sleep may play in the development and/or maintenance of daytime challenges.

Chapter 10

Collateral Treatment Outcomes for Participant Children and Parents

The outcomes of the systematic review (Chapter 9) suggest that improvements in children's sleep following a BSI may be associated with improvements in children's daytime functioning and wellbeing, including reductions in daytime stereotypy, challenging behaviour and other internalising and externalising symptoms (e.g., anxiety and hyperactivity). Since the publication of this review (Hunter et al., 2020), I co-operated in authoring another study by McLay, France, Blampied, Hunter, and colleagues (2021) which examined whether BSI result in collateral benefits for children on the autism spectrum and their parents. This study extends the results of the case analysis by McLay, France, Blampied, van Deurs, et al. (2021; described in Chapter 2) that reports the outcomes of BSI for 41 children on the autism spectrum.

In the McLay, France, Blampied, Hunter, et al. (2021) study, change in children's daytime challenging behaviour, autism symptom severity, and maternal and paternal ratings of personal sleep quality, symptoms of depression, anxiety and stress, and relationship quality, were assessed using the CBCL, GARS-3, PSQI, the DASS-21 and RQI, respectively, pre- and post-treatment. For the majority of child participants, sleep problem severity significantly reduced following the BSI (as described in McLay, France, Blampied, van Deurs, et al., 2021). Further, results showed improvements in children's internalising and externalising symptoms, and autism symptom severity. In particular, reductions in children's restricted and repetitive behaviour, and improvements in social interaction, were found following intervention (McLay, France, Blampied, Hunter, et al., 2021). Small improvements were also found for maternal personal sleep quality and parental stress (McLay, France, Blampied, Hunter, et al., 2021).

A recent systematic review and meta-analysis of randomised controlled trials by Phillips et al. (2020) identified similar findings, reporting a small-moderate effect of BSI on total behaviour problems (not maintained at follow-up), and on inattention/hyperactivity at follow-up, in children with neurodevelopmental disorders including autism. Comparatively, no significant improvements were found in other internalising and externalising problems pre- to post-treatment, indicating a need for further research in this area.

Given the detrimental effects of poor sleep in children and parents, and the findings of Hunter et al. (2020) and McLay, France, Blampied, Hunter, et al. (2021), it is important to assess the range of benefits (in addition to improved sleep) that may accrue following a BSI. This includes examining potential collateral benefits for parents. This chapter presents data on the collateral effects of the BSI in participant children and parents from studies 1, 2 and 3, to address the following research questions:

1. Are there any collateral benefits of the selected behavioural sleep treatments?
    a. Are there improvements in children's daytime functioning and behaviour?
        i. Are there changes in the core symptoms of autism, including stereotypy and social communication?
        ii. Are there changes in overall autism symptom severity?
        iii. Are there changes in in internalising or externalising symptoms?
    b. Are there improvements in parental functioning and wellbeing?
        i. Does parents' own sleep improve?
        ii. Are there changes in symptoms of depression, anxiety, and stress?
        iii. Does parents' relationship quality improve?

Method

Measures

Secondary Outcome Measures – Children

The GARS-3 and CBCL were administered pre- and post-treatment to evaluate the impact of a BSI on children's autism symptom severity, including stereotypy, and internalising and externalising problems, respectively.

The Gilliam Autism Rating Scale – Third Edition. The GARS-3 is a 58-item informant (i.e., parent or professional) rating scale used to assess autism symptom severity and the probability that an individual is on the autism spectrum. It is designed for use with individuals aged 3-22 years. The GARS-3 includes six subscales: Restricted/Repetitive Behaviours (RRB), Social Interaction (SI), Social Communication (SC), Emotional responses (ER), Cognitive Style (CS) and Maladaptive Speech (MS; Gilliam, 2014). Informant ratings are scored on a 4-point Likert scale (0 = not at all like the individual; 1 = not much like the individual; 2 = somewhat like the individual; 3 = very much like the individual). Subscale scores are summed to yield an overall Autism Index score, with higher scores indicative of more severe symptoms and a higher probability that an individual is on the autism spectrum. Probability estimates are categorised as Unlikely, Probable or Very likely.

The GARS-3 is based on DSM-5 diagnostic criteria (APA, 2013) and the Autism Index score can be classified according to descriptors that reflect likely support requirements (Level 1 = minimal support required; Level 2 = requiring substantial support; Level 3 = requiring very substantial support; Gilliam, 2014). A severity score is not given for individuals who are scored as 'Unlikely' to be on the autism spectrum. The GARS-3 has been used extensively throughout research and clinical practice, and has high validity and reliability ratings, including internal consistency (α = .79 to .94 for the subscales), and

interrater reliability (r = .71 to .85) and test/re-test reliability (r = .76 to .90; Gilliam, 2014). The six subscale scores and Autism Index Score are reported on in this analysis.

The Child Behaviour Checklist. The CBCL is a standardised parent-report measure that assesses internalising and externalising problems in children and adolescents. Two parent-report forms were used in accordance with the age range of participants; the CBCL 1.5-5 years and the CBCL 6-18 years (Achenbach & Rescorla, 2000; 2001). For both forms, parents indicate the degree to which problem behaviours were present for their child over the past 6-months on a 3-point Likert scale (0 = not true; 1 = somewhat or sometimes true; 3 = very true or often true). Total scores on syndrome subscales combine into two major scales of Internalising Problems and Externalising Problems (Achenbach, 1991).

The CBCL 1.5-5 years consists of 100 age-relevant items; the Internalising Problems subscales include Emotionally Reactive, Anxious/Depressed, Somatic Complaints, and Withdrawn. The Externalising Problems subscales include Attention Problems and Aggressive Behaviour. A Total Problems score is the summation of the Internalising and Externalising scales and the Sleep Problems subscale. The CBCL 6-18 years consists of 113 age-relevant items; Internalising Problems subscales include Anxious/Depressed, Withdrawn/Depressed and Somatic Complaints. The Externalising Problems subscales include Rule-Breaking Behaviour and Aggressive Behaviour. The Total Problems score is the summation of the Internalising and Externalising scales, and three remaining subscales: Social Problems, Thought Problems, and Attention Problems.

For both forms, higher scores indicate greater problems. The Internalising, Externalising and Total Problems *T*-scores are interpreted as falling within a normal (i.e., non-clinical; *T*-score < 65), borderline (i.e., sub-clinical/at-risk: *T*-score 65-69) or clinical (*T*-score > 70) range. The CBCL has been used extensively throughout research and clinical

practice and has good internal consistency (α = .63 to .79) and high test/re-test reliability (*r* = .90; Achenbach & Rescorla, 2000; 2001).

Secondary Outcome Measures – Parents

The PSQI, DASS-21 and RQI were administered pre- and post-treatment to examine the impact of a BSI on parental sleep quality, mental health, and relationship quality (in two-parent households), respectively.

The Pittsburgh Sleep Quality Index. The PSQI is a 19-item adult self-report measure of sleep quality (Buysse et al., 1989). Respondents rate their sleep habits over the past month using a 4-point Likert scale pertaining to the frequency of sleep disturbance (0 = not during the past month; 1 = less than once a week; 2 = once or twice a week; 3 = three or more times a week). Seven components, including sleep quality, sleep latency, sleep duration, sleep efficiency, sleep disturbance, medication use and daytime dysfunction, are scored to give a global PSQI score (Buysse et al., 1989). The range for the Global score is 0-21, with higher scores indicative of poorer sleep; a Global score of > 5 indicates poor quality sleep (Buysse et al., 1989). The PSQI has been widely used in research and clinical practice, including with parents of children on the autism spectrum (Hodge et al., 2013; Hoffman et al., 2008; Meltzer, 2008). The PSQI has good validity (α = .83) and test/re-test reliability (*r* = 0.85; Buysse et al., 1989).

The Depression Anxiety Stress Scales – 21. The DASS-21 is a 21-item self-report questionnaire used to assess the severity of symptoms of depression, anxiety, and stress in adults (Lovibond & Lovibond, 1993). The DASS-21 is a short-form version of the DASS-42; the short-form version was used in the present research to reduce the burden of written assessment material for families. Respondents rate the extent to which they have experienced symptoms over the past week on a 4-point Likert scale (0 = never; 1 = sometimes; 2 = often;

3 = almost always). Ratings yield a total score for each of the three dimensions (depression, anxiety and stress), as well as an overall total score. Higher scores are indicative of higher psychological distress (Lovibond & Lovibond, 1993). Scores are categorised as normal, mild, moderate, severe, or extremely severe for depression (0-4 = normal; 5-6 = mild; 7-10 = moderate; 11-13 = severe; $\geq$ 14 = extremely severe), anxiety (0-3 = normal; 4-5 = mild; 6-7 = moderate; 8-9 = severe; $\geq$ 10 = extremely severe) and stress (0-7 = normal, 8-9 = mild; 10-12 = moderate; 13-16 = severe; $\geq$ 17 = extremely severe).

The DASS-21 scores can be multiplied by two to allow comparison with the DASS-42 classification system. In the present research, the DASS-21 raw scores (i.e., scores were not doubled) were interpreted in accordance with the severity categories described above. The DASS-21 has been widely used throughout research and clinical practice to assess parental wellbeing, including with parents of children on the autism spectrum (e.g., Firth & Dryer, 2013; Lai et al., 2015). The DASS-21 has good convergent and discriminative validity and internal consistency (α =. 82 to .93; Henry & Crawford, 2005).

The Relationship Quality Index. The RQI is a 6-item adult self-report questionnaire assessing relationship quality and satisfaction (Norton, 1983). Respondents rate the extent to which they agree with statements describing their relationship on a 7-point Likert scale (1 = very strongly disagree; 2 = strongly disagree; 3 = disagree; 4 = neither agree nor disagree; 5 = agree; 6 = strongly agree; 7 = very strongly agree). One item pertaining to global relationship satisfaction is rated on a 10-point Likert scale (ranging from 1 = unhappy to 10 = perfectly happy). Items are scored to yield a total RQI score (range 6- 45). Higher scores are indicative of higher relationship satisfaction, with scores $\leq$ 29 signalling relationship distress. The RQI has strong convergent and discriminative validity, and moderate to good internal consistency (α = .68 to .85; Norton, 1983).

Procedure

Parents were provided with paper versions of the questionnaires prior to commencing treatment. In two parent-households, one parent completed the CBCL and GARS-3, and both parents completed the PSQI, DASS-21 and RQI independently. Questionnaires were returned by post prior to the commencement of baseline. Immediately upon the completion of treatment, the same questionnaires were posted to parents again (i.e., the post-treatment assessment). To ensure consistency, the same parent who completed the CBCL and GARS-3 completed them post-treatment. Questionnaires were returned by post within six weeks of the end of treatment. In some cases, failure to return questionnaires or complete all questionnaire items resulted in missing data, as outlined below.

Data Analysis

Changes in GARS-3, CBCL, PSQI and DASS-21 scores were analysed using modified Brinley plots (Blampied, 2017). Modified Brinley plots are scatterplots that depict change in an individual's dependent variable data from pre- to post-intervention, viewed in a context displaying change across a group of participants (Blampied, 2017). Data are presented as coordinate pairs across two time points; pre-treatment scores on the X-axis, and post-treatment scores on the Y-axis. Individual's data points may fall on, above or below a 45^0 diagonal line. When an individual's data points lie on or near the diagonal line (i.e., X = Y), then there is little or no evidence of therapeutic change (Blampied, 2017). If they fall above or below the line (i.e., if pre- and post-treatment scores differ) this shows the degree of change (depending on the direction of therapeutic change [i.e., an increase or decrease]) proportional to the deviation from the line for each individual, as well as replication of change across participants (Blampied, 2017).

For all measures in the present study (except the RQI), the direction of therapeutic change was a reduction in scores (i.e., below the diagonal line). The RQI was not presented

on a modified Brinley plot because, owing to single parent households, the sample size was too small; instead, RQI data are presented in Table 10.5 and results are discussed separately below. Effect size measures (e.g., Cohen's d) were not able to be calculated because of the small sample size.

Results

The CBCL, GARS-3 and PSQI were administered to 8/12 participant families across Studies 1-3. These measures were not administered to the parents of Emma, Jorge, Mirasol, or Nikolay in Study 1 owing to the pilot nature of this study. Max's pre-treatment GARS-3 scores were missing, and thus he was not included in the GARS-3 analysis (n = 7). The DASS-21 was administered to 9/12 participant families, excluding the parents of Jorge, Mirasol, and Nikolay because of the pilot nature of Study 1. The RQI was administered to 5/12 families, owing to either the pilot nature of Study 1 (it was not given to the parents of Emma, Jorge, Mirasol, or Nikolay) or single parent households (it was not given to the parent of Max, Bella and Tessa).

