EVALUATING THE EFFECTIVENESS OF FUNCTION-BASED BEHAVIOURAL SLEEP INTERVENTIONS FOR THE TREATMENT OF ANXIETY RELATED SLEEP DISTURBANCE IN AUTISTIC YOUTH

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Abstract

The prevalence of anxiety disorders, and sleep disturbance is elevated among autistic youth. However the ways in which anxiety, autism characteristics, and sleep disturbance interact with each other, in the context of an individual's environment and family relationships is still unclear. The reciprocal influences between these variables is diverse, and multi-faceted. Despite the relatively common co-occurrence of sleep disturbance and anxiety in autistic and non-autistic individuals, these conditions continue to be treated separately, with minimal evidence of effective integrated treatment for anxiety related sleep disturbance. A greater understanding of the bi-directional relationship between anxiety and sleep in autistic individuals, and the development of minimally sufficient treatment of anxiety-related sleep disturbance is needed. This study evaluated the effectiveness of Functional Behavioural Assessment (FBA) - informed behavioural sleep interventions for the treatment of anxiety related sleep disturbance, utilising single-case-research design. Study One is presented as a case study with one 13-year-old participant. Study Two is a single-case multiple-baseline AB design involving three participants aged 13-16 years. The effects of sleep treatment on children's anxiety, daytime behaviour, and quality of life, as well as parents' sleep quality and well-being was examined. All four participants were adolescents, with a diagnosis of Autism, and parent or self-reported sleep-interfering anxiety. Participants' interventions were informed by FBA, and individualised according to the needs of the child and their family. In Study One, treatment included relaxation strategies and faded parental presence procedures. Study Two included adjustment of participants' sleep/wake cycle for all participants, with one participant also requiring sleep hygiene modifications. Results indicated substantial improvements in sleep for all participants who completed treatment, with improvements largely maintained up to 10 weeks following treatment. Findings related to anxiety were variable, with some indication of both reductions and increases in perceived anxiety

following treatment. Positive changes were evident overall in participants' daytime behaviour, quality of life, and parent well-being post-treatment, with mixed results relating to parents' sleep quality. Parents and participants indicated that treatment was acceptable, cost effective, reasonable, and understandable. Families found treatment time-consuming, but worthwhile due to the benefits of effective treatment, and families felt well-supported by clinicians throughout the treatment process. Overall, this study provides evidence of the effectiveness of behavioural sleep interventions for treating sleep disturbance in anxious autistic youth, and highlights the utility of FBA informed interventions for providing individualised, responsive treatments, uniquely tailored to the needs of autistic individuals and their family.

Chapter 1: Introduction and Literature Review

Sleep Disturbance and Anxiety in Autistic Children

Sleep difficulties are common in autistic children, with prevalence rates ranging from 50-80% (Cuomo et.al., 2017; Devnani & Hegde, 2015; Mazurek & Petroski, 2015; Rzepecka et.al., 2011). This compares with 9-50% of non-autistic children (Cuomo et al., 2017; Kotagal & Broomall, 2012; Williams et.al., 2015). The most common types of sleep problems in autistic children include difficulty falling asleep (sleep onset delay), night awakenings, and circadian rhythm disturbances (Kotagal & Broomall, 2012; Mazurek & Petroski, 2015; Richdale & Shreck, 2009). Poor sleep efficiency (time asleep compared to time spent in bed), unwanted co-sleeping, and early waking are also frequently reported (Cuomo et.al., 2017).

Untreated sleep problems are likely to continue into adulthood (Herrmann, 2016). Approximately two thirds of children experience ongoing sleep issues, if left untreated (Cuomo et.al., 2017; Hodge et.al., 2014). Persistent sleep problems can negatively affect children's daytime functioning, cognitive development, learning, and overall health (Leahy & Gradisar, 2012, Herrmann, 2016). Research has demonstrated a strong link between sleep problems and anxiety in non-autistic children, and autistic children (Mazureck et.al., 2015; Nadeau et.al., 2015; Rzepecka et.al., 2011). In fact, Hollway and colleagues (2013) suggested anxiety was the strongest predictor of sleep problems in autistic children.

The relationship between sleep disturbance and anxiety is complex; a dance between the high states of arousal characteristic of anxiety, and the lower states of arousal necessary for sleep (Brown et.al., 2018; Dahl 1996; Papadimitriou & Linkowski, 2005). Sleep disturbing anxiety in autistic children can be exacerbated by core autism characteristics. For example, sensory sensitivities, inherent arousal dysregulation, circadian rhythm disturbances,

and behavioral characteristics such as rigid adherence to routines, ritualistic behaviours, and difficulty with transitions or identifying social 'bedtime' cues (Mazureck et.al., 2015; Nadeau et.al., 2015). Wiggs and Stores (2004, p. 377) term anxiety related sleep disturbance as: "sleeplessness associated with specific verbalised concerns or fears, with overt signs of anxiety such as distress, autonomic nervous system activity, and muscular tension". In this study, autistic individuals who appear to be experiencing anxiety according to psychometric evaluation, and self or parent report indicates anxiety may be interfering with sleep, will be included in this research.

Evidence clearly supports behavioural approaches for treating sleep disturbance, however there is little research investigating the effects of behavioral sleep treatment on children with co-occurring sleep disturbance and anxiety (Cuomo et.al., 2017; Turner & Johnson, 2012; Vriend et.al., 2011; Wiggs & Stores, 2004). Common behavioural sleep interventions for autistic children include reinforcement of positive sleep hygiene routines, modifying sleep associations, and adjusting sleep environments and schedules to promote sleep conducive behaviours (McLay et.al., 2021; Pattison et.al, 2020). Research indicates these interventions are effective in addressing problems with sleep onset and maintenance (Cuomo et.al., 2017; Rigney et.al., 2018, Souders et.al, 2017). Very little research has investigated the integration of traditional sleep and anxiety treatments, with sleep disturbance or anxiety disorders predominantly being addressed separately (Souders et.al., 2017). The aim of this study is to investigate the effectiveness of behavioural sleep interventions in children with autism, where assessment indicates that anxiety is a presenting factor, and appears to be interfering with sleep .

In this chapter, autism, sleep disturbance and anxiety are defined, behavioural sleep and anxiety treatments are described, and current treatment evidence is discussed. The literature review will explore the nature and presentation of sleep disturbance and anxiety in

autistic children, and highlight the reciprocal relationship between competing states of arousal associated with anxiety and sleep. The importance of FBA is also noted, as are limitations of existing research, highlighting the importance of continued research in this area.

Autism Spectrum Disorder

Definition

Autism is a complex neurodevelopmental condition, characterised by difficulties with verbal and non-verbal communication, social interaction, and repetitive restricted behaviors or interests. Many autistic individuals also experience unusual sensory responses to environmental stimuli, either avoiding sensory input that is experienced as overwhelming, or seeking sensory stimulation which is perceived as pleasant and calming (APA, 2018; Souders et.al., 2017). The combination and presentation of symptoms is unique to the individual and can range from having no verbal language, to experiencing difficulties with back-and-forth conversation, making eye contact, understanding facial expressions, social cues, and more complex social expectations (APA 2018). Behavioural symptoms often present as repetitive or stereotyped motor movements such as lining up toys, hand-flapping, insistence on sameness, or intolerance of uncertainty (APA 2018, Chaudhuri & Chatterjee, 2015). Cognitive ability can also vary, with some estimates suggesting 46% of individuals have average to above average intellectual ability (IQ >85), 23% lie in the borderline range (IQ 73-85), and 31% meet criteria for intellectual disability (IQ<73) (Rzepecka et.al., 2011, Souders et.al., 2017). Core characteristics of autism are evident from as young as 1-2 years of age. The unique tapestry of an autistic child's individual characteristics can significantly impact their daily life, and that of their family (Batshaw et.al, 2019; Carr, 2016).