The Gilliam Autism Rating Scale – Third Edition

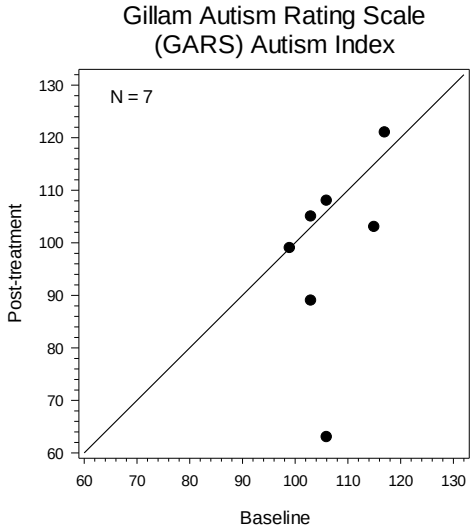
The modified Brinley plots showing children's (n = 7) Autism Index and subscale scores are presented in Figures 10.1 and 10.2, respectively, with the pre- and post-treatment GARS-3 subscale and total scores presented in Table 10.1. Results show that the Autism Index scores reduced post-treatment for three children. For two children, this change was clinically significant; Tessa and Elsie's score moved from Level 3 (Requiring Very Substantial Support) to Level 2 (Requiring Substantial Support) and Level 1 (Minimal Support Required), respectively. The third child's (Bella) score reduced but remained at Level 3. For the other children, the Autism Index score was unchanged (Eddy) or increased (Davie, James, Finn) following treatment. The RRB subscale showed a narrow range at

baseline, with three clear improvements (Eddy, Elsie, James) and one clear deterioration (Davie). The other subscales showed a wider distribution of baseline scores.

There was no clear change on the SI subscale except for a reduction for Eddy, who had the highest baseline score. On the SC subscale, two high baseline scores improved (Tessa, Bella), the low baseline score deteriorated (Elsie), and other scores showed little to no change. On the ER subscale, two scores improved (Tessa, Bella), one deteriorated (Eddy), and the rest were largely unchanged. On the CS subscale, one score improved (Bella), two deteriorated (Eddy, James), and for the rest there was marginal or no change. On the MS subscale, one low baseline score improved (Elsie), one high baseline score deteriorated (Davie), and the rest were largely unchanged.

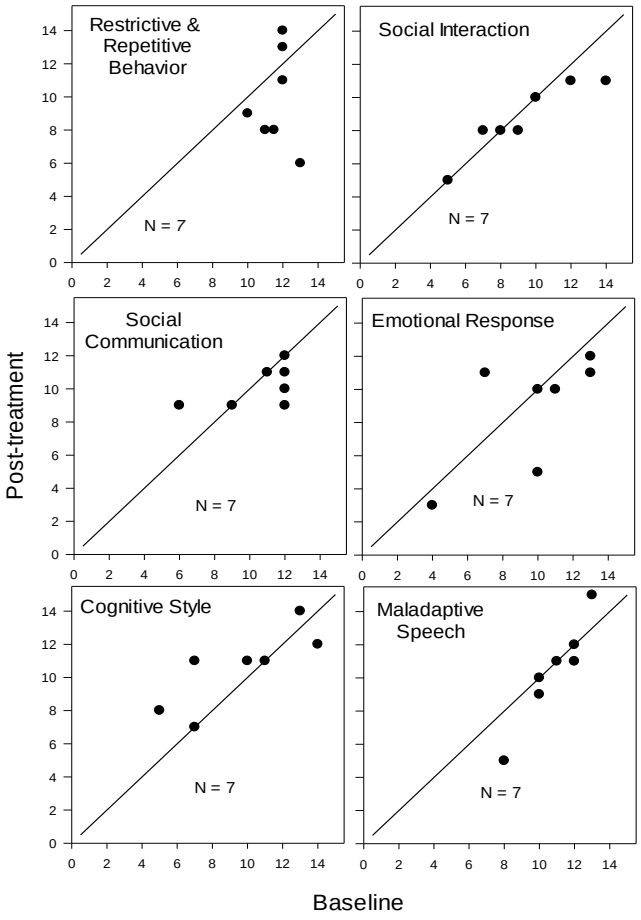
Overall, at a group level, the highest degree of change, with the highest number of replications across participants, were in restricted/repetitive behaviours and emotional responses. It is important to note, however, that as some children's scores increased following intervention, not all change was beneficial.

Figure 10.1.



Modified Brinley Plot Showing Change from Pre- to Post-Treatment on the Gilliam Autism Rating Scale –Third Edition Autism Index Scores

Figure 10.2.



Modified Brinley Plot Showing Change from Pre- to Post-Treatment on the Gilliam Autism Rating Scale –Third Edition Subscale Scores

Table 10.1. *Pre- and Post-Treatment GARS-3 Scores*

	GARS-3													
	RRB		**SI**		**SC**		**ER**		**CS**		**MS**		**Autism Index**	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Davie	12	14	12	11	11	11	13	12	10	11	13	15	117	121
Max	-	8	-	7	-	12	-	14	-	14	-	10	-	108
Eddy	11	8	14	11	12	11	7	11	5	8	10	10	99	99
Tessa	10	9	9	8	12	10	10	5	11	11	10	9	103	89*
Bella	12	11	7	8	12	9	13	11	14	12	12	11	115	103
James	11	8	10	10	12	12	10	10	7	11	12	12	103	105
Elsie	13	6	5	5	6	9	4	3	7	7	8	5	106	63*
Finn	12	13	8	8	9	9	11	10	13	14	11	11	106	108

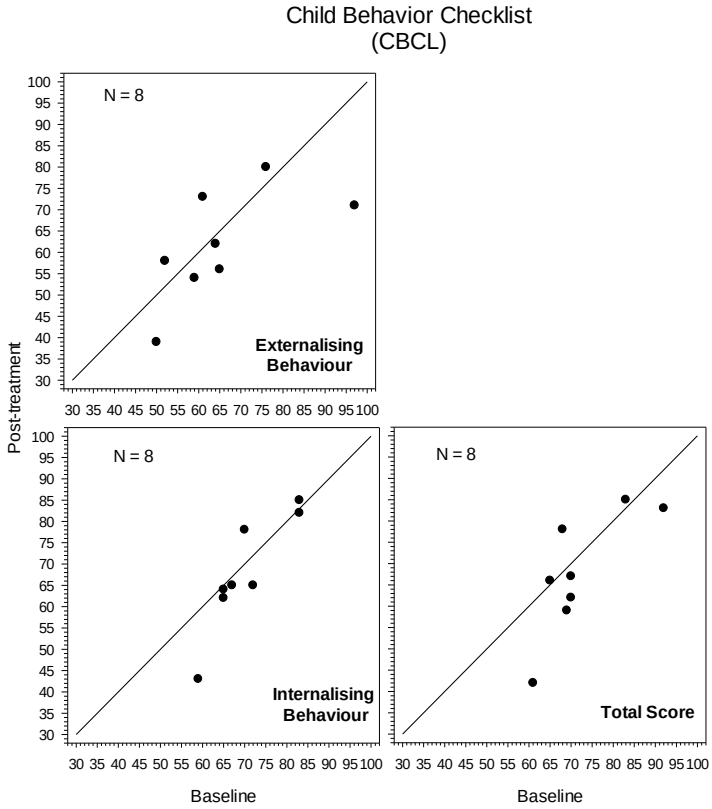
Note. CS: cognitive style; ER: emotional responses; GARS-3: Gilliam Autism Rating Scale – third edition; MS: maladaptive speech; RRB: restricted/repetitive behaviours; SC: social communication; SI: social interaction; *clinical improvement. The direction of clinical change is a reduction in scores.

The Child Behaviour Checklist

The modified Brinley plot showing children's (n = 8) CBCL scores is presented in Figure 10.3, with pre- and post-treatment scores presented in Table 10.2. The internalising, externalising and total scales showed a wide distribution of scores at baseline. On the internalising scale, there was clear improvement in two scores (Elsie, Finn), one clear deterioration (Davie) and the rest showed little to no change. Tessa showed clinically significant change as her score moved from the clinical to borderline range post-treatment, while for the other children there was no change in clinical severity.

On the externalising scale, four scores improved (Tessa, Bella, Elsie, Finn) and three deteriorated (Max, Eddy, James). For Davie, change was marginal but clinically significant. Davie and Tessa's scores moved from the clinical range to the borderline and normal range, respectively. Four total scores improved (Tessa, Bella, Elsie, Finn) with one clear deterioration (Davie), while others' scores were largely unaffected. For Tessa and Finn change was clinically significant, as their scores moved from the clinical to normal and borderline range, respectively. Overall, all children showed improvement in at least one category, except for Max whose scores deteriorated post-intervention (remaining in the clinical range).

Figure 10.3.



Modified Brinley Plot Showing Change from Pre- to Post-Treatment on the Child Behaviour Checklist Internalising, Externalising and Total Scores

Table 10.2. *Pre- and Post-Treatment CBCL Scores*

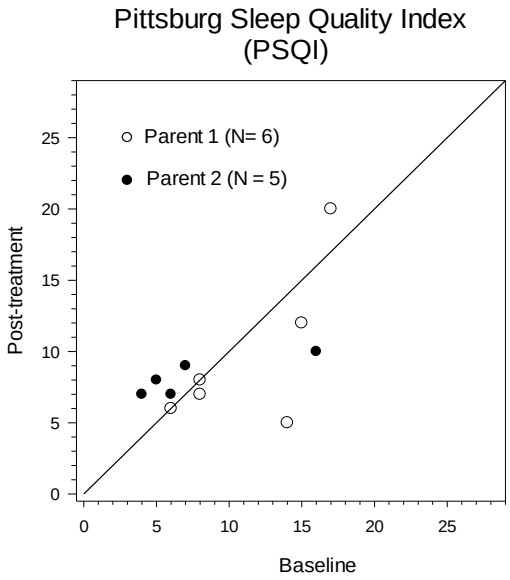
	CBCL					
	Internalising		**Externalising**		**Total**	
	Pre	Post	Pre	Post	Pre	Post
Davie	70	78	64	62*	68	78
Max	83	85	76	80	83	85
Eddy	65	64	61	73	70	67
Tessa	65	62*	65	56*	69	59*
Bella	83	82	97	71	92	83
James	67	65	52	58	65	66
Elsie	59	43	50	39	61	42
Finn	72	65	59	54	70	62*

Note. CBCL: Child Behaviour Checklist; *clinical improvement. The direction of clinical change is a reduction in scores.

The Pittsburgh Sleep Quality Index

The modified Brinley plot showing parents' (*n* = 11) PSQI Global scores is presented in Figure 10.4, with the pre- and post-treatment scores presented in Table 10.3. For Davie, James, and Elsie both parents' (parent 1 and 2) scores are presented. For Max, Eddy, Tessa, Bella, and Finn only one parent's scores are presented owing to a single-parent household or missing data. In general, a majority of parents reported poor sleep (> 5) before and after intervention. Results showed that three parents (of Max, James, and Finn) perceived their own sleep to improve following intervention, with the highest degree of change reported for James' mother (parent 1). For the parents of four children (Davie, Bella, James, and Elsie) scores deteriorated post-treatment, while the rest were largely unchanged. Change varied across and within parent-pairs, for example for James and Elsie one parent's score improved and the other parent's score deteriorated.

Figure 10.4.



Modified Brinley Plot Showing Change from Pre- to Post-Treatment on the Pittsburgh Sleep Quality Index Global Scores

Table 10.3. *Pre- and Post-Treatment PSQI Global Scores*

		PSQI	
		Pre-treatment	**Post-treatment**
Davie	**P1**	8	8
	P2	4	7
Max	**P1**	15	12
Eddy	**P1**	-	14
	P2	6	7
Tessa	**P1**	6	6
Bella	**P1**	17	20
James	**P1**	14	5
	P2	7	9
Elsie	**P1**	8	7
	P2	5	8
Finn	**P1**	-	6
	P2	16	10

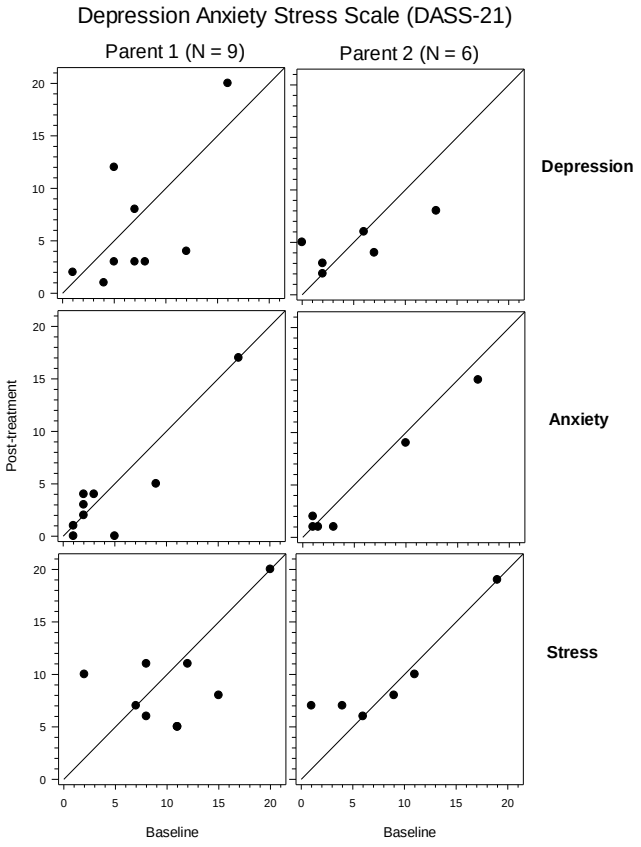
Note. PSQI: Pittsburgh Sleep Quality Index; P1: parent 1; P2: parent 2. The direction of clinical change is a reduction in scores.

The Depression Anxiety Stress Scales – 21

Parents' (n = 15) depression, anxiety, and stress scores are shown on a modified Brinley Plot in Figure 10.5, with the pre- and post-treatment scores presented in Table 10.4. For Emma, Davie, Eddy, James, Elsie and Finn, the DASS-21 was completed by both parents (i.e., parent 1 and 2), and for Max, Tessa, and Bella it was completed by the solo parent (parent 1). A majority of parents reported low anxiety scores at baseline, while the depression and stress baseline scores were more widely spread. On the depression scale, scores for 6/15 parents (of Emma, Davie, Eddy, Elsie, and both of Finn's parents) showed clear improvement, three (of Davie, Max, and Bella) showed clear deterioration, and the rest were largely unchanged. On the anxiety subscale, the scores of 3/15 parents (of Eddy and both of Finn's parents) improved, with the other scores showing little to no change. On the stress scale, clear improvements were seen for 3/15 parents (of Eddy, Tessa, and Finn), four scores (of Emma, Max and both of Davie's parents) deteriorated, and the rest were generally unchanged.

In many cases, improvement was clinically significant, with parents' scores falling into a lower category of severity post-intervention. In terms of depression, this included one parent (Finn) moving from the severe to moderate range, and five parents moving from the severe (Eddy), moderate (Davie, Elsie, and Finn) and mild (Tessa) ranges into a normal range. In terms of anxiety, one parent (James) moved from the extremely severe into the severe range, one parent (Finn) moved from the severe to the mild range, and one parent (Eddy) moved from the mild to a normal range. On the stress subscale, one parent (Eddy) moved from the severe to mild range, and three parents moved from the moderate (Tessa, Finn) and mild (Elsie) ranges into a normal range. Overall, some degree of improvement was evident for at least two parents across all domains, and results showed that in general, parents tended to report greater symptoms of depression and stress than anxiety.

Figure 10.5.



Modified Brinley Plot Showing Change from Pre- to Post-Treatment on the Depression Anxiety and Stress Scale -21 Scores

Table 10.4. *Pre- and Post-Treatment DASS-21 Scores*

		DASS-21					
		D		A		S	
		Pre	Post	Pre	Post	Pre	Post
Emma	**P1**	4	1	1	0	7	7
	P1	6	6	3	1	4	7
Davie	**P1**	7	3*	2	3	2	10
	P2	0	5	1	1	1	7
Max	**P1**	5	12	2	4	8	11
Eddy	**P1**	12	4*	5	0*	15	8*
	P2	2	2	1	1	6	6
Tessa	**P1**	5	3*	2	2	11	5*
Bella	**P1**	16	20	17	17	20	20
James	**P1**	7	8	3	4	12	11
	P2	2	3	10	9*	11	10
Elsie	**P1**	1	2	1	1	8	6*
	P2	7	4*	1	2	9	8
Finn	**P1**	8	3*	9	5*	11	5*
	P2	13	8*	17	15	19	19

Note. A: anxiety; D: depression; DASS-21: Depression Anxiety Stress Scale -21; P1: parent 1; P2: parent 2; S: stress; *clinical improvement. The direction of clinical change is a reduction in scores.

The Relationship Quality Index

Parents' pre- and post-treatment scores on the RQI are presented in Table 10.5. The scores for Eddy's parents and one of Elsie's parents were not interpretable owing to missing data. Davie's parents reported a high level of satisfaction with their relationship; this remained largely unchanged following intervention. James and Elsie's parents reported a moderate-high level of relationship satisfaction, which increased for one parent (parent 1 for Elsie and parent 2 for James) per couple. Finn's parents indicated relationship distress (≤ 29) but higher satisfaction was reported for parent 2 post-intervention.

Table 10.5. *Pre- and Post-Treatment RQI Scores*

	Parent 1		Parent 2	
	Pre	Post	Pre	Post
Davie	44	43	43	44
Eddy	39	-	43	-
James	31	32	33	36
Elsie	35	40	-	40
Finn	24	21	26	34

Note. The direction of clinical change is an increase in scores.

Discussion

This chapter evaluated whether the BSI described in Studies 1-3 produced collateral benefits for children and parents in several domains known to be associated with sleep disturbance. This included children's autism symptom severity, including stereotypy and social communication, and internalising and externalising problems (H. Adams, Matson, Cervantes, et al., 2014; Lindor et al., 2019; Richdale & Schreck, 2009; S. Cohen, Conduit, et al., 2014), as well as parental depression, anxiety, stress, relationship quality and sleep quality (Martin et al., 2019; Meltzer, 2011). The results of the BSI in Studies 1-3 indicated that improvements in at least one aspect of children's sleep occurred in 11/12 children, with the 12th child showing improvement during follow-up but not intervention.

The results of the present analysis show that clear collateral improvements occurred in overall core characteristics of autism (3/7 children), including restricted and repetitive behaviours (3/7 children), emotional responses (2/7 children) and social communication (2/7 children). Improvements were observed in externalising symptoms and total scores (4/8 children, respectively) and internalising scores (2/8 children). However, for some children on some measures post-treatment scores did not change or deteriorated (e.g., cognitive style for 2/7 children, and externalising scores for 3/8 children increased). The outcomes for most children on the GARS-3 and CBCL were mixed, showing some improvement, some deterioration, or no change across sub-domains. Three children (Tessa, Bella, and Elsie) showed consistent improvement without deterioration in scores (except for the SC subscale for Elsie). One child (Davie) showed consistent deterioration without clear improvement across any sub-domains.

For parents, personal sleep quality was generally perceived as poor, and improved in only 3/11 parents, while the scores for 4/11 parents deteriorated. Symptoms of depression improved in 5/15 parents, and symptoms of anxiety and stress improved in 2/15 and 3/15

parents, respectively. However, for some parents, scores did not change or deteriorated (3/11 parents for depression and stress, respectively). Improvement in relationship satisfaction was reported by 3/7 parents, with deterioration reported by one parent.

The findings of this chapter align with those reported in Hunter et al. (2020; Chapter 9) and McLay, France, Blampied, Hunter, et al. (2021), suggesting that improvements in children's stereotyped behaviours and internalising and externalising symptoms may occur following a BSI. Further, McLay, France, Blampied, Hunter, et al. (2021) reported improvement in children's overall symptom severity, and small improvements in parental stress, which also align with the findings of this analysis. Such findings underscore the importance of routinely assessing sleep in relation to children's daytime challenges, since sleep problems may be underpinning problem behaviour, and effectively treating pediatric sleep disturbance (Levin & Scher, 2016; Reynolds et al., 2012; Reynolds & Malow, 2011; Roussis et al., 2021). Sleep is a core mechanism for supporting adaptive functioning and behaviour (S. Cohen, Conduit, et al., 2014), thus improving sleep problems may produce a cascade of improvements across untargeted domains of functioning (Delahaye et al., 2014; Lindor et al., 2019; Richdale & Wiggs, 2005; Roussis et al., 2021; Tudor et al., 2012). Importantly, however, the finding that many children and parents' scores showed no change or worsened following treatment suggests that collateral effects do not universally occur following a BSI, that changes may vary in the types and magnitude of effects, and that change may not always be beneficial.

In this analysis, DASS-21 scores indicated high psychological distress for several parents (particularly Bella and Finn's parents), emphasising the clinical importance of assessing parental wellbeing in relation to child sleep disturbance (Johnson et al., 2018; Varma et al., 2021). Although it is not clear that psychological distress in participant parents directly resulted from child sleep problems, there is a wealth of evidence associating child

sleep disturbance with parental depression, anxiety, stress, and fatigue, particularly among mothers (Chu & Richdale, 2009; Gallagher et al., 2010; Johnson et al., 2018; Martin et al., 2019; Meltzer & Mindell, 2008; Meltzer, 2011).

Parental wellbeing is also an important consideration in understanding the factors that may contribute to the development and maintenance of child sleep problems. For example, as discussed in Chapter 1, when parent wellbeing is diminished, parents may be more likely to respond to children in ways that accidentally reinforce, rather than reduce, sleep disturbance (e.g., allowing extended screen time), or strain parent-child interactions, which may unsettle children's sleep (Johnson et al., 2018; Levin & Scher, 2016; Martin et al., 2019; Meltzer & Montgomery-Downs, 2011; Varma et al., 2021). For example, the presence of parental depression increases the likelihood that coercive parent-child interactions may develop and be maintained (G. Patterson, 1982). Further, implementing a behavioural intervention is likely to be more challenging for parents if their coping is reduced, particularly if there is also an absence of partner or family support (e.g., a solo parent with other children; Carnett et al., 2020; Meltzer, 2016). These reasons add to the importance of assessing parental sleep and wellbeing when assessing children's sleep and to the importance of improving children's sleep, as this may provide a means to improve parents' own sleep quality and wellbeing.

The mechanisms underlying post-intervention improvements for children and parents are unclear, and not necessarily linked to improved sleep. For example, as discussed in Hunter et al. (2020; Chapter 9), parents may learn to generalise behavioural management strategies from the sleep context to daytime behaviours. Further, sleep outcomes and collateral effects across participant children varied, and it is not clear how these variables may be interlinked, for example, whether improving particular sleep problems (e.g., SOD or NWs) may affect specific areas of daytime functioning (e.g., internalising or externalising symptoms). In this analysis, DASS-21 scores revealed changes in symptoms of depression,

anxiety, and stress for many parents, whilst changes in parent ratings of personal sleep quality were minimal, despite many parents indicating poor personal sleep at baseline. This suggests that parents' own sleep quality is unlikely to be the only mechanism through which parental wellbeing may be improved. The reasons why some scores deteriorated post-treatment are also unclear and may reflect external stressors on parents and families.

Results of the present analysis found that clear improvements in stereotypic behaviour occurred in three children post-intervention. A limited number of studies have examined the effects of a BSI on autism symptom severity, including stereotypy, with mixed results. Several studies have assessed change in children's restricted and repetitive behaviours using the RBS-R (Malow et al., 2014; Loring et al., 2018), the Aberrant Behaviour Checklist (ABC; Malow et al., 2014; McCrae et al., 2020) and Parental Concerns Questionnaire (PCQ; Reed et al., 2009), as well as the GARS-3 (McLay, France, Blampied, Hunter, et al., 2021). All studies reported some level of improvement except for Loring et al. (2018), who found no change on the RBS-R following a sleep intervention. The range of measures used across studies to assess autism symptom severity may (partially) account for different findings, as some measures assess symptoms of autism more thoroughly than others (Veatch et al., 2017), and may therefore be more sensitive to detecting change. For example, the GARS-3 is an autism-specific measure, while the PCQ and ABC include only single items related to stereotypy. The RBS-R does not include any items that assess vocal stereotypy (Chebli, 2016), which is a primary reason why the GARS-3 was selected over the RBS-R to assess autism symptom severity in children in this thesis.

In general, results across the aforementioned studies as well of the results of the present research indicate that improving children's sleep may reduce children's stereotypy, providing a promising avenue for future research. Future research could also help to determine whether reduced levels of (daytime) stereotypy also reduce children's sleep-related

stereotypy, and subsequently, their sleep problems. Such findings would help to shed light on the broader, bidirectional nature of the relationship between sleep and stereotypy (i.e., across settings of day and night).

Limitations of this study include the small sample size (particularly as not all children and parents were administered all measures) and a lack of secondary sources of data to triangulate parent-report (e.g., from the child's teacher). There is a potential for parental bias in perceiving and reporting improvements in other problems as a reflection of perceived improvements in children's sleep (i.e., a Halo effect; McLay, France, Blampied, Hunter, et al., 2021). Therefore, results should be interpreted with caution. Importantly, it is not possible to determine whether changes in children's sleep directly resulted in collateral child and parent improvements. It is important that future research investigates the nature of the relationship between treating children's sleep problems and the range of possible secondary effects, including the mechanisms underlying this relationship, and the reasons why inconsistencies may occur in the range and magnitude of collateral effects.

Finally, further research is also needed employing a wider range of measures than those used within this thesis, as changes in other untargeted areas (e.g., child quality of life) may have occurred but gone unmeasured. Collecting observational data is also important to detect change in children's behaviour that may not be detected by pre/post psychometric assessment measures (McLay, France, Blampied, Hunter, et al., 2021). Further, the duration between pre- and post-treatment assessment may affect the range and magnitude of collateral effects observed; it is possible that collateral changes take time to emerge or may be short-term in their effect. Therefore, it is important that future research assess the types and durability of collateral effects across a range of short- and long-term follow-up periods. The overall findings of this chapter in relation to the findings of Chapter 9 are discussed in the next chapter.