While there are fundamental commonalities among the core features of autism, the presentation of autism characteristics, and their impact on daily life can vary greatly according to the child's age, temperament, cognitive and language abilities, and dynamics of family systems (Batshaw et.al., 2019; Carr, 2016). Early identification, intervention and support for autistic children and their families can have a significant effect on parent and child well-being, and overall future outcomes (Carr, 2016; Devnani & Hegde, 2015; Souders et.al., 2017).

Prevalence

Recent evidence suggests 1 in 54 children and young people in New Zealand are autistic (Bowden et.al., 2020), with autism affecting more than 40,000 New Zealanders (Autism Guidelines, 2016, Bowden et.al., 2020). US estimates state 1 in 43 children are diagnosed with autism (Xu et.al., 2018). Presently, more boys are diagnosed with autism than girls, at a ratio of 4:1 (Johnson et.al., 2016).

Aetiology

The cause of autism is largely undetermined, and is presently considered to be multifactorial. Research suggests autism may result from a dynamic, complex combination of genetic, environmental, and biological factors. This involves interactions between multiple gene arrangements and environmental influences that likely result in alterations in brain structure and function (Carnassi et.al., 2019; Fakhoury, 2015; Ratajczak, 2011).

Genetic factors

Evidence suggests a strong genetic component is involved in the development of autism (Fakhoury, 2015). Both family and twin studies reveal a complex but clear contribution of non mendelian genetic factors involved in autism, with heritability up to 80% (Chaste & Leboyer, 2012; Fakhoury, 2015). The influence of genetic factors in the

development of autism appears to be polygenic, and there is considerable debate around the exact nature of genetic factors in the expression of autism within families (Chaste & Leboyer, 2012). Some research suggests different symptoms of autism are due to separate threads of genetic influence. Other evidence implies symptom domains reflect various behavioural manifestations of autism which link to a single quantitative neurological difference (Chaste & Leboyer, 2012). However, recent research indicates differences in brain development in autistic children clearly extends beyond single areas of neurological functioning, with evidence implying brain-wide differences in early neural development in autistic children (Carnassi et.al., 2019; Geoffray et.al, 2016; Gliga et.al., (2014).

The most established method for investigating autism susceptibility genes is whole-genome sequencing (WES). The employment of WES in research has revealed a connection between the fragile X protein, and autism-associated genes (Fakhoury, 2015). Chromosomal microarray analysis has also evidenced differences in GABA receptor subunit genes GABRB3, GABRA5 and GABRG3, which appear to be associated with abnormal brain development involved in the pathophysiological expression of autism (Fakhoury, 2015). Additionally, selective candidate gene analysis has identified mutations in serotonergic genes and neuroligins which increase the risk of autism, and are linked to depression, and synaptogenesis (Fakhoury, 2015). The SLC6A4 gene, associated with anxiety and attention has been particularly noted as associated with the expression of behavioural symptoms in autism, with links to somatosensory functions, and social deficits in autistic individuals (Sudhof, 2008; Wiggins et.al., 2014; Fakhoury, 2015). Such findings demonstrate the continued efforts of researchers to understand the role genetics plays in autism (Chaste & Leboyer, 2012; Fakhoury, 2015; Gliga et.al., 2014).

Environmental factors

Alongside advances in our understanding of genetics in autism, evidence is increasingly identifying environmental factors, and gene-environment interactions which may contribute to the increasing prevalence of autism (Chaste & Leboyer, 2012; Gliga et.al., 2014). Some evidence suggests autistic children may react differently to certain environmental stimuli than non-autistic individuals (Chaste & Leboyer, 2012). Environmental factors may interact directly with susceptibility genes, leading to epigenetic alterations in genetic expression, increasing the likelihood of autism developing (Fakhoury, 2015). A small number of medications have been identified as possible causative agents in the incidence of autism such as valproic acid, thalidomide, and misoprostol, as well as maternal infections from rubella, and cytomegalovirus (Persico-Merelli, 2014). Some studies have highlighted a possible association between tobacco and alcohol exposure, and certain structural brain anomalies observed in autistic children (Eliasen et.al., 2010; Tran et.al., 2013). Other literature suggests nutritional disorders, air pollutant exposure, maternal infection during pregnancy, and poor socioeconomic status are potential factors influencing the predisposition towards autism (Chaste & Leboyer, 2012; Lyall et.al, 2014). Research is continuing to reveal possible links between autism and several environmental factors however, to date, no single factor within the environment is identified as influential enough to significantly predispose an individual towards autism (Fakhoury (2015).

Autism and Challenging Behaviour

Although not part of the diagnostic criteria for autism, challenging behaviour is an almost universally reported feature of daily life for families with autistic children. Literature suggests 82% - 92% of autistic children exhibit some type of challenging behaviour (Cervantes et.al., 2013; Hattier et.al., 2011; Matson & Minshawi, 2007; O'Nions et.al., 2018). This is thought to be more acute and demanding than for non-autistic children, or those with

other developmental conditions, including intellectual disability (Blacher & McIntyre, 2006; Estes et.al., 2009; O'Nions et.al.; 2018). A number of factors may explain challenging behaviours in autistic children; including language and communication difficulties, differences in social information processing, emotional regulation difficulties, sensory sensitivities, rigid thinking, and low tolerance for uncertainty and frustration (Bearss et.al., 2016; Bitsika & Sharpley, 2016; Marquenie et.al., 2011; O'Nions et.al., 2018; Rodgers et.al, 2012; Martinez et.al., 2016, Schaaf et.al, 2011).

Challenging behaviour in autistic children often includes disruptive behaviour, aggression towards others, stereotyped behaviours, pica, running away, self-injury, property damage, and inappropriate behaviour in public (Cervantes et.al., 2013; Kanne & Mazurek, 2010; O'Nions et.al., 2018). Parents report managing behaviours which require constant supervision, include risk of physical harm, accentuate the child's differences, or draw embarrassing attention as the most problematic (Kanne & Mazurek, 2010; O'Nions, 2018). Challenging behaviours are a source of significant distress for many families raising autistic children, and impact strongly on children's quality of life. Challenging behaviour can have a significant impact on parent mental and physical health problems, marital stress, anxiety, sleep disturbance, and out-of-home placement (Bitsika & Sharpley, 2016; Kanne & Mazurek, 2010; Lucyshyn et.al., 2015; Rzepecka et.al., 2011). Evidence indicates that in the absence of intervention and support, challenging behaviours tend to continue into adolescence and adulthood for autistic individuals (Conroy et.al., 2005; Goldman et.al., 2011).

One of the most frequently reported challenging behaviours which contributes to family stress, and reduced quality of life for autistic individuals is sleep disturbance (Goldman et.al., 2011; Hollway & Aman, 2011; Hollway et.al., 2013; Rzepecka et.al., 2011; Souders et.al, 2017). Research suggests there is a reciprocal relationship between challenging behaviours, and sleep deprivation. Sleep deprivation tends to increase the

occurrence of challenging behaviour, whilst expressions of challenging behaviour are associated with a higher incidence of sleep disturbance in autistic children (Cohen et.al., 2014; Mazzone et.al., 2018; Richdale et.al., 2000; Rzepecka et.al., 2011; Wiggs & Stores, 1996; Vriend et.al, 2011). As such, effective treatment of sleep disturbance in autistic children represents significant potential for reducing family stress, improving quality of life, and future outcomes for autistic children and their families (Cohen et.al., 2014; Goldman et.al., 2011; Meltzer, 2008).