Chapter 11

General Discussion

This Chapter discusses the overall findings relating to each of the research questions in the research covered in this thesis, including the clinical implications of these findings and directions for future research. First, this chapter discusses the effectiveness of the function-based interventions to treat sleep problems and sleep-related stereotypy in participant children, and parent perceptions of the acceptability of the selected treatments (Studies 1-3). This chapter then examines the overall findings regarding the nature of stereotypy in relation to sleep disturbance, including characteristics of behaviour (Study 4), and the potential relationship between stereotypy and sleep disturbance (Studies 3 and 4). Following this, the collateral effects of BSI as identified in the systematic review of the existing literature (Chapter 9) and in relation to the outcomes of Studies 1-3 are discussed. Finally, limitations of the present research are considered, as well as a conclusion formed from the results.

Sleep is essential to our overall functioning and wellbeing (S. Cohen, Conduit, et al., 2014). For children on the autism spectrum who experience high rates of sleep problems, which can be chronic and severe, the negative effects of poor sleep can be extensive, negatively affecting physical, psychological, emotional, and behavioural wellbeing, as well as autism symptom severity including stereotypy (Jan et al., 2008; Phillips et al., 2020; Richdale & Schreck, 2009; S. Cohen, Conduit, et al., 2014; Schreck et al., 2004; Spruyt & Curfs, 2015; Tudor et al., 2012). Further, sleep problems in children do not occur in isolation and can affect family members, particularly parents who attend to children to manage sleep problems (Chu & Richdale, 2009; Hoffman et al., 2008; Johnson et al., 2018; Kodak & Piazza, 2008; Martin et al., 2019; Meltzer, 2008; Meltzer & Montgomery-Downs, 2011). It is widely agreed that the causes of sleep problems in children on the autism spectrum are multifactorial, involving biological, psychological, social, medical, and environmental factors, either alone

or in combination (Cortesi et al., 2010; Mazzone et al., 2018; Richdale & Schreck, 2009; S. Cohen, Conduit, et al., 2014). Inclusive within these contributing factors, sleep problems often have a behavioural basis (Blampied & France, 1993). Extant research shows that behavioural interventions can effectively reduce sleep disturbance in TD children, but there is comparatively little research investigating the efficacy of BSI for children on the autism spectrum (McLay, France, Blampied, van Deurs, et al., 2021; Meltzer & Mindell, 2014; Rigney et al., 2018; Spruyt & Curfs, 2015).

In recent research investigating the efficacy of BSI in children on the autism spectrum, FBA is increasingly being used to guide the selection of individualised intervention strategies. For children on the autism spectrum, FBA may reveal stereotypy as a core characteristic of autism contributing to sleep problems (Mazzone et al., 2018; Reynolds & Malow, 2011). It is currently unclear whether established BSI, which predominantly involve techniques known to be effective for TD children, can effectively reduce sleep-related stereotypy, or whether additional strategies or adaptations are required to ensure BSI are responsive to the needs of children on the autism spectrum (Pattison et al., 2020; Phillips et al., 2020; Spruyt & Curfs, 2015). Further, little is known about the characteristics of sleep-related stereotypic behaviours in children on the autism spectrum, including whether and how stereotypy may contribute to sleep disturbance (Hundley et al., 2016).

The primary aims of the present research were three-fold, namely: (1) to investigate the effectiveness of function-based BSI to treat stereotypy in the context of sleep problems in children on the autism spectrum, and to examine the maintenance of treatment effects and acceptability to parents of the selected treatments; (2) to investigate whether and how stereotypy may contribute to sleep disturbance, and the characteristics (e.g., type, topography, function) of sleep-related stereotypy across individuals; and (3) to examine whether treating

sleep problems produces collateral benefits for children and parents, including change in children's daytime stereotypy.

The three empirical studies reported on in this thesis included 12 children (5 girls and 7 boys aged 3-10 years), all with formal diagnoses of ASD. A fourth, qualitative study involved thematic analysis of the clinical assessment reports of 15 children on the autism spectrum who engaged in stereotypy in the sleep context (aged 3-15 years; including the children in Studies 1-3). Collectively, the outcomes of this research provide preliminary evidence that function-based BSI, involving a variety of antecedent- and consequence-based strategies, can effectively reduce sleep-related stereotypy and sleep problems in children on the autism spectrum, and demonstrate the social validity and feasibility of the selected strategies within the sleep context. Further, results suggest that treatment effects are generally maintained over time (up to 40 weeks post-treatment). The outcomes of this research also help to shed light on the nature of stereotypy in relation to sleep disturbance, including highlighting potential ways in which stereotypy may affect children's sleep.

Study 1 (pilot study) demonstrated the utility of FBA to target BSI to the underlying causes of sleep problems and underscored distinctive challenges that arise in treating sleep-related stereotypy where reinforcement is automatic and not amenable to direct, external manipulation. Study 2 demonstrated that BSI, including procedures that manipulated the MOs for sleep and for stereotypy, effectively reduced sleep-related stereotypy and sleep problems in all three children. Study 3 revealed mixed results for treatments targeting stereotypy in isolation, including that ambient white noise did not reduce vocal stereotypy for one child, and showed that stereotypy may not interfere with sleep for all children. Study 4 characterised the types of stereotypic behaviours performed by children at night and revealed that stereotypy may differentially affect sleep across and within children. Study 4 also highlighted several ways that parents may manage stereotypy when it occurs in the sleep

context. Finally, an examination of the collateral effects resulting from BSI in the research literature, and from Studies 1-3, suggested that BSI may produce a number of beneficial, untargeted effects for children and parents, including in children's daytime stereotypy. Given there are limitations (discussed below) of the studies in the present research, however, results are tentative and await further direct and systematic replication. The findings of this research are discussed in depth below.

The Effectiveness of Function-Based Behavioural Interventions to Treat Sleep Problems and Sleep-Related Stereotypy in Children on the Autism Spectrum

Function-based BSI were used to treat sleep problems in 12 children; of these children, 10/12 presented with sleep-related stereotypy, and stereotypy was a dependent variable for 7/12 children. For three children, stereotypy was not measured independently of other sleep-related variables (Mirasol and Davie in Study 1), or insufficient data prevented the evaluation of treatment effects on stereotypy (Finn in Study 3). The overall effectiveness of the selected treatments on children's sleep problems and sleep-related stereotypy are discussed below.

Effectiveness of the Function-Based Interventions on Sleep Problems

Improvements in at least one aspect of children's sleep were evident for 11/12 children following treatment. This included a reduction in SOD (9/11 children), CCs (7/7 children), and in the frequency (7/10 children) and duration (7/10 children) of NWs. Improvement in NWs was also observed for one child (Tessa); however, this change could not be attributed to treatment. Additional treatment outcomes included a reduction in unwanted co-sleeping (2/2 children, with co-sleeping eliminated in one family), night-time breast-feeding (1/1 child), and independent sleeping (1/1 child). Sleep problems across children were reduced with moderate-large treatment effects. For the 12th child (Max),

improvements were not observed during treatment, however, a reduction in SOD and NWs was reported during follow-up.

Treatment effects across children were found to be largely maintained at follow-up, except for three children for whom SOD (but not other sleep variables) deteriorated, and one child for whom CCs deteriorated. Conversely, for some children, further improvement in sleep-related variables was evident during follow-up compared to the treatment phase. This highlights the possibility that some effects may take longer to emerge and underscores the importance of assessing the durability of treatment effects over time (e.g., at long-term follow-up). The overall results of Studies 1-3 suggest that BSI informed by FBA can effectively reduce a range of sleep problems (e.g., SOD, CCs, NWs, unwanted co-sleeping) for children aged 3-10 years, with differing nationalities, communicative abilities, and co-occurring conditions.

These findings are concordant with the expanding number of studies demonstrating the efficacy of function-based BSI to treat sleep problems in children on the autism spectrum (e.g., Didden et al., 2002; Freidman & Luiselli, 2008; Jin et al., 2013; McLay, France, Blampied, van Deurs, et al., 2021; McLay, France, Knight, et al. 2019; P. Moore, 2004; van Deurs et al., 2019; Weiskop et al., 2001; 2005), and the effectiveness of BSI irrespective of age, type of sleep problems or autism characteristics (Carnett et al., 2020; Cuomo et al., 2017; Keogh et al., 2019; Pattison et al., 2020; Phillips et al., 2020; Rigney et al., 2018). A majority of studies have examined the effectiveness of BSI in children aged 5-13 years (Phillips et al., 2020; Spruyt & Curfs, 2015), with fewer studies focussing on children under five years (K. Turner & Johnson, 2013). The results of the present research demonstrate the feasibility and preliminary effectiveness of function-based BSI in younger children (7/12 children were $\leq$ 5 years). It is important to know whether BSI are effective in younger children because early

intervention is critical to preventing long-lasting sleep problems in children and families (K. Turner & Johnson, 2013; Phillips et al., 2020; Scantlebury et al., 2018).

Of some note, BSI were particularly well suited to the participant children who predominantly had limited verbal ability, by using non-verbal methods to alter behaviour (K. Turner & Johnson, 2013; Kodak & Piazza, 2008; Richdale & Wiggs, 2005). This emphasises the utility of BSI to change children's sleep-related behaviour without requiring verbal or cognitive mediation, given that such skills may not be developed in children on the autism spectrum who are of a younger age and/or have limited cognitive ability and/or limited/no-verbal ability (Lickel et al., 2012).

There are both strengths and limitations of interventions informed by FBA. The individualised nature inherent to function-based interventions increases the likelihood that treatment will be effective for an individual but limits its generalisability in that treatments cannot be prescribed across all or most cases, nor implemented with only moderate levels of clinician monitoring and support. It may be the case, however, that some function-based treatments are generally likely to be effective when certain sleep-related problem behaviours are indicated in FBA; for example, that sleep restriction may effectively reduce sleep-related stereotypy regardless of its type and/or function(s). Further research is needed to help to translate function-based treatment research into routine clinical practice.

All 12 children presented with multiple sleep problems, including SOD and/or NWs. This is in line with research showing sleep problems in children on the autism spectrum frequently co-exist and commonly include difficulties with sleep onset and maintenance (Cortesi et al., 2010; Cotton & Richdale, 2006; Liu et al., 2006; Reynolds & Malow, 2011; Richdale & Schreck, 2009; Rigney et al., 2018; Roussis et al., 2021; Singh & Zimmerman, 2015; Souders et al., 2017). FBA in Studies 1-3 showed antecedent factors contributing to sleep problems across children included: (a) poor sleep hygiene (e.g., the absence of or a

dysfunctional bedtime routine); (b) insufficient sleep pressure (e.g., owing to late and inconsistent wake times and/or daytime naps); and (c) inconsistent and/or inappropriate S^D for sleep (e.g., parental presence during SOL, a bedroom TV). Consequences for sleep-interfering behaviour included positive (e.g., parent attention, tangible items), negative (e.g., escape from the demand to go to sleep) and automatic reinforcement, often in combination. These findings are consistent with extant research showing that sleep problems in children on the autism spectrum are often multiply determined and require multimodal, individualised treatment (Didden et al., 2002; Goldman et al., 2012; McLay, France, Blampied, van Deurs, et al., 2021; Spruyt & Curfs, 2015).

In accordance with FBA, the primary objectives of treatment across participant children were to: (a) improve stimulus control for sleep; (b) ensure that children's physiological state at bedtime was sleep-conducive, including that (i) motivation to initiate and maintain sleep was high (i.e., with sufficient sleep pressure) and (ii) competition from other motivational states was low; (c) to disrupt the contingency between sleep-interfering behaviour and its reinforcers; and (d) teach and/or strengthen behavioural quietude as an adaptive replacement behaviour (Blampied, 2013a; Hanley et al., 2014; Kodak & Piazza, 2008; McLay, France, Blampied, van Deurs, et al., 2021; Pattison et al., 2020; Rigney et al., 2018).

A range of antecedent- and consequence-based interventions were used to address sleep problems, including improved sleep hygiene practices (e.g., a consistent and calming bedtime routine), environmental modifications (e.g., removal of lightbulbs or toys from the bedroom), visual cues (e.g., a Gro-clock^{TM} to provide salient S^D for sleeping or waking), social stories, a bedtime pass, sleep/wake re-scheduling, the faded bedtime procedure, unmodified and modified extinction procedures (e.g., faded parental presence, planned ignoring) and positive reinforcement. These techniques were largely used without alteration

to the established procedures but were tailored to suit individual child/family needs and preferences (e.g., utilisation of children's special interests within social stories). In order to address automatically maintained stereotypy, additional strategies were required within the function-based treatments, which are discussed below.

Effectiveness of the Selected Function-Based Interventions on Sleep-Related Stereotypy

Sleep-related stereotypy reduced in 6/7 children following treatment, and effects were maintained for each of the five children for whom stereotypy was assessed at follow-up. For the seventh child (Max), the frequency and duration of stereotypy worsened during treatment. Although the reasons for this are unclear, it is possible that the removal of a bedroom TV increased the salience and desirability of stereotypy in the absence of stimulation from the TV. Another consideration is that Max's parent reported that his anxiety about attending school increased during intervention. Anxiety may contribute to sleep disturbance in autism (Mazurek & Petroski, 2015; Richdale & Baglin, 2015), and anxiety may drive stereotypic behaviour (Schreck, 2021; Russell et al., 2019). It is possible that Max's stereotypy increased in relation to increased anxiety regardless of (or in addition to) changes to the bedroom environment. These findings strongly suggest there are several complexities that require further investigation to improve our understanding of sleep-related stereotypy, including that care must be taken to avoid inadvertently increasing stereotypy when targeting other aspects of children's sleep. It also emphasises that the role of children's emotions in relation to sleep problems is an important consideration within FBA (Blampied, 2013a).

For all children, FBA indicated that stereotypy was likely to produce automatic reinforcement because it typically occurred when children were alone in their bedroom. This hypothesis was developed in accordance with the research literature, which suggests behaviour is likely to be automatically reinforced when it persists in the absence of social consequences (Akers et al., 2020; Iwata et al., 1982; 1994; Kennedy et al., 2000; Querim et

al., 2013; Rapp & Lanovaz, 2016). In four children, a secondary social function was hypothesised because stereotypy also frequently resulted in parent attention (e.g., a parent would lie down with the child) and thus the possibility that stereotypy served a social function could not be excluded. This finding is in line with research literature suggesting that children's stereotypy may serve multiple functions, including both non-social and social consequences (Kennedy et al., 2000; S. Patterson et al., 2010; Scalzo et al., 2015; Wilke et al., 2012).

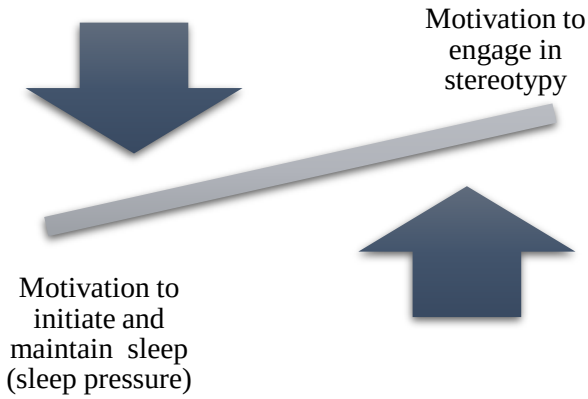
Interventions used to treat sleep-related stereotypy were selected based upon: (1) what is effective for treating daytime stereotypy and effective for sleep problems; (2) what it is feasible to implement in the sleep context; and (3) the outcomes of FBA. Antecedent-based interventions involved sleep restriction including the faded bedtime procedure (5 children) and a set wake time (3 children) as an EO for sleep, while pre-bedtime satiation (5 children) and white noise (4 children) were used as an AO for stereotypy. Modified and/or unmodified extinction procedures were used to disrupt the contingency between stereotypy and its putative reinforcers (7 children). This included removing access to items associated with sleep-related stereotypy (3 children), a faded parental presence procedure and/or planned ignoring to remove putative social consequence for stereotypy (4 children), and sensory extinction (1 child). Social stories and/or positive reinforcement were used alongside extinction procedures to model and strengthen an adaptive replacement behaviour (e.g., lying quietly in bed) for stereotypy (Cunningham & Schreibman, 2008; Lanovaz et al., 2013; Lydon et al., 2017). Function-based treatments for stereotypy are commonly multimodal, as treatments that combine antecedent- and consequence-based strategies are found to be more effective at reducing stereotypy than use of these strategies alone (Akers et al., 2020; DiGennaro Reed et al., 2012; Mulligan et al., 2014).

Sleep Restriction and Pre-Bedtime Satiation. The multicomponent nature of the BSI (including strategies targeted to treat stereotypy) for four children (Emma, Eddy, Tessa, and Bella) means it is not possible to determine whether stereotypy was directly reduced, or whether sleep problems diminished which indirectly decreased stereotypy. For example, pre-bedtime satiation and sleep restriction procedures were used in conjunction for five children to both increase their motivation for sleep (i.e., via mild sleep deprivation) and decrease their motivation to engage in sleep-related stereotypy (i.e., via satiation). It is possible that the sleep restriction procedure effectively removed the opportunity for children to engage in stereotypy, because the time that children spent in bed awake was greatly reduced (i.e., that sleep restriction indirectly treated stereotypy).

Studies show that sleep restriction procedures can effectively treat multiple sleep problems simultaneously, including SOD, NWs, and sleep-interfering behaviours (Kodak & Piazza, 2011; Piazza & Fisher, 1991a; 1991b; Piazza et al., 1997) which may include stereotypy. Sleep restriction may have utility in treating stereotypy because children's motivation to engage in sleep-related stereotypy is likely to be intimately affected by the degree to which children are motivated to enter into and sustain sleep. As discussed in Chapter 1, a child who is put to bed too early (i.e., before they are 'ready' to go to sleep) may struggle to fall asleep (K. Turner & Johnson, 2013) and be more likely to engage in sleep-interfering behaviour, which may further delay sleep. This is also likely to result in stimulus control problems, where bedroom-related S^D become associated with a state of arousal rather than sleep, making the onset of sleep more difficult (Blampied & Bootzin, 2013; Mindell & Owens, 2015). Viewed in this way, stereotypy may be understood as a motivational state problem, in that it may be particularly likely to occur when reinforcers associated with stereotypy are salient, desirable, and more immediately available than the delayed reinforcement of sleep (Blampied, 2013a; Blampied & Bootzin, 2013). This underscores the

importance within the FBA of considering the motivational processes underlying children's behaviour when assessing and treating sleep-related stereotypy and sleep problems (see Figure 11.1).

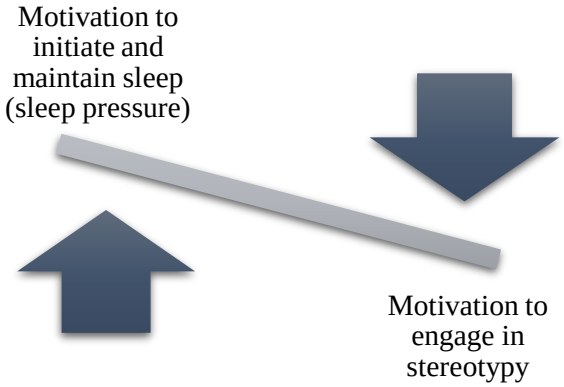
Figure 11.1.



Sleep-related stereotypy may be more likely to occur when children's motivation for sleep is insufficient

If MOs are identified in the FBA as contributing to stereotypy and sleep problems, then any intervention needs to ensure that motivation to go to sleep (sleep pressure) peaks at the scheduled bedtime (e.g., by sleep/waking scheduling or sleep restriction procedures), and that competition from other motivational states is low (e.g., by allowing them to be satiated prior to bedtime; see Figure 11.2). Indirectly treating stereotypy with sleep restriction procedures may offer a feasible means to reduce stereotypy that is particularly challenging to manipulate, such as vocal stereotypy. For example, James' vocal stereotypy did not reduce when directly targeted with white noise (discussed below) but was eliminated concomitantly with the eradication of NWs (i.e., treatment appeared to remove the opportunity for him to engage in vocal stereotypy).

Figure 11.2.



A function-based behavioural intervention may include procedures that manipulate the motivating operations for sleep to ensure a child's physiological bedtime state is sleep-conducive

Further, sleep restriction procedures may improve stimulus control for sleep by strengthening the bedroom environment as S^D for sleep (Bootzin, 1977; Piazza & Fisher, 1991a; 1991b) and enabling children to experience falling asleep without stereotypy. For instance, Bella's stereotypy remained reduced despite her SOL increasing at follow-up, which suggests it is possible she learned to change her behaviour during the sleep onset period. The reinforcement of sleep may strengthen behavioural quietude over time, if children are regularly able to fall asleep without, or with minimal, engagement in stereotypy. Further, procedures that manipulate the MOs for sleep maybe particularly feasible for the treatment of stereotypy in the sleep context because they do not require moment-by-moment parent control after a child is bid goodnight.

It is noteworthy that the faded bedtime procedure and pre-bedtime satiation seemed to complement one another in terms of the feasibility of implementation of these strategies. The extended period before bed (i.e., owing to the delayed bedtime) provided opportunity for children to receive pre-bedtime access to putative reinforcement for sleep-interfering behaviours including stereotypy. It is not possible to determine however, if and by how much

children were satiated prior to bed, and satiation may well have varied across and within children. For instance, Eddy was encouraged by his parents to engage in stereotypy prior to bed, but the extent to which he did is unclear. In comparison, parent-report suggests that Finn used his trampoline on a nightly basis for 20-30 min prior to bed. It may therefore be assumed that satiation is more likely to have occurred for Finn than for Eddy. It is also possible that fatigue from exercise, rather than satiation, had an abolishing effect on sleep-related stereotypy (Lang, Koegal, et al., 2010; Rapp & Lanovaz, 2016). The frequency, intensity, and duration of children's pre-bedtime engagement in stereotypy may have also differed on different nights, possibly producing a different magnitude of effects on stereotypy. The research literature for daytime stereotypy and the study by Jin and colleagues (2013) suggests that AOs (e.g., satiation or exercise) may be an effective means to reduce subsequent engagement in stereotypy. Thus, the mechanisms by which EO and AO may affect sleep-related stereotypy, and the potential magnitude of effects, warrants further investigation.

It is also possible, but unable to be verified (because stereotypy was only monitored in the bedroom), that children's stereotypy was displaced across settings (e.g., occurring in the lounge rather than the bedroom; Lanovaz et al., 2013), rather than being reduced absolutely. For example, Emma's magazines were relocated to the lounge where she was able to freely engage with them outside of her bedroom. As noted in Chapter 5, teaching children the parameters (e.g., timing and location) under which is it acceptable to engage in stereotypy may be a more reasonable goal than eliminating stereotypy (Akers et al., 2020; Hundley et al., 2016). Further, enabling children to engage in stereotypy in settings other than the bedroom is an important consideration given that stereotypy may serve a beneficial purpose for some children (e.g., to be calming and enjoyable; McCarty & Brumback, 2021). Allowing

children to engage in stereotypy sometimes may benefit their ability to refrain from the behaviour at other times.

White Noise (Matched Stimulation). White noise was used as a form of matched stimulation to target vocal stereotypy for Mirasol and Davie (Study 1), and James and Finn (Study 3). It was hypothesised that white noise would have a satiating effect on stereotypy and thus reduce children's motivation to engage in this behaviour (i.e., an AO), and that it would provide a consistent S^D for sleep. In comparison to pre-bedtime satiation, white noise was used continuously in the child's bedroom overnight. The ease with which ambient white noise can be implemented overnight in the home setting made it an attractive treatment option (Forquer & Johnson, 2005; K. Turner & Johnson, 2013; Knight & Johnson, 2014; Rosalez et al., 2020). White noise did not directly affect the duration of James' vocal stereotypy, nor other sleep variables. Interestingly, however, his parents anecdotally reported that white noise reduced the intensity (i.e., volume) of his vocalisations, which suggests it is possible that some beneficial effects occurred that went unmeasured. As discussed in Study 3, there are several reasons why white noise may have been ineffective, including that satiation did not occur, or that vocal stereotypy was reinforced by consequences other, or in addition to, auditory stimulation (e.g., the content of vocalisations relating to an enjoyable memory).

The inability of VSG to capture audio was a limitation of the present research that meant that the effects of white noise on vocal stereotypy for Davie, Mirasol and Finn were unable to be determined. Despite this limitation, white noise appeared to reduce sleep problems including the duration of NWs for Davie, and (to further reduce) SOD and CCs for Finn. One possibility is that white noise reduced children's vocal stereotypy allowing them to initiate and/or resume sleep more efficiently, however, this cannot be verified. Alternatively, as discussed in Study 1, it is possible that parents became less aware of children's wakeful

behaviour (e.g., Davie's parents closed their bedroom door when white noise was in use) and therefore were less able to report it.

It is interesting to note that for Finn, white noise and the trampoline (i.e., AO procedures) appeared to reduce his SOD and CCs, while the faded bedtime (i.e., an EO procedure) had little to no effect on these dependent variables. This suggests, that sleep restriction as an EO may not be equally effective in reducing stereotypy for all children. One reason for this may be that Finn experienced severe SOD, and therefore the faded bedtime procedure was likely limited in its utility to increase sleep pressure (e.g., it was not possible to delay his bedtime past his natural sleep onset owing to how late he fell asleep). Comparatively, procedures presumed to produce an abolishing effect on his stereotypy (e.g., the trampoline) may have been relatively more effective.