Sleep in Autistic children

Function of Sleep

Sleep serves a critical function in healthy development and well-being throughout the lifespan (Souders et.al., 2017). Although the exact purpose of sleep is still not clearly understood, evidence suggests sleep is essential for healthy brain development, immune function, consolidation of memory, and cyclical restoration of physical and mental energy (Souders et.al., 2017; Wiggs, 2007). Sleep deprivation has been linked to a higher incidence of anxiety, depression, behaviour difficulties, and poor academic performance in neurotypical children (Bagley & El-Sheikh, 2013, Lushington et.al., 2013). Sleep difficulties are one of the most commonly reported problems among parents of autistic children (Cortesi et.al., 2010; Cuomo et.al., 2017, Devnani & Hegde, 2015; Herrmann, 2016; Hodge et.al., 2014; Mazurek & Petroski, 2015; Kuhn & Kennedy, 2018; Vriend et.al., 2011; Wiggs & Stores, 2004), and evidence indicates persistent sleep difficulties have a significant impact on children's daytime functioning, learning, physical and mental health, and cognitive development (Goldman et.al., 2011; Herrmann, 2016; Leahy and Gradisar, 2012; Turner and Johnson, 2012; Vriend et.al., 2011). Given the significant impact of sleep on children's development, health and wellbeing, sleep disturbance represents an issue of serious concern for autistic children and their families.

Course and Prevalence of Sleep Disturbance

Without effective treatment, sleep problems in autistic children tend to continue into adolescence and adulthood (Herrmann, 2016), with estimates suggesting approximately 63% of autistic children experience ongoing sleep issues, if left untreated (Cortesi et.al., 2014; Cuomo et.al., 2017; Hodge et.al, 2014). By contrast, sleep difficulties in typically developing children are more likely to resolve with developmental maturation, independent of intervention (Baker et.al., 2013; Hodge et.al., 2014; Richdale and Prior, 1994; Siverston et.al., 2012). In light of the increased vulnerability towards sleep difficulties for autistic children, and the continuation of these difficulties without intervention, the identification and development of effective treatments for sleep difficulties in autistic children is a pressing need.

According to current literature, sleep onset delay (SOD), and night waking (NW) are the most prevalent sleep disturbances reported by families with an autistic child (Herrmann, 2016; Hodge et.al., 2014; Malow et.al., 2014; Mazzone et.al., 2018; Richdale & Shreck, 2009). Bedtime resistance, circadian rhythm disturbance, unwanted co-sleeping, poor sleep efficiency and early morning waking are also commonly reported (Cuomo et.al., 2017, Kotagal & Broomall, 2012; Mazzone et.al., 2018; Souders et.al., 2017).

Classification of Sleep Disturbance

The International Classification of Sleep Disorders – Third Edition (ICSD-3) places major sleep disorders into seven categories: insomnias, sleep-related breathing disorders, hypersomnias of central origin, circadian rhythm disorders, parasomnias, sleep-related movement disorders, and other (secondary) sleep disorders (Sateia, 2014; Thorpy, 2012). The most prevalent sleep disturbances experienced by autistic children can be defined within three major categories: parasomnias, secondary sleep disorders, and insomnias (Herrmann,

2016; Krakowiak, 2008 Thorpy, 2012). Parasomnias include night terrors, sleep walking, nightmares, bruxism, or sleep related movement disorders which intrude on sleep (Herrmann, 2016; Krakowiak, 2008). Secondary sleep disorders relate to conditions that result from physical or psychological disorders which impair sleep such as Depression, Anxiety or Sleep Apnoea. Insomnias involve difficulties falling asleep, staying asleep (night waking), waking too early, or poor quality sleep, despite adequate opportunity and conditions for sleep. Sleep insomnias result in significant impairment in daytime functioning, related to insufficient sleep quantity and quality (Herrmann; 2016; Krakowiak, 2008; Thorpy, 2012). Difficulties falling asleep (SOD) and night waking (NW) are the most commonly reported disturbances in both typically developing children, and autistic children (Herrmann, 2016; Krakowiak, 2008), and include bedtime resistance, prolonged sleep onset with 'curtain call' behaviour (i.e., bids for parental attention), and sleep onset association disorder (Herrmann, 2016; Krakowiak, 2008; Thorpy, 2012). Extended or frequent night waking, reactive co-sleeping, and early morning awakening are also frequently reported (Goldman et.al., 2012; Herrmann, 2016; Krakowiak, 2008; Malow et.al., 2006; Richdale, 2013).

Types of Sleep Problems in Autistic Children

Bedtime Resistance /Curtain Calls

Bedtime resistance is characterised by active resistance to bedtime routines, where a child may refuse to go to bed, or stall sleep and separation from a parent (e.g., with curtain call behaviours such as making repeated requests for another story, or a glass of water (Kotagal & Broomall, 2012; Meltzer, 2010; Vriend & Corkum, 2011). Stressed and tired parents may inadvertently maintain these bedtime difficulties by responding to the child's requests and giving them repeated attention (Meltzer 2010; Wiggs, 2008).

Sleep-onset association disorder

Sleep onset association disorder describes a child's reliance on external stimuli to initiate and maintain sleep, for example the presence of a parent, breast feeding, or the need to be rocked, patted, or held in order to get to sleep (Thorpy, 2012). This reliance on an external stimulus usually affects both sleep onset and night wakings, where the child requires the presence of the stimuli to re-initiate sleep if they wake in the night. This often prompts parents to again provide the necessary sleep cue, as the child is not able to self-soothe and return to sleep on their own (Broomall and Kotagal, 2012; Krakowiak, 2008; Meltzer, 2010; Mindell et.al., 2006; Thorpy, 2012).

Co-sleeping

Co-sleeping can be defined as a parent or parents sharing the same sleeping space as their child, and can be either intentional, or reactive (Dodd & Jackiewicz, 2015). Intentional co-sleeping involves a choice by families to bed-share as the best sleeping arrangement for their family. This is most often a choice guided by cultural preferences, societal values, or socio-economic status (Goldberg & Keller, 2007; Ma et.al., 2014; Thoman, 2006). Intentional co-sleeping is thus guided by choice, and is not considered a sleep problem in this context.

Reactive co-sleeping refers to the occurrence of bed-sharing where parents have conceded to co-sleeping with their child due to difficulties with the child's capacity to initiate sleep independently at sleep onset or when waking in the night, even though separate sleeping arrangements are reported as the parents' preference (Goldberg & Keller, 2007; Kose et.al., 2017). Co-sleeping is more frequently reported as a problem by parents of autistic children than parents of typically developing children, predominantly within the younger and middle childhood years (Kose et.al., 2017; Lindbom, 2019).

Circadian Rhythm Sleep Disorders

A number of recent studies have reported an association between sleep disturbances and circadian dysregulation in autistic children (Carmassi et.al., 2019; Mullegama et.al., 2015). Six main circadian sleep disorders are classified by the ICSD-3. Circadian sleep disorders include shift work disorder, jet-lag disorder, delayed sleep-wake phase disorder, advanced sleep-wake phase disorder, non-24hr sleep-wake rhythm disorder, and sleep-wake disorder not otherwise specified (Sateia, 2014). Evidence suggests the most commonly occurring circadian rhythm disorders seen in autistic children are delayed sleep-phase disorder, and irregular sleep/wake rhythm disorder (Goto et.al., 2017; Mullegama et.al., 2015; Nicholas et.al., 2007; Yang et.al., 2016). Literature reflects a link between these particular expressions of circadian dysrhythmicity and behavioural manifestations of delayed sleep onset, bedtime resistance, and frequent night waking in autistic children (Carmassi, 2019, Glickman, 2010; Hodge et.al., 2014).