The use of white noise to target sleep-related vocal stereotypy in participant children was supported by the sleep as well as the stereotypy research literature. Nevertheless, the effectiveness of white noise (and other forms of auditory stimulation such as music) to reduce sleep-related vocal stereotypy and sleep problems in children on the autism spectrum requires further investigation, including the mechanisms underlying its effects. For example, it is also possible that white noise improves sleep in part by providing a consistent S^D for sleep onset and the resumption of sleep if a child wakes (Forquer & Johnson, 2005; France et al., 2018; Stores, 1996). Importantly, the results of Studies 1 and 3 showed that white noise was well tolerated by most children and parents. Although the effects of white noise for Mirasol were unable to be determined, her mother perceived it to be the most effective treatment component. Similarly, James' parents perceived white noise to improve his sleep, despite data showing that white noise had no clear impact on his sleep problems.

These findings align with existing research suggesting white noise has high social validity (Forquer & Johnson, 2005; K. Turner & Johnson, 2013; Knight & Johnson, 2014;

Rosalez et al., 2020), and that parents may perceive white noise to be effective in the absence of evidence supporting its efficacy (McLay et al., 2020). Overall, white noise did not effectively reduce vocal stereotypy for one child but did appear to reduce sleep problems in two children, highlighting the need for further investigation into its impact on sleep in children on the autism spectrum (including its impact on sleep-related vocal stereotypy), particularly as the mechanism underlying these changes remains unclear.

Extinction Procedures. Extinction procedures included the removal of items associated with sleep-related stereotypy from the bedroom (3 children); a parental presence procedure and/or planned ignoring to remove putative social consequences for stereotypic behaviour (4 children); and a sleep sack to prevent motor stereotypy from occurring (i.e., regarded as sensory extinction; one child). The removal of items associated with stereotypy were core components of the BSI for Emma and Tessa because they engaged in prolonged periods of RMO (with magazines and toys, respectively). Results showed that Emma and Tessa's stereotypy reduced following intervention, although the specific contributing effects of extinction cannot be determined owing to the multicomponent nature of the BSI for these children. It is noteworthy that for Emma, while RMO appeared to reduce, her body-rocking did not, suggesting (as expected) that extinction specifically targeted the consequences maintaining RMO but not her motor stereotypy.

For James, the removal of parent attention did not appear to affect the duration of his vocal stereotypy, suggesting that his stereotypy was unlikely to be (partially) socially maintained and supporting the hypothesis that it provided him with non-social, automatic reinforcement. This underscores the difficulty in ascertaining the specific consequences that may be maintaining children's stereotypy in the sleep context. Nevertheless, as discussed in Chapter 8, if parents consistently respond to sleep-related stereotypy then it may be necessary to eliminate social consequences for behaviour as well as addressing underlying sensory

consequences (Cunningham & Schreibman 2008). Parents may inadvertently reinforce children's 'awake' behaviour (e.g., by providing attention in response to NWs) even if their response does not impact stereotypy per se. Removing parent attention may help to reduce sleep problems and indirectly reduce opportunities for stereotypy to occur.

The sleep sack (in combination with a hug pillow and social story) appeared to reduce Elsie's stereotypy (and SOD and CCs), while the frequency and duration of her NWs remained unchanged. As discussed in Study 3, the mechanisms for this effect on stereotypy are not clear, including whether sensory extinction occurred and enabled her to fall asleep more efficiently, or whether treatment helped to reduce her SOD and consequentially removed the opportunity for her to engage in sleep-interfering (CC and stereotypic) behaviour. A reduction in SOL may have also indirectly reduced stereotypy during NWs, via improved stimulus control for behavioural quietude (i.e., she experienced and was reinforced for falling asleep without stereotypy).

Overall, the extent to which social contingencies may have partially reinforced stereotypy in children in Studies 1-3 are unclear and warrant further investigation in function-based treatment research. Given the complex nature of the stereotypy-sleep relationship, it may be the case that experimental functional analysis is required to adequately understand the relationship in any specific case, in addition to or instead of FBA. A recent review by Akers et al. (2020) on interventions for motor stereotypy in individuals on the autism spectrum recommended that a screener (Querim et al., 2013), consisting of a series of no-interaction conditions, may have pragmatic utility in determining whether stereotypy is automatically maintained or whether a full experimental functional analysis is needed. If stereotypy decreases when behaviour is ignored (i.e., in a no-interaction condition), then experimental functional analysis can be conducted to determine a putative social function (Akers et al., 2020). The results of the present research suggest that FBA may have provided an effective

screener for identifying automatically maintained sleep-related stereotypy (i.e., based on whether stereotypy persisted when children were alone in their bedroom), but that further analysis would be needed to rule out a (partial) putative social function.

As discussed in Chapter 3, there are challenges to implementing experimental functional analysis in the sleep context, including that highly controlled and structured analysis is not necessarily feasible (e.g., requiring implementation overnight) nor ethical (e.g., intentionally reinforcing problem behaviour; Blampied, 2013a; Blampied & Bootzin, 2013). Further research is required to determine how the functions of sleep-related stereotypy may be determined in the sleep context, which would inform function-based treatment. RMO in particular is likely to be maintained by extrinsic (i.e., tangible) reinforcement in addition to intrinsic (i.e., sensory) consequences, necessitating the removal of such items to disrupt external reinforcement contingencies, and allow exposure to natural sleep-controlling S^D, thus making the onset of sleep more likely (Blampied, 2013; Blampied & France, 1993; Delemere & Dounavi, 2018; Jin et al., 2013).

Social Stories and Positive Reinforcement. The use of social stories (conceived of as primarily a procedure for modelling desirable behaviour) and/or positive reinforcement within the BSI was in line with extant sleep and stereotypy intervention research suggesting it is necessary to target and strengthen an adaptive replacement behaviour wherever problem behaviour is reduced (Beavers et al., 2013; Blampied, 2013a; DiGennaro Reed et al., 2012; Hanley et al., 2014; Kodak & Piazza, 2008; Rapp & Vollmer, 2005a). This is particularly important whenever extinction procedures are used, to help to mitigate any possible undesirable side effects such as an extinction burst (DiGennaro Reed et al., 2012; Rapp & Vollmer, 2005a). Further, it is important to target and strengthen an adaptive replacement behaviour for stereotypy, given that a reduction in a targeted form of stereotypy may lead to

an increase in an untargeted form of stereotypy (Lanovaz et al., 2013; Lydon et al., 2017; Rapp & Vollmer, 2005a).

Although the specific effects of social stories on children's sleep-related behaviour including stereotypy are not able to be determined, they were generally well liked by participant parents and children, supporting literature suggesting that social stories have high social validity when used in a family setting (P. Moore, 2004; Test et al., 2011). Given that sleep disturbance is associated with diminished social communication and interaction in children on the autism spectrum (H. Adams, Matson, Cervantes, et al., 2014; Johnson et al., 2018; Richdale & Schreck, 2009; Schreck et al., 2004; Roussis et al., 2021; Veatch et al., 2017), social stories may help children to interpret social expectations related to going to bed and to sleep, including what to do if a preferred reinforcer is unavailable.

Acceptability to Parents of the Selected Function-Based Interventions

Aside from evaluating the effectiveness of treatment, it is also important to assess whether treatments are socially meaningful and acceptable to families (Callahan et al., 2017; Finn & Sladeczek, 2001; Kazdin, 2000; P. Moore, 2004; Wolf, 1978). Assessing social validity is particularly important to BSI given that parents must act as primary intervention agents within their own home. Results of the TARF-R showed a moderate-high level of treatment acceptability overall (range = 87-119/119). Parents generally viewed treatment as effective and reasonable, and indicated that procedures were easy to understand and able to be implemented at no financial cost. The level of disruption/time was viewed more moderately across parents, with some parents perceiving negative side effects of intervention and indicating they were less willing to implement treatment procedures. Interestingly, parents who perceived negative side effects of intervention also had lower treatment fidelity scores (particularly during follow-up), suggesting that social validity may play an important

role in parents' willingness to implement a behavioural treatment programme correctly and consistently.

The outcomes of the post-treatment interviews largely reflected that of the TARF-F scores, suggesting that parents generally perceived treatments to be effective and a good family fit. Many parents appreciated learning skills to change their child's behaviour and reported that they were pleased with the level of collaboration at all phases. These findings are consistent with previous research showing that BSI are valued by parents and help to increase parents' knowledge and skills to manage children's sleep-related behaviour in the home-setting (Cuomo et al., 2017; Hastings, 2002; Kirkpatrick, Louw et al., 2019; Pattison et al., 2020; Prata et al., 2018). The findings also emphasise the importance of including parents as consumers in the design of their own treatment programmes, to ensure BSI are feasible, practical, and socially meaningful to the families who benefit from them (Jin et al., 2013; K. Turner & Johnson, 2013; Kirkpatrick, Louw, et al., 2019; Moes & Frea, 2002; Sanders & Burke, 2014).

An important consideration to arise from the outcomes of the TARF-R and post-treatment interviews is the on-going requirements for parents to implement a research-based behavioural sleep programme including managing data collection in many cases over long periods of time. Treatment duration across participant children ranged from 29-106 nights/4-15 weeks (median = 49 nights/7 weeks) for those families that completed treatment. The length of time to implement treatment and carry out data recording daily was perceived less favourably by some parents. This suggests that care must be taken to streamline data collection methods to minimise parental burden, particularly in families who may be facing multiple complexities. Conversely, some parents felt the intensive intervention support was what enabled their treatment goals to be met.

Summary of the Effectiveness of Function-Based Behavioural Interventions to Treat Sleep Problems and Stereotypy and Directions for Future Research

The sleep context presents both unique challenges and opportunities for interventions directed at stereotypy. Despite the range of evidence-based strategies available for the treatment of daytime stereotypy and for sleep problems, many interventions for daytime stereotypy are not feasible for implementation in the sleep context, and many of the established BSI techniques are not suitable to target behaviour that is automatically maintained. The results of the present research provide preliminary evidence to support procedures that manipulate the MOs for stereotypy and sleep, although further research is needed to determine the mechanisms for these effects. Other strategies such as matched stimulation also warrant further investigation for the treatment of sleep-related stereotypy because they demonstrate theoretical promise, are feasible for implementation in the sleep context, and have high social validity. Future research is needed to replicate and extend the current findings as well as to investigate other evidence-based techniques that were not examined within this thesis. For example, the results of Chapter 5 suggested that RIRD (appropriately adapted for use in the sleep context) may be used to target motor stereotypy particularly during the sleep onset period but was not used for any participant children in the present research. There is also a clear need for translational research to help to generalise such findings to routine clinical practice, as discussed above.

The Nature of Stereotypy in Relation to Sleep Disturbance

Characteristics of Sleep-Related Stereotypic Behaviours

The type and topography of sleep-related stereotypy in participant children was examined using thematic analysis in Study 4 (Chapter 8). This was designed to examine the nature of sleep-related stereotypy in more depth, by drawing on parents' insights and

understanding of children's behaviour in the home setting. The results of the thematic analysis revealed a diverse range of behaviours were performed across and within children, broadly including motor and vocal behaviours, and to a lesser extent RMO. In terms of body parts, motor movements involved (a) the head, trunk, and shoulders (i.e., bouncing [on bottom], body-rocking [standing or in a prone position], body-rolling [in a supine position] and head-banging); (b) the arms/legs (i.e., flicking legs, waving arms); and (c) hands/fingers (i.e., hand-flapping, twiddling fingers; Goldman, Wang, et al., 2009). Vocal stereotypy consisted of non-word sounds (i.e., throat clearing, humming, croaking, groaning, 'babbling', non-contextual laughter, and squealing), repetition of words (i.e., numbers and letters), and scripting (i.e., television phrases, song lyrics). RMO involved toys (e.g., lining toys up), twiddling items (e.g., a pen), and mouthing (e.g., magazines) or sniffing (a scarf) objects.

Approximately 50% of the 15 children in Study 4 engaged in more than one form of stereotypy, with motor and vocal stereotypy (e.g., bouncing and squealing) being the most frequent combination. These findings exemplify the heterogeneous nature of stereotypy (Cunningham & Screibman, 2008; Rapp & Vollmer, 2005), and supported extant research suggesting that stereotypy can transverse day and night and accompany sleep problems (Jin et al., 2013; Hundley et al., 2016; Malow et al., 2006; McLay, France, Blampied, van Deurs, et al., 2021; Richdale & Schreck, 2009; Weiskop et al., 2005).

The Relationship between Stereotypy and Sleep Disturbance

Research suggests that the severity of sleep problems in children on the autism spectrum can be categorised as ranging from mild-moderate (e.g., SOD > 1 hour, and/or NWs of short duration) or severe (e.g., SOD $\geq$ several hours, prolonged NWs or EMW; Roussis et al., 2021; Sikora et al., 2012). In accordance with these distinctions, the severity of sleep problems for participant children may be considered to fall within the moderate-severe range (estimated to affect between 20-30% of children on the autism spectrum; Roussis et al.,

2021). It is noteworthy that several children's sleep problems may even be considered extremely severe; for example, Emma and Finn took up to seven hours to fall asleep, and Tessa displayed NWs of up to five hours in duration. Given the severity of sleep problems in these children, and the well-established association in the research literature between sleep problem severity and autism symptom severity (Lindor et al., 2019; Schreck et al., 2004), it is perhaps unsurprising that stereotypy accompanied sleep problems in 10/12 children in this thesis. Sleep-related stereotypy may represent a robust, but under-researched, phenomenon, and may be a particular feature of sleep disturbance in children whose sleep problems are clinically severe. Further, stereotypy (particularly RSM behaviours) is associated with younger age and lower nonverbal IQ (Goldman, Wang, et al., 2009; Hundley et al., 2016; Hollway & Aman, 2011), so knowing how to treat stereotypy in the context of sleep problems may be particularly relevant to supporting sleep in younger children, and/or those with limited verbal and/or cognitive ability (K. Turner & Johnson, 2013).

Despite the well-established association between sleep problems and autism symptom severity, little is known about the specific nature of this relationship (Mazurek & Sohl, 2016; Mazzone et al., 2018; S. Cohen et al., 2018; Sannar et al., 2018; Schreck et al., 2004). Evidence suggests the autism-sleep relationship is bidirectional and dynamic, yet the nature of these interactions is largely unstudied (Schreck & Richdale, 2020). In this thesis, a behavioural model of sleep problems (Blampied & France, 1993; see Chapter 1) was used to identify factors that may disrupt a child's sleep onset behaviour chain and contribute to the development and maintenance of sleep problems. A behavioural framework is important because it can identify factors underlying sleep disturbance that are modifiable, which therefore present critical targets for behavioural treatment.

In Chapter 5, the behavioural model was extended to accommodate autism-associated behaviours that FBA may reveal as contributing to sleep disturbance, specifically, to consider

how stereotypy may interfere with sleep. It was proposed that stereotypy has the potential to interfere with sleep onset if it inhibits a child's ability to establish and maintain behavioural quietude for a sufficient period to enable sleep onset to occur. Stereotypy may also interfere with the re-initiation of sleep following a NW. Thereby, stereotypy may contribute to SOD and/or the duration of NWs by delaying sleep onset (Hundley et al., 2016; Jin et al., 2013).

Interestingly, daytime stereotypy has been associated with specific types of sleep problems including SOD (Tudor et al., 2012), sleep fragmentation (Goldman, Surdyka, et al., 2009), and reduced sleep duration (Sannar et al., 2018; Schreck et al., 2004; Tudor et al., 2012; Veatch et al., 2017). This highlights the reciprocal nature of the interactions between sleep problems and stereotypy, in that stereotypy may both contribute to and be exacerbated by sleep problems. As another example, results of the present research showed that NWs appeared to provide opportunity for children to engage in vocal stereotypy, which seemed to delay or impede their ability to return to sleep. NWs may therefore serve as a vulnerability factor that renders children at risk of making contact with contingencies of automatic (and possibly socially-mediated) reinforcement, which in return may partially account for the development of prolonged NWs (Blampied & France, 1993).

Future research could help to determine whether increased daytime stereotypy in the presence of sleep problems increases the frequency and/or severity of children's night-time stereotypy, possibly placing children at further risk of disrupted sleep. Given that our sleep cycles are regulated by homeostatic and circadian rhythm processes over a 24-hour period, a comprehensive account of the sleep-stereotypy relationship must include consideration of day-night relationships. Longitudinal research could also help to shed light on how the sleep-stereotypy relationship may change in relation to age, particularly because the types of sleep problems (Goldman et al., 2012; Sivertsen et al., 2012) and severity of stereotypy (MacDuffie et al., 2020) that children exhibit may be subject to change as children mature. In the present

research, children performed the same forms of stereotypy on a nightly basis, with these behaviours tracked over an average period of two months. The one exception was Elsie, for whom no stereotypy was evident when the family returned to the study following a period of non-participation. The reason for this change is unclear but may reflect natural changes in behaviour over time. There is a strong need for longitudinal research from birth through adolescence to explicate the natural history of sleep and stereotypy and their interactions in children on the autism spectrum.

Stereotypy as Sleep-Interfering or Sleep-Conducive

A major finding of Study 4 was that different types of stereotypy might differentially affect sleep across and within children, underscoring the importance of using FBA to inform individualised case conceptualisations to treat sleep-related stereotypy. Vocal stereotypy and RMO were primarily classed as sleep-interfering (along with some forms of motor stereotypy, e.g., simultaneous bouncing and squealing), while in some cases, motor stereotypy appeared to be sleep conducive, predominantly when it involved repetitive movement of the head, trunk, and shoulders (e.g., body-rocking or rolling).

There may also be other topographical distinctions between stereotypic behaviours that promote versus disrupt sleep. For example, RMO occurred when participant children were out of bed, in contrast to sleep-conducive stereotypy that tended to occur when children were in bed and often in a prone position. These forms of motor stereotypy were therefore more similar to 'settling behaviour' than RMO, which may have prevented sleep occurring by virtue of the fact that children were up and out of bed. Vocal stereotypy is more perplexing however, given that it frequently occurred when children were in bed and lying down, but was predominantly viewed by parents as sleep-interfering.

Another consideration is the temporal occurrence of stereotypy in relation to sleep onset; sleep-conducive motor stereotypy occurred immediately prior to sleep onset (i.e., during the wake-sleep transition). In comparison, children often ceased sleep-interfering stereotypy for a period before falling asleep. For instance, following prolonged periods of RMO, Tessa re-made her bed on the floor each night where she would then lie quietly before falling asleep. The extent to which children ceased vocalising before falling asleep is not clear. As discussed in Study 4, sleep-conducive stereotypy may represent a learned variety of self-settling behaviour for some children (i.e., instead of behavioural quietude), which may fall under operant control, if sleep itself acts as a reinforcer.

Further, sleep-interfering RMO and vocal stereotypy were largely reported to occur during the day as well as at night, and may therefore be viewed as an extension of the same behaviour across settings (of day and night). In comparison, sleep-conducive motor stereotypy was reported to occur only in the sleep context and may therefore have served a function more specifically related to sleep (e.g., self-settling).

The potential differences between sleep-conducive and sleep-interfering stereotypy seem to be most evident for Emma and Finn. Both children exhibited severe SOD and engaged in prolonged periods of 'sleep-interfering' stereotypy (bouncing and squealing for Finn, RMO for Emma) prior to falling asleep. Overall, there may be differences in the type and topography of stereotypy that differentially affect sleep across and within children, which may include the physical location stereotypy occurs in (e.g., in bed or out of bed), its temporal proximity to sleep onset, and the degree of relatedness to daytime stereotypy.

Importantly, the degree of overlap between sleep-conducive stereotypy and RMD as discussed in Chapter 4 warrants further attention, including whether these behaviours are the same phenomenon, and of shared or distinct aetiology. It is also not clear whether sleep-conducive stereotypy relates differently to initial sleep onset compared to the re-initiation of

sleep following a NW. For example, Eddy engaged in sleep-conducive motor stereotypy during the sleep onset period but not NWs.

As discussed in Study 4, the distinction between sleep-interfering and sleep-conducive stereotypy carries important implications for BSI. Sleep-interfering stereotypy may represent a form of 'play', for which the underlying issue may be motivational (as discussed above), in that the reinforcer of stereotypy is more salient, immediate, and desirable than the delayed reinforcer of sleep (Blampied & France, 1993; Blampied & Bootzin, 2013; Rapp et al., 2017). Accordingly, treatment may focus on increasing children's motivation for sleep and decreasing children's motivation to engage in sleep-interfering behaviour.

In contrast, sleep-conducive stereotypy may represent a learned type of self-settling behaviour in some children, for which a greater focus of treatment may need to be placed on teaching sleep-facilitating replacement skills (e.g., relaxation leading to behavioural quietude). Models such as social stories may assist children's learning in this regard. Further, as previously noted, increasing children's motivation to fall asleep (e.g., via mild sleep deprivation) may ensure a more rapid sleep onset and enable children to experience falling asleep without stereotypy. An important consideration is that if children are using repetitive movement to fall asleep, then limiting or preventing movements from occurring may actually increase SOL. For example, research suggests that RMD may serve a sleep-initiation role and its absence may increase SOL in some children (Blunden & Nair, 2009; Hoban, 2003). It may be important (in addition to strengthening a replacement behaviour and ensuring a rapid sleep onset), to allow children to satiate their desire for repetitive movement prior to falling asleep, perhaps in an alternative, more adaptive manner (e.g., bouncing on a trampoline rather than in bed).

The Collateral Effects of Behavioural Sleep Interventions

Sleep plays a vital role in all aspects of daily functioning, and therefore represents a pivotal target for treatment. Given the strong associations between sleep problems and autism symptom severity, internalising and externalising problems, and parents' own sleep quality and wellbeing (Hoffman et al., 2005; S. Cohen, Conduit, et al., 2014; Schreck et al., 2004; Sivertsen et al., 2012; Tudor et al., 2012; Meltzer, 2008), it is important to assess whether BSI produce collateral benefits in such areas for children and parents. Chapter 9 presented a systematic review evaluating the empirical evidence investigating the collateral benefits of BSI in children (range = 2-18 years) on the autism spectrum. Chapter 10 presented an analysis of the collateral benefits of BSI for children and parents in Studies 1-3. In the systematic review, 10/10 studies reported improvement in at least one aspect of children's sleep, and 8/10 studies reported collateral improvement in at least one area of children's daytime functioning, including a reduction in daytime stereotypy, internalising (e.g., anxiety) and externalising (e.g., challenging behaviour) symptoms, and improved child quality of life. This study was the first to review the collateral effects of BSI in children on the autism spectrum, suggesting that treating sleep problems may positively affect children's daytime functioning and wellbeing. The results of Chapter 10 offered preliminary support of those of the systematic review and recent research (McLay, France, Blampied, Hunter, et al., 2021).

Importantly, however, Chapter 10 also showed that many children and parents' scores on relevant measures did not change or deteriorated, suggesting that treating children's sleep problems does not universally nor consistently produce untargeted benefits across children and parents. Similarly, the results of the systematic review found that collateral benefits were not uniformly reported by all studies, and the types of collateral benefits found differed. This finding is somewhat unsurprising given that children on the autism spectrum commonly vary in their response to behavioural treatments in general (Lombardo et al., 2019; Singh &

Zimmerman, 2015; Vivanti et al., 2014), and is consistent with the finding that group means may conceal instances of deterioration while implying improvement on average (Blampied, 2017). In the present research, the degree of improvement in children's sleep problems varied, including that for some children sleep problems were eliminated, for others the sleep problems were reduced, and in some cases, the sleep problems did not improve.

The mechanisms for collateral change require further investigation, including why effects may differ across children and parents, and the types and magnitude of collateral change in relation to the type and magnitude of change in children's sleep. It may be that different areas of functioning are more directly affected by sleep problems than others. Interestingly, the findings of the systematic review, the analysis in Chapter 10, and the study by McLay, France, Blampied, Hunter, et al. (2021) demonstrated improvements in children's daytime stereotypy, which may suggest that sleep problems and the severity of stereotypy are closely linked. As discussed in Chapters 9 and 10, the types of measures that studies choose to incorporate will affect the types of collateral effects that may be detected, with some measures being more sensitive to change in specific areas than others. In the post-treatment interview, many parents anecdotally reported beneficial side effects of intervention including improved morning routines, tidier and calmer households, and increased school attendance, which were outcomes that were not assessed by any psychometric measures in Studies 1-3.

As discussed in Chapter 10, it is also possible that collateral effects are time sensitive, possibly taking a long time to emerge, or they may be highly transient, and/or affected by post-treatment changes in life circumstances and experiences. Overall, the collateral effects that may occur for children and parents following sleep treatment are intriguing, and place importance on considering the role of sleep problems underlying daytime challenges.

Limitations

A number of methodological limitations may compromise the external validity of the results of the present research. First, the empirical studies in this thesis included a small number of participants, which limits the number of replications of treatment and its effects. The heterogeneity of the participants in terms of language and cognitive ability, autism symptom severity, strengths and abilities, as well as the variation in the home contexts, family situation and culture (amongst a wide range of other contextual/ecological variables), is a further limitation that means it may not be possible to relate results to other children on the autism spectrum, even when there are similar presenting complaints. However, rather than parcelling out these factors as may be done in group-based design, it is thought that the use of a case study design and FBA methodology which purposefully retained individual data allowed for a richer description of how treatment can be individualised for children with unique sets of needs across a variety of contexts. Further, the diversity that is inherent across individualised cases may enhance the generality of intervention; the broad method (i.e., function-based, individualised interventions) by which treatments were designed may have generality, even though no specific treatment can confidently be said to work with all or any specific cases.