Delayed sleep-phase disorder

Delayed Sleep-phase Disorder is characterised by a delayed circadian phase where core body temperature, melatonin rhythms, initiation of the sleep period, and morning awakening are delayed, and usually misaligned with societal expectations (Drake, 2010, Glickman, 2010, Wyatt et.al., 2006). This disorder involves a lengthened intrinsic circadian period, even in the presence of normal external, or 'entrainment' cues (Drake, 2010).

Irregular sleep/wake rhythm disorder

Irregular sleep/wake rhythm disorder manifests as a lack of circadian rhythm, with multiple short sleep periods taken throughout the day/night cycle. Interestingly, children with this pattern of sleep may sustain a comparatively normal total amount of sleep over 24 hours, but the sleep schedule is highly fragmented. This sleep disorder is most commonly seen in

children with neurological conditions involving damage to a mechanism important in circadian regulation, the suprachiasmatic nuclei (SCN) (Glickman, 2010; Drake, 2010). This polyphasic sleep pattern is similar to infant sleep, possibly suggesting a link between circadian sleep disturbances in autistic children, and differing maturation rates within the circadian system (Glickman, 2010; Mazzone et.al., 2018). Literature suggests the usual response to entrainment signals in autistic children may be affected by hypo or hyper sensitivities to sensory information, particularly sound and sight cues, and decreased attention and understanding of social cues associated with sleep routines (Dawson et.al., 1998; Glickman, 2010; Talay-Ongan & Wood, 2000).

Causes of Sleep Problems in Autistic Children

A number of factors appear to contribute to the onset and maintenance of sleep disturbance in autistic children, including biological, psychological, and social factors, often related to core characteristics of autism (Herrmann, 2016; Kotagal & Broomall, 2012; Mazzone et.al., 2018; Nadeau et.al., 2015). Richdale and Shreck (2009), suggest a biopsychosocial framework for understanding the causes of sleep difficulties in autistic children (Cortesi et.al., 2010, Kotagal & Broomall, 2012, Popodopoulus et.al., 2015, Richdale & Shreck, 2009).

Biological Factors

Melatonin and Cortisol Production. Evidence suggests certain biological factors contribute to sleep disturbance in children with autism (Cortesi et.al., 2010; Mazurek & Petroski, 2015; Richdale & Schreck, 2009; Souders et.al., 2017). For example, individuals with autism appear to have atypical patterns of cortisol and melatonin production (Corbett et.al., 2006; Cortesi et.al., 2010; Herrmann, 2016, Kotagal & Broomall, 2012; Melke et.al., 2008; Richdale &Prior, 1992; Souders et.al., 2017; Wiggs & Stores, 2004). Blood, salivary

and urinary levels of melatonin have been shown to be significantly lower in autistic individuals in several studies (Carmassi, et.al., 2019; Devnani &Hegde, 2015; Glickman, 2010; Herrmann, 2016; Mazzone et.al., 2018; Melke et.al., 2008). A growing number of autistic children are accessing exogenous melatonin therapy, with a number of studies demonstrating the effectiveness of exogenous melatonin in improving SOD, NW, and early morning awakening (Devnani & Hegde 2015; Souders et.al., 2017). Some studies reflect mixed results, suggesting melatonin therapy loses its effectiveness over time, or has no effect, particularly when forms of behavioural intervention are not employed (Malow et.al., 2011; Souders et.al.,2009; Souders et.al., 2017). Melatonin dysregulation, including delays in melatonin peak, diminished production of melatonin, and altered melatonin gene expression also appears to play a role in circadian rhythm sleep disorders in autistic children (Carmassi et.al., 2019; Glickman, 2010; Herrmann, 2016; Mazzone et.al. 2018; Souders et.al.2017).

Circadian Dysregulation. Biochemical secretions (e.g. melatonin and cortisol) as well as behavioural patterns are involved in regulating the circadian cycle, orchestrated by the neurobiological 'master clock', located in the hypothalamus: the suprachiasmatic nuclei (SCN) (Carmassi et.al., 2019; Glickman, 2010). The endogenous SCN rhythm is also synchronised by environmental signals, particularly the 24 hr day/night cycle (Carmassi et.al., 2019, Glickman, 2010). Other extrinsic cues which appear to influence these internal rhythms include food intake, sound, social cues, exposure to stress, and behavioural conditioning (Carmassi et.al., 2019; Chang et.al., 2009; Geoffray et.al., 2016). A number of studies have noted links between sleep disturbance and circadian dysregulation in autistic children, yet the exact nature of this link remains unclear. Recent research implies bi-directional influences are at play, whereby sleep disturbance may not only exacerbate circadian dysregulation, but genetic differences in autistic children may directly impair circadian function, playing a causal role in both sleep disturbance, and intensifying

characteristics of autism, in a self-reinforcing feedback loop (Carmassi et.al., 2019; Geoffray et.al., 2016; Glickman, 2010; Nicholas et.al., 2007; Yang et.al., 2015; van der Heijden et.al., 2018).

Autism-related Psychosocial Factors

Numerous studies link sleep disturbance in autistic children to core characteristics of autism such as communication differences, difficulties with reciprocal social communication, responding to social cues, and behavioural adherence to strict routines or rituals (Herrmann, 2016; Kotagal & Broomall, 2012; Lindor et.al., 2019; Mazzone et.al., 2018; Nadeau et.al., 2015; Richdale & Shreck, 2009). Some studies note sleep disturbance is more prevalent among those with more prominent characteristics of autism (Herrmann, 2016; Lindor et.al., 2019; Richdale & Schreck, 2009), however sleep disturbance can be a significant problem for individuals across the full range of the autism spectrum (Lindor et.al., 2019; Mazzone et.al., 2018; Veatch, Maxwell-Horn, Malow, 2015). According to Hollway & Aman (2011), characteristics of autism appear to increase an individual's vulnerability to sleep difficulty, and predisposes autistic children to sleep disturbance in the presence of certain environmental factors.

Language/ Social Communication. Sleep difficulties such as bed-time resistance, and sleep onset delay can be associated with language or social communication differences. Some autistic children may struggle to recognise social and environmental cues which prepare them for bedtime, such as not recognising parents' behavioural and verbal prompts for them to go to bed (Herrmann, 2016; Kotagal & Broomall, 2012; Mazzone et.al., 2018; Nadeau et.al., 2015). Autistic children may have difficulty attending to and understanding instructions to prepare for and proceed to bed, particularly if instructions are given without warning, or in the absence of a visual prompt (Kotagal & Broomall, 2012; Malow et.al., 2014). Additionally, some autistic children may find it a challenge to communicate their

needs such as expressing pain or discomfort that may affect their ability to sleep (Reed et.al., 2009; Singh & Zimmerman, 2015).

Behavioural Differences. Repetitive, ritualistic behaviours, difficulty shifting from one activity to another, or insistence on sameness can also interfere with bedtime routines. For example, variations in specific before-bed routines can cause distress for some autistic individuals, and hinder the process of settling to bed if routines are not strictly followed. Strong attachment to specific routines or objects can also present difficulties with night-waking, where some autistic children may have difficulty returning to sleep independently if a routine is not repeated, or an object or person is not present to re-initiate sleep in the night (Reed et.al., 2009; O'Nions et.al., 2018; Souders et.al., 2017). Autistic children may perseverate on activities, and struggle making the shift from one task to another, which can further delay the process of getting to bed and settling to sleep (Mazzone et.al., 2018; Richdale & Shreck, 2009; Souders et.al., 2017).

Sensory Processing Differences. Sensory processing differences and arousal dysregulation are characteristic features of autism, and appear to play a role in exacerbating difficulties with sleep onset and maintenance (Hollway et.al., 2011; Hollway et.al., 2013; Mazurek & Petroski, 2015; Mazzone et.al., 2018; Souders et.al., 2017). Environmental stressors which impact sleep difficulties for autistic children include both over-responsivity, and under-responsivity to sensory stimuli (Hollway et.al., 2013; Mazzone et.al., 2018).

Sensory over-responsivity is characterised by heightened responses to everyday sensory experiences often involving visual, auditory or touch stimuli (Carpenter, et.al., 2019). Autistic children may be extra sensitive to light, noises, smells, or tactile input, which can interfere with their ability to settle to sleep. Conversely, children who experience under-responsivity to sensory stimuli may require high levels of touch, visual or vestibular sensory input to relax and fall asleep (Mazurek & Petroski, 2015; Reynolds et.al., 2012; Souders

et.al., 2017). When autistic children experience sensory discomfort, they frequently resort to coping strategies which interfere with, rather than induce relaxation and sleep. This may include repetitive stereotypical behaviours, bedtime resistance, or demands for parental attention (Hollway et.al., 2013; Herrmann, 2016; Mazurek & Petroski, 2015; Richdale & Shreck, 2009; Souders et.al., 2017). These contrasting sensory processing differences add to the complexity of sleep disturbance in autistic children (Cortesi et.al., 2010; Hollway, Aman & Butter, 2013; Mazzone et.al., 2018; Souders, 2017).

Arousal Dysregulation. Theories on the role of dysregulated arousal states in autistic children have long been hypothesised, with an early study by Hutt and colleagues in 1964 (cited in Souders et.al., 2017), suggesting autism involves heightened states of arousal, which may directly interfere with sleep. Recent evidence suggests genetic indicators related to hyperarousal, play a role in the incidence of sleep disturbance in non-autistic and autistic children (Hollway & Aman, 2011; Malow and McGrew, 2008; Mazurek & Petroski, 2015; Richdale, 1999; Souders et.al., 2009; Souders et.al., 2017). Studies are emerging which demonstrate the possibility of differences in brain function and subsequent neural development of arousal regulatory systems in autistic children (Geoffray et.al., 2016; Goto et.al., 2017; Mazzone et.al., 2018; Yang et.al., 2016). For example, evidence indicates a number of unique and diverse variations within neuronal networks which regulate hormonal fluctuations related to arousal state (Carnassi et.al., 2019; Geoffray et.al., 2016; Glickman, 2010; Mazurek & Petroski, 2015; Mazzone et.al., 2018; Puzino et.al., 2018). These neurobiological systems subsequently have a significant impact on arousal regulation (Puzino et.al., 2018; Wuyts et.al., 2012), and the initiation and maintenance of sleep in individuals with autism (Carnassi et.al., 2019; Geoffray et.al., 2016; Glickman, 2010; Mazurek & Petroski, 2015; Mazzone et.al., 2018).

Psychological Factors

Bi-directional mechanisms between sleep disturbance and psychological factors such as Major Depression, Post-Traumatic Stress Disorder, Bipolar Disorder and Generalised Anxiety Disorder have been identified in the literature, but are not yet fully understood (Fang et.al., 2019; Moore, 2012). In the past, sleep disturbance was regarded as a core secondary symptom of depression. However, more recent evidence has noted sleep disturbance often precedes depression, recognising it as a risk factor in the occurrence of certain mood disorders such as depression and anxiety (Cox & Olatunji, 2016; Fang et.al., 2019; Riemann et.al., 2020). Literature suggests various mechanisms may be at play in the relationship between sleep disturbance and depression (Fang et.al., 2019). For example, specific inflammatory markers known to impact sleep and mood have been identified, and circadian rhythm dysfunction has also been linked to depression, as well as sleep disorders (Carmassi et.al., 2019; Fang et.al, 2019). Recent evidence has suggested genetic factors may also be involved (Fang et.al., 2019). Autistic children and youth are more at risk that the non-autistic population for developing sleep disturbance, as well as mood disorders such as Major Depression, and Generalised Anxiety Disorder (Mayes & Calhoun, 2009; Mazurek & Petroski, 2015). Although recent literature has established links between mood disorders and sleep disturbance in autistic children and youth (Hollway & Aman, 2011; Mazurek & Patroski, 2015), there is a paucity of research elucidating the nature of the relationship between these factors in autistic children. The relationship between anxiety and sleep disturbance in autistic youth will be explored in more depth later in this chapter.

Behavioural Model of Sleep Disturbance: The Role of Learning and Reinforcement

Behavioural theory provides a useful framework for understanding how, and why sleep disturbances occur. Positive and negative reinforcement can play a primary role in the occurrence and maintenance of sleep disturbances (France and Blampied, 1993). Both parent

and child can be caught in a cycle of unhelpful responding, which creates and maintains sleep disturbing behaviors. For example, a child who has learned the positive association of falling asleep with their parent present is likely to engage in behaviors which ensure this condition (parental presence) is met, such as crying or getting out of bed in order to avoid the negative experience of falling asleep alone (France et.al., 2003). The maintenance of this coercive cycle of parent-child interactions can be exacerbated by anxiety. Behavioral avoidance, or 'escape conditioning' is the primary function of anxiety-related learned behaviours. Within the context of sleep, a child who is fearful of being left alone, and successfully avoids this anxiety provoking situation by behaving in a way which obtains the presence of their parent to fall asleep, is caught in a cycle of behavioural avoidance.

Learned associations of fear and subsequent avoidance are powerful mechanisms in the onset and maintenance of anxiety disorders, and anxiety-related sleep disturbance (Helbig-Lang & Peterman, 2010; Pittig et.al., 2018). Associative fear involves the repeated pairing of a stimulus with an aversive (unpleasant) unconditioned stimulus. Over time, this pairing results in an individual learning that a particular stimulus is fearful or unpleasant, and behaviours which result in avoidance of that feared stimulus provide immediate relief from distress. Ultimately, the individual learns that avoidance is the most effective way to ensure their feared consequence does not eventuate. The experience of anxiety is often accompanied by a distorted perception of threat, where a harmful outcome is strongly feared, yet realistically unlikely to eventuate, or cause harm. This distorted perception intensifies feelings of anxiety, and reinforces escape behaviours (active avoidance of the threatening object or circumstance) (Thompkins, 2013).

Children who learn that escape behaviors reliably cause the removal of unpleasant stimuli are strongly motivated to consistently repeat the learned behaviour to ensure they avoid the distress of their anxiety inducing consequence (escape conditioning). For example

in the context of sleep disturbance, a child's escape behavior (crying out for a parent) consistently results in the parent responding with their presence to calm the child, causing the removal of unpleasant stimuli (separation from a parent). Through this cycle of behaviour, the child also learns they are not able to calm their own anxious feelings, and without a parent present to calm their anxiety, they will become overwhelmed with distress they cannot resolve independently (Helbig-Lang & Peterman, 2010). The belief that they are not able to achieve sleep without the calming mechanism of the parent's presence is continually reinforced (Helbig-Lang & Peterman, 2010). The parent's response inadvertently rewards, or reinforces their child's learned escape behaviour every time their presence and reassurance is made available to alleviate their child's distress, and establish sleep in the short term (France et.al., 2003). This cycle of behavior negatively reinforces both child and parent through avoidance of distress, and simultaneously provides positive reinforcement through the consequence of early sleep onset. To continue avoiding distress and achieving sleep onset, both child and parent are likely to continue this cycle of behavior (France et.al., 1996). This cycle may also be repeated if a child wakes in the night, and is once again unable to independently calm their distress, and initiate sleep without a parent's presence (France et.al, 2003). Over time, a child's reliance on external forces (parental reassurance and guidance) to assuage their anxiety reduces their ability to initiate or maintain sleep independently (Nadeau et.al., 2015; France and Blampied, 1993).

Anxiety

Defining Anxiety

The experience of anxiety is considered a part of normal human development and life experience for all individuals (Kerns et.al., 2017; Thompkins, 2013). Cognitive, physiological, and behavioural mechanisms are involved in anxious responding, including intrusive, repetitive thoughts about an event, feelings of fear and stress, a pounding heart,

sweating, rapid breathing, feeling faint or shaky, and attempts to avoid, escape, or control fearful situations (Elwood et.al., 2012; Thompkins, 2013; von-Steensel et.al., 2011).

Although some use the terms anxiety and fear interchangeably, the literature tends to identify distinctive differences between the two. Fear is primarily viewed as an adaptive, acute response to a salient threat; a response elicited upon factual, sensory input, with a predominantly autonomic arousal of our 'fight or flight' instinct (Tovote et.al., 2015; DSM-V, APA, 2013). Anxiety tends to represent a more diffuse, pervasive reaction to stimuli which is not well defined, such as general apprehension about the future, or anxiety about what might happen (Silverman et.al., 2011; Thompkins, 2013).

Cognitively, anxiety elicits an unrealistic perception of uncontrollability over aversive events. Physiological responses involve heightened autonomic arousal. Behavioural manifestations of anxiety consistently present as escape or avoidance behaviors (including cognitive avoidance) in the presence of an anxiety-provoking stimulus, or partial avoidance, with sufferers enduring a situation with obvious distress, such as pacing, handwringing, or clinging to a caregiver with urgent requests for support (Silverman and Field, 2011; Thompkins, 2013; Zvolensky et.al., 2000). Safety behaviours may also be incorporated as a coping mechanism, such as excessive checking of a parent's presence for a child with separation anxiety, or avoiding looking down and holding one's breath while riding an elevator, for someone with a specific phobia of elevators (Tompkins, 2013). An adaptive, anxious response in new situations and challenges is considered developmentally normal in individuals as they negotiate new or unfamiliar contexts in the course of life, and the characteristics and developmental trends of normative anxiety are similar across cultures (Pittig et.al., 2018; Silverman & Field, 2011).

Anxiety Disorders

Disordered or impairing anxiety is more often characterised by hypervigilant anticipation of future threat; a diffuse, persistent response to an imprecise, anticipated perception of threat (DSM-V, APA, 2013; Lang et.al., 2010). In contrast to typical experiences of anxiety, disordered anxiety presents symptoms beyond expected developmental levels, disproportionate to the actual threat posed, and is persistent, irrational, and negatively impacting on everyday functioning and quality of life (Kerns & Kendall, 2013; Lang et.al., 2010; Pittig et.al., 2018; South et.al., 2017).

Anxiety disorders are associated with three or more symptoms including restlessness, edginess, fatigue, concentration difficulties, irritability, muscle tension, anxious responses to specific situations or objects, and sleep disturbance (APA, 2017; Keefer et.al., 2018). The DSM-V includes seven categories related to anxiety: Generalised Anxiety Disorder (GAD), Separation Anxiety Disorder (SAD), Specific Phobia (SP), Agoraphobia, Panic Disorder, Social Anxiety Disorder (Social Phobia), and Selective Mutism (DSM-V, 2013). All anxiety disorders cause significant distress, and disruption to daily functioning and developmental progress for those who experience them, and their families (Keefer et.al., 2018; Silverman & Field, 2011; South et.al., 2017; Williams et.al., 2015).

The Development and Maintenance of Anxiety Disorders

The experience of impairing levels of anxiety in children and adolescents has risen over the past two decades, and is now recognised as one of the most common, developmentally disruptive, and socially distressing mental health challenges faced by the current generation of youth (Rapee et.al., 2009; Silverman & Field, 2011; White et.al., 2014). Various biological, psychological, and psychosocial factors appear to contribute to the development of anxiety disorders in children, including genetic and neurobiological

influences, individual temperament and cognitive style, and other environmental familial influences such as parent child interactions, parenting style or personality, and significant life stressors (Carr, 2016; Rapee et.al., 2009).

Genetic, Neurobiological Factors

Research indicates children with a parent or first-degree relative who has an anxiety disorder are significantly more likely to develop an anxiety disorder themselves (Carr, 2016; Kerns et.al., 2017; Rapee et.al., 2009). This involves possible genetic, and neurobiological characteristics. Twin studies support the hypothesis for a genetic influence in anxiety disorders, with evidence of an association between anxiety disorders and various candidate genes identified, although not clearly specified (Carr 2016; Rapee et.al., 2009). Neuroimaging technology has contributed to the identification of neuro-biological factors contributing to the development of anxiety disorders. This includes evidence of atypical variations in neuroanatomical systems associated with threat processing, and stress-response hypersensitivities (Carr, 2016; Zantvoord et.al., 2013). Similarly, evidence continues to grow around the influence of chronic exposure to trauma or high stress environments on a child's developing brain, particularly regulation of the stress-response system located within the amygdala (Caspi et.al., 2010; van der Kolk, 2007).

Individual Temperament and Cognitive Style

Several studies have noted certain personality traits increase the likelihood of a child developing anxiety disorders, when compared to children with other temperamental traits (Buss & Kiel, 2013; Fox & Pine, 2012; Rapee et.al., 2009). Evidence indicates behavioural inhibition including traits of harm avoidance, reticence approaching and communicating with strangers, tendency to remain close to safety figures and withdrawal in the face of novelty, was associated with increased risk for development of anxiety disorder in children (Buss &

Kiel, 2013; Fox & Pine, 2012; Kampman, Vikki, Jarvenausta, Leinonen, 2014). Children with traits of novelty seeking and sociability appeared to be significantly less vulnerable towards developing anxiety disorders (Buss & Kiel, 2013; Fox & Pine, 2012; Kampman et.al., 2014). Some evidence suggests cognitive features such as attentional bias towards threat is important in the development and maintenance of anxiety disorders (Rapee et.al., 2009). For example, preliminary research has found that attentional bias towards threat in early childhood predicted patterns of social withdrawal and later development of clinical anxiety (Bus & Kiel, 2013, Fox & Pine, 2012, Rapee et.al., 2009).

Familial Environmental Influences

Research indicates a number of environmental family-related variables may play a role in the development of anxiety disorders in children and adolescents (Rapee, 2012). Currently, evidence points most clearly to the influence of parent-child interactions on the development of anxiety disorders (Rapee et.al., 2009; Rapee, 2012; Sahithya & Raman, 2021). Parent-child interactions characterised by an overprotective parenting style, negative or critical parenting, and highly permissive parenting have been linked to increased odds of anxiety disorder developing in children and youth (Rapee, 2012; Sahithya & Raman, 2021). Parental expression of anxious responding has also been linked to the development of anxiety disorders in children (Lawrence et.al., 2019; Sahithya & Raman, 2021). By contrast, a parenting style characterised by warmth, affection and authoritativeness was associated with a reduced incidence of anxiety disorder in children (Rapee, 2012; Sahithya & Raman, 2021).

Research on the impact of chronic life stressors and complex childhood trauma on clinical levels of anxiety in children and youth continues to grow (van der Kolk; 2007), with evidence of a significant link between chronic exposure to stress and trauma in the early developmental years, and the development of anxiety and other disorders in childhood, adolescence, and adulthood (Beesdo-Baum & Knappe, 2012; Dye, 2018; Thibaut, 2022).

The impact of specific negative life events on the development of anxiety disorders is less clear, with some research indicating significant negative life events are not associated with increased vulnerability towards anxiety disorders in children (Hovens et.al., 2015), whilst other research suggests childhood life events impact on the development of anxiety disorders in youth, when mediated by other risk factors for anxiety disorder such as parent-child interactions, and childhood temperament traits (Broeren et.al., 2013; Rapee et al., 2009). Overall, evidence suggests a number of factors influence the development of childhood anxiety disorders, with both family-related heritable, and environmental factors playing a significant role (Beesdo-Baum & Knappe, 2012; Rapee et.al., 2009; Silverman & Field, 2011; Thibaut, 2022). Autistic children are not only impacted by these same risk factors, but literature suggests specific characteristics of autism may increase their vulnerability towards experiencing anxiety disorders (Kerns et.al., 2017; White et.al., 2014; South & Rodgers, 2017; Wood & Gadow, 2010).

Anxiety Disorders in Autistic Children

Prevalence

Many autistic children have co-occurring physical and psychological conditions (Carr, 2016; Cohen et.al., 2014). Common co-occurring physical health conditions include enuresis, encopresis, gastrointestinal problems, food and other allergies, and feeding problems. Psychological conditions which commonly co-occur include ADHD, ODD, CD, Depression, Sleep Disturbance, OCD, and Specific Phobias (Carr, 2016; Cohen et.al., 2014). Among the most common co-occurring psychological conditions are anxiety disorders (Hallett et.al., 2016; Kotagal & Broomall, 2012).

Research suggests up to 84% of autistic children experience high levels of distress related to anxiety, and approximately 42-55% of young people with autism are diagnosed

with an anxiety disorder (Delli et.al., 2018; Keefer et.al., 2018; Hallett et.al., 2013; Lecavalier et.al., 2014; O'Nions et.al., 2018; South & Rodgers, 2017; van Steensel, et.al., 2011; Williams et.al., 2015). In contrast, prevalence rates of anxiety disorders in the general child/adolescent population ranges from 2.2 - 20% (Kerns et.al., 2017; Rapee, et.al., 2009). A number of factors are thought to contribute towards higher prevalence rates of anxiety in autistic children, including differences in social motivation, sensory sensitivities, emotional regulation challenges, difficulties with interoception, and intolerance of uncertainty (Kerns et.al., 2017; Mazurek & Petroski, 2015; Rzepecka et.al., 2011; Souders et.al., 2017; von-Steensel et.al., 2011).

Autism Characteristics and Anxiety Disorders

The development and maintenance of anxiety disorders in individuals with autism appear to share several overlaps with typically developing individuals, as well as atypical presentations which may be more attributable to characteristics of autism (Kerns & Kendall, 2013; Kerns et.al., 2017; South & Rodgers, 2017; Wood & Gadow, 2010). For example social avoidance, differences in social reciprocity, and compulsive/ritualistic behaviours are challenges faced by both autistic and non-autistic individuals with anxiety, particularly OCD, or social phobia (Kerns & Kendall, 2013; Ollendick & White, 2013). Additionally, cognitive distortions, (i.e. 'all or nothing' thinking, dwelling on perceived threats), and physiological hyperarousal are a common feature of anxiety disorders, regardless of autism diagnosis (Britton et.al., 2010; Ollendick & White, 2013; Kerns & Kendall, 2013; Kerns et.al., 2017; Pittig et.al., 2018; South & Rodgers, 2017).

Anxiety disorders have been characterised as 'high arousal disorders', (Cuomo et.al., 2017; Mazurek & Petroski, 2015; Nadeau et.al., 2011; Rzepecka et.al., 2011; Souders et.al., 2017). Literature indicates autistic children exhibit differences in the functioning of the autonomic nervous system, similar to that seen in other children with anxiety disorders,

including sympathetic nervous system activity, which increases their susceptibility to anxiety disorders. (Souders et.al., 2017, Mazzone et.al., 2018). Kushki and colleagues (2013), provided evidence of autistic children exhibiting heightened autonomic responses (heart rate, electrodermal activity and skin temperature), in anxiety provoking conditions when compared to typical peers.

While several aspects of anxiety disorders in youth with autism appear to present similarly to that of non-autistic individuals, it is possible the aetiology of these presenting behaviours may differ (Kerns & Kendall, 2013; Wood & Gadow, 2010; South & Rodgers, 2017). Core characteristics of autism can result in stressful or negative experiences which induce anxiety, such as poor perspective-taking contributing to negative social experiences, sensory over-responsivity triggering discomfort, pain and avoidance in the presence of everyday environmental stimulus, or disruption of adherence to specific routines creating distress and anxiety (Kerns & Kendall, 2012; Wood & Gadow, 2010; South & Rodgers,2017; South et.al., 2017). Autistic features such as social motivation differences, sensory sensitivities, and intolerance of uncertainty may contribute uniquely to the development and maintenance of anxiety disorders in autistic individuals (Green et.al., 2012, Moree & Davis, 2010, South et.al., 2017).

Social Motivation Differences

Avoidance of social interactions in socially anxious non-autistic youth is primarily anxiety-related, yet it can be difficult to determine whether social avoidance for an autistic individual may be differently motivated (Wood & Gadow, 2010). Both non-autistic youth, and autistic individuals experience anxiety in social contexts, however, for the non-autistic individual this is commonly related to fears of humiliation, negative evaluation, or performance fears. For autistic individuals, evidence suggests social anxiety is more often related to difficulty understanding and interpreting emotional or social cues. This then

creates anxiety provoking confusion or uncertainty in social interactions, leading to social avoidance (Kerns et.al., 2017; Moree & Davis, 2010; South et.al., 2017; Wood & Gadow, 2010). Research further indicates some autistic youth experience distressing social anxiety without explicit fears related to negative evaluation or worry about social performance (Gillott and Furness, 2001; Kerns et.al., 2014; Kerns et.al., 2017).

Sensory Over-sensitivity

Sensory sensitivity causes an individual to be highly sensitive to various stimuli in their social, and physical environment (Uljarevic et.al., 2016). Autistic children with sensory over-sensitivity often show signs of distress in noisy or visually complex environments, or when touched unexpectedly (Green & Ben-Sasson, 2010, Uljarevic et.al., 2016). A reciprocal relationship between atypical sensory function and anxiety appears to be a feature of anxiety experienced by autistic youth (South & Rodgers, 2017). Empirical evidence suggests sensory over-responsivity increases the incidence of impairing levels of anxiety in the autistic community (Green & Ben-Sasson, 2010; Rodgers et.al., 2016). This includes incidences of behavioural avoidance, and neurological evidence of heightened cortisol responses in the presence of sensory over-stimulation (Corbett et.al., 2016; Green et.al., 2015; South & Rodgers, 2017). Some studies found autistic youth who experienced moderate to severe sensory over-sensitivity reported significantly higher anxiety scores than those individuals without (Sasson et.al., 2008; Uljarevic et.al., 2016). Although evidence points towards a relationship between sensory over-responsivity and anxiety, the causal mechanisms involved remain unclear (Corbett et.al., 2016; Green & Ben-Sasson, South & Rogers, 2017; Rodgers et.al., 2016).

Intolerance of Uncertainty

Intolerance of uncertainty, or insistence on sameness relates to a heightened sensitivity to uncertainty in everyday situations, and a tendency to perceive unfamiliar or unpredictable events as a threat (Uljarevic et.al., 2016). Research suggests intolerance of uncertainty is featured in the presentation of most individuals with an anxiety disorder, particularly generalised anxiety disorder (Boulter et.al., 2014; Carleton et.al., 2012; Dugas & Koerner, 2005). Intolerance of uncertainty is also a distinctive characteristic of autistic individuals (South & Rodgers, 2017; Wood & Gadow, 2010). An insistence on sameness related to anxiety and worry in the face of changes to daily schedules, novel situations or unfamiliar experiences is almost universally experienced by autistic youth (Gotham et.al., 2013; Ozsivadjian et.al., 2012, Kerns et.al., 2017). Some evidence indicates a significant relationship between intolerance of uncertainty and anxiety severity in autistic children, and suggests intolerance of uncertainty has a significant influence on the maintenance of anxiety in autistic, and non-autistic children (Boulter et.al., 2014; Kerns et.al., 2017).

Diagnosis of Anxiety in Autistic Children

The diverse evidence regarding the nature and presentation of anxiety in individuals with autism, has raised questions regarding whether anxiety constitutes a co-occurring condition in autistic individuals, or whether it is more representative of characteristic features unique to autism (Bearss et.al., 2016; Ollendick & White, 2013; South et.al., 2017). For example, literature suggests repetitive, compulsive behaviours, or ruminative, fixated cognitions which feature as symptoms of anxiety disorders, may in fact be more representative of an underlying cognitive disposition in individuals with autism (Kerns & Kendall, 2013; Wood & Gadow, 2010; South &Rodgers, 2017). Other characteristics associated with autism such as diminished awareness of internal emotional states (limited interoception), challenges in expressing abstract concepts, or reduced motivation to report on

these symptoms may impact the accuracy of diagnostic instruments in detecting anxiety, particularly those which rely on self-report data (Wood & Gadow, 2010, South & Rodgers, 2017). Thus a body of research (Wood & Gadow 2010) suggests core features of autism may be misidentified as co-occurring anxiety, and highlight the possibility of existing diagnostic instruments for anxiety eliciting inaccurate representations of the true nature of anxiety in autistic children and youth (Kerns & Kendall, 2013; Kerns et.al., 2017; Ollendick & White, 2013; Williams et.al., 2015; Wood & Gadow (2010).

Although research exploring the bi-directional relationship between autism and anxiety continues to grow, a deeper understanding of the nature of this relationship has yet to be fully understood. One aspect of this relationship is firmly established; anxiety-related challenges add a significant burden to daily life for many autistic individuals, and their families (Kerns & Kendall, 2013; Ozsivadjian et.al., 2012; O'Nions et.al., 2018; South & Rodgers, 2017; South et.al., 2017; Williams et.al., 2015).

Autism, Anxiety and Sleep Disturbance – Reciprocal Relationships

Several studies highlight the association between anxiety in autistic children and sleep problems including bedtime resistance, sleep onset and maintenance, and difficulties sleeping independently (Hollway et.al., 2013; Mazurek & Petroski, 2015; Rzepecka et.al., 2011; Souders et.al., 2017). A study by Hollway and colleagues (2013) found anxiety was the strongest predictor of sleep disturbance in autistic children, over and above other externalizing and internalizing disorders, sensory sensitivities, autism severity and gastrointestinal problems. Autistic children recognised as 'poor sleepers' tended to have more internalizing symptoms than autistic children classed as 'good sleepers' (Hollway et.al., 2013). Other studies found autistic children with average to above-average IQ appear to be prone to anxiety-related sleep disturbance such as fear of the dark, separation anxiety, worries due to social anxiety, nightmares, and pre-sleep arousal generated by anxious cognitions

(Hollway et.al., 2013; Paavonen et.al., 2008; Richdale & Baglin, 2013; Williams et.al., 2015). Some studies suggest a link between anxiety, sleep disturbance and level of intellectual functioning in autistic children (Rzepecka et.al., 2011; Hagopian & Jennett, 2008). Other studies indicate a consistent relationship between sleep disturbance and anxiety, regardless of intellectual functioning (Giannotti et.al., 2008; Hollway, et.al., 2013; Taylor et.al., 2012). Clearly, the association between sleep problems, autism and anxiety is multi-faceted and complex (Nadeau et.al., 2015), with evidence of reciprocal influences between various characteristics apparent, in anxious autistic youth with sleep disturbance (Hollway et.al., 2013; Uhde et.al., 2009).

Aetiological Factors

Evidence suggests both sleep and anxiety may be regulated by interconnected neurobiological activities. The fact that sleep disturbance and anxiety disorders are so commonly co-occurring in both autistic and non-autistic individuals suggest the possibility of shared neurobiological features (Lindor et.al., 2019; Tsypes et.al., 2013; Uhde et.al., 2009). There are several possibilities to consider in explaining the nature of this relationship, particularly in presentations of Generalised Anxiety Disorder (GAD), where the sleep architecture and nature of sleep difficulties between patients with primary GAD, and those with primary insomnia are almost identical (Tsypes et.al., 2013; Uhde et.al., 2009). This suggests the possibility that neurobiological influences in anxiety and sleep difficulty may rest along the same continuum – representing the same neurobiological features, with a spectrum of presenting difficulties which are influenced by a variety of stimuli (Carpenter et.al., 2019; Tsypes et.al., 2013; Uhde, 2009). A relationship of this nature (shared aetiology) may be indicated if treatment of either component – anxiety, or sleep disturbance in autistic children, results in alleviation of both conditions – sleep disturbance, and anxiety symptoms (Carpenter et.al., 2019; Uhde, 2009).

An alternative hypothesis is that both sleep disturbance and anxiety are significantly, synergistically influenced by a third factor, such as autism characteristics (Hollway et.al., 2011b; Hollway et.al., 2013; Nadeau et.al., 2015; Uhde et.al., 2009; Mazzone et.al., 2018). Hollway and colleagues (2011b) hypothesised that autism characteristics represent significant vulnerability factors which predispose children to sleep disturbance, and anxiety. Within the context of autism, either or both conditions may be triggered or exacerbated by environmental stressors, which increase the likelihood of maladaptive coping mechanisms being employed by the child, and caregivers. Thus, the risk of autistic individuals developing internalising disorders such as anxiety, or depression and associated sleep disturbance, is heightened. Results from this and other studies demonstrate evidence of reciprocal influences within physiological, emotional, and behavioural spheres related to autism characteristics, but clear evidence of the exact nature of these relationships remains elusive, with mixed, inconclusive results (Brown et.al., 2018: Hollway et.al., 2011b; Nadeau et.al., 2015). Indeed, the links between sleep disturbance, autism characteristics and anxiety suggest a complex relationship, with autism and anxiety impacting children's sleep in numerous ways (Nadeau et.al., 2015; Souders et.al., 2017). Sleep disturbance in anxious autistic youth occurs as a result of numerous possible internal and environmental factors, with the cause in any particular individual likely to be multi-factorial. However, evidence suggests a number of intrinsic and extrinsic factors render autistic youth uniquely vulnerable to sleep disturbance, and anxiety (Hollway & Aman, 2011b; Nadeau et.al., 2015; Souders et.al., 2017).

Physiological Factors: The Primary Role of Arousal States

Anxiety, sleep disturbance and some characteristics of autism are strongly influenced by high states of physiological arousal. Physiological states of arousal are largely regulated by circadian and homeostatic sleep/wake cycles, the pendulation between high and low states

of arousal throughout the rhythm of day and night (Brown et.al., 2018; Dahl, 1996; Mazurek & Petroski, 2015; Souders et.al., 2017). Sleep