There are also limitations arising from the design of interventions, including not being able to use a controlled single-case design such as a multiple-baseline design across all participants. While an AB approach fits with the sequential nature of function-based treatments, it cannot rule out cumulative sequence effects where a treatment effect is detected, which limits the ability to draw conclusions about the specific effects of treatment strategies.

Another limitation is the incomplete or missing data that limits the validity and reliability of the findings. Sleep diary data were collected across all participants; however,

parents did not always complete sleep diaries. Some parents were also unable to report information on dependent variables because they were unaware of behaviour when the child was not visible or audible. This was a particular problem with measuring stereotypy, because stereotypy typically occurred when children were alone in their bedroom and thus parents were often unable to report it. This difficulty is noted in the research literature; for example, NWs may occur without parental knowledge if the child does not signal they are awake (Blampied, 2013a; Jan et al., 2008; M. Moore et al., 2017; Richdale & Schreck, 2009; Sadeh, 1999).

Studies that compare parent-report of sleep outcomes with instrumented sleep measures (e.g., VSG, actigraphy) show that parents most accurately report SOL, while corroboration with other sleep problems (e.g., NWs) is less consistent (Bangerter et al., 2020; Lambert et al., 2016). Many studies rely heavily on parent report, which on its own may be subject to bias or inaccuracy (Bangerter et al., 2020; Kotagal & Broomall, 2012; Sadeh, 1999; Spruyt & Curfs, 2015; Stores & Wiggs, 1998). For example, parents may over- or under-report sleep problems because of their attributions and beliefs about a child's sleep and/or their own coping (e.g., highly stressed parents may over-report sleep problems; Bangerter et al., 2020; Richdale & Schreck, 2009). This underscores the importance of instrumentally recorded observations such as VSG to triangulate data and circumvent bias, and help to quantify treatment effects (Blampied, 2013b; Blampied & Bootzin, 2013; Pattison et al., 2020; Spruyt & Curfs, 2015).

Heavy reliance on VSG where parent-report was not possible also resulted in missing or incomplete data sets in some cases. VSG was frequently relied on to record stereotypy; when VSG was not recorded, or the child moved out of camera range, then information on stereotypy was lost. A key limitation of the present research was the inability to capture audio recordings which, given the often heavy reliance on VSG to measure stereotypy, meant that

vocal stereotypy could not be recorded. James' case presented an exception owing to his parents being consistently aware of and able to report the duration of his vocalisations (e.g., because of sleeping next to him).

The difficulty in measuring vocal stereotypy is reflected more widely across the research literature; for example, measures frequently used to assess stereotypy lack items pertaining to repetitive vocalisations (e.g., the RBS-R; Chebli et al., 2016). The early childhood version of the RBS (RBS-EC; Wolff et al., 2016) includes one item pertaining to vocal stereotypy; however, the age range of this measure limits its validity in older children. Given that up to 85% of children on the autism spectrum may engage in vocal stereotypy (Mayes & Calhoun, 2011), there is a clear need for future research to develop measures that specifically assess repetitive vocalisations. Advancements in technology may improve the ability to capture audio recordings overnight in the home setting, which would help to establish the reliability of parent-report. A further limitation of the present research was that IOA for dependent variables (including stereotypy) could not always be calculated, because of missing parent-report or VSG data.

Conclusion

Notwithstanding the aforementioned limitations, the findings of the present research offer several important contributions to the research literature. First, it demonstrates the preliminary effectiveness, maintenance, feasibility, and social validity of a variety of function-based behavioural treatments for sleep problems including preliminary evidence for strategies that may effectively reduce automatically maintained sleep-related stereotypy. Second, it illustrates the nature of sleep-related stereotypic behaviours, including the type and topography of behaviour across and within individuals, which has scarcely been documented in the research literature. Third, it offers conceptual insights into the potential role of stereotypy in relation to sleep disturbance, including that stereotypy may differentially affect

sleep across and within children, which may have differing implications for behavioural treatment. Fourth, it shows that improving children's sleep may generate beneficial collateral effects for children and parents.

Overall, the findings show that a range of stereotypic behaviours may contribute to difficulties with sleep onset and/or sleep maintenance and indicate that function-based BSI that include strategies that specifically target automatically maintained behaviour may effectively reduce stereotypy in the context of sleep disturbance. Further, improving children's sleep problems may have positive implications for their daytime functioning including autism symptom severity. Future research is needed to better understand the dynamic and multifaceted nature of stereotypy in relation to sleep problems in children on the autism spectrum, including treatment studies with larger sample sizes and robust methodology to extend the findings of the present research.

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Appendix A: Ethics Approval

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

HUMAN ETHICS COMMITTEE
Secretary, Rebecca Robinson
Telephone: +64 03 369 4588, Extn 94588
Email: human-ethics@canterbury.ac.nz

Ref: HEC 2018/47

23 July 2018

Dr Laurie McLay
Health Sciences
UNIVERSITY OF CANTERBURY

Dear Laurie

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

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

Best wishes for your project.

Yours sincerely

pp. R. Robinson

Professor Jane Maidment
Chair
University of Canterbury Human Ethics Committee

Appendix B: Parent Information Sheet

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

Information for Parents/Caregivers

This research has been assessed and approved by the University of Canterbury Human Ethics Committee (HEC 2014/150).

Dear Parent/Caregiver,

We are a group of researchers at the University of Canterbury. Dr Laurie McLay is a 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 autism 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, and Jolene Hunter, Jacqui Knight, and Jenna van Deurs are student's in Child and Family Psychology who will be working on their theses as part of this project.

We would like you to consider allowing your child with autism or Asperger's syndrome to participate in this research study. The primary purpose of this study is to investigate the effectiveness of treatments for sleep disturbance in children with autism or Asperger's syndrome. These treatment options include non-traditional approaches (e.g., white noise) as well as modified behavioural approaches. 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' experiences in using the treatments and any changes to their lives, or their child's lives, which result.

If you agree to allow your child to be a part of this study, we will meet with you to discuss your child's sleep behaviour with you and find out more about him/her and your family. This initial meeting will last for approximately 1 hour. 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 a commonly used questionnaire in order to obtain information about your child's sleep behaviour and the effects of treatment. It will take approximately 15 minutes to complete this 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 meeting which will last 1-1 ½ hours.

To help us gather further information about your child's sleep patterns we will bring a video camera to your home for some nights over the course of the programme, that is capable of recording all night sleep. This method will allow us to measure sleep behaviour at times when an adult is not present. We will demonstrate and explain how to use the video equipment for gathering information.

As a part of this study we would also like to investigate the experiences of families in implementing treatments for sleep disturbance, those treatments that they consider to be most acceptable, and the impact of successful treatment of sleep problems on parent and child wellbeing and quality of life. In order to do this we will ask you to complete some questionnaires about your 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 or in a clinic at the University of Canterbury.

When information about your child's sleep behaviour has been gathered, treatment will commence. You will be offered a choice of treatment options. The treatment will be implemented for up to four weeks. 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 non-traditional approach, or alternatively, a modified behavioural approach to treatment can be implemented. If you would prefer to use a behavioural approach from the beginning then this is also an 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 be over a period of 3-4 months.

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.

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 me:
Phone: 64 (3) 364-2987 ext. 7176
Email: laurie.mclay@canterbury.ac.nz

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

Appendix C: Child Information Sheet

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

Children's Information Sheet

Hello. My name is Jolene Hunter and I am an intern 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 your parent/s about ways to help you to sleep better. This means that I will be coming to your house, or your parent/s will be coming to see me at the University.

I will ask you to wear a special watch called an actigraph sometimes. This will help me to understand the times that you are awake and asleep. There will also be a video camera in your bedroom sometimes. This will also 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.

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

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

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

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

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

Appendix D: Parent Consent Form

Consent Form for Parents/Caregivers

This research has been assessed and approved by the University of Canterbury, Human Ethics Committee.

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 understood the information that was given to me about this study.

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.

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 someoone 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 myself and my child 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:_____

Please return this form to Jolene Hunter.

Appendix E: Child 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 _____.

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

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

I know that if I want to stop at any time or if I do not want to be a part of this project anymore, that will be fine. I can tell Jolene or my parents.

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

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 Jolene Hunter.

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

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: Flyer

Does your child have autism or features of autism, and sleep problems?

You may be eligible to receive treatment through the nation-wide Autism and Sleep Study, being conducted through the University of Canterbury

- Our research is investigating (a) the effectiveness of treatments for sleep disturbance for children with autism (b) the impact of successful treatment on parent and child well-being (c) the effectiveness of treatments for sleep-interfering repetitive behaviours

- Our research team is led by Dr Laurie McLay and Associate Professor Karyn France, and consists of Child and Family Psychology and PhD students

- Sleep treatments can include a range of strategies, including both non-traditional approaches (such as white noise) and behavioural interventions

- Treatment options will be outlined for you, and the final decision will be yours. If the approach is unsuccessful, alternative treatment options will be offered

- The research will be conducted in the family home or at a University clinic, and will be implemented by parents, with support and guidance through skype and phone calls from the researchers

If you or somebody you know might be interested in participating in this study and you would like further information, please contact:

Dr Laurie McLay
School of Health Sciences, University of Canterbury
Phone: (03) 369 3522
Email: laurie.mclay@canterbury.ac.nz
Jolene Hunter (PhD Candidate & Registered Intern Psychologist)
School of Health Sciences, University of Canterbury
Phone: (03) 366 7001 and ask for extension 3696
Email: Jolene.hunter@pg.canterbury.ac.nz

Appendix H: Sleep Diary Template

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

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
1st Nigh 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 Nigh 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 Nigh 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

Appendix I: Post-Treatment Interview Questions

- How did you find the intervention overall, and the process?
- What is it that you (both) did, that you feel made a difference?
- How do you make sense of the improvement?
- Did the child's progress/improvement have an impact on you personally, if so- how?
- What impact did it have on the child/ the rest of your family?
- On a scale of 1-5 with 5 as the worst the sleep problems could possibly be, what level of impact do you see the sleep problems as having currently (1 being 'no problem' and 5 being 'very much a problem- big impact')
- Did you notice any other changes in your child's behaviour in response to intervention?
- Any suggestions for how our process could have been improved?
- Any other comments you would like to make

Appendix J: Overview of Clinical Interview Content

Introduction

- Welcome, overview of the research team
- Limits of confidentiality
- Agenda

Child and family information

- Ethnicity/culture
- Important family members
- Typical daily routine and schooling
- Child's interests/strengths
- Medication (child)
- Sleeping arrangements in household
- Other service involvement

Sleep problems

- Type, topography
- Typical night (from dinner-breakfast)
- Sleep environment
- Behaviour while awake
- Parent response to behaviour
- Frequency/intensity/duration of problem behaviour
- Exceptions to the problem
- What makes it better/worse

History of Sleep Problem

- Onset
- Developmental and family context at onset
- Changes over time
- Past management strategies (and to what effect)

Child's Developmental History

- Life before pregnancy
- Conception

- Pregnancy
- Birth
- Postnatal adjustment
- Early years – temperament, attachment, adjustment
- Developmental milestones
- Starting preschool and school
- Physical health
- Major events
- Diagnosis

Parent relationship

- Attributions for sleep problems
- Ability to work as a team
- Ability to see eye to eye (+ re management of sleep)
- Current relationship strength

Parent wellbeing

- Current wellbeing
- Impact of sleep problems
- Other stressors
- Management strategies
- Sources of support

Set Sleep Goals

Appendix K: Supplementary Information (S1)

Children's Sleep Habits Questionnaire Scores

CSHQ total scores were available for 10/15 children in this study; the parents of one child were administered the SF-CSHQ. Results revealed that 8/10 children's scores exceeded the clinical cut-off.

Table S1. *Children's Sleep Habits Questionnaire (CSHQ) total scores at the time of pre-treatment assessment*

Participant	CSHQ Total Score
1	13 (SF)
2	40*
3	53*
4	50*
5	60*
6	37
7	52*
8	64*
9	45*
10	45*
11	-
12	-
13	-
14	-
15	-

*Note.*SF: short-form CSHQ; *Above clinical cut-off

Appendix L: Supplementary Information (S2)

Stereotypy Questions within the Clinical Interview

(a) What does the repetitive behaviour look/sound like (i.e., what does your child do?)

(b) When, where (e.g., in bed), how often, and for how long, does this behaviour occur?

(c) What do you think your child gains from this behaviour?

(d) What (if anything) prompts the behaviour to occur?

(e) How do you respond when your child does this behaviour? What effect does your response have on the behaviour?

(f) Does your child do anything else alongside this behaviour? If so, please describe.

(g) Are there other times throughout the day or night when your child does this? (e.g., NWs if not previously reported)

(h) What impact (if any) does the repetitive behaviour have on your child's ability to fall asleep/stay asleep?

(i) What impact (if any) do the sleep problems have on your child's repetitive behaviour (day and/or night)?

(j) What have you tried in the past (if anything) to disrupt the behaviour (and to what effect)?

(k) Please describe your child's daytime repetitive behaviour; how is this the same/different from their night-time repetitive behaviour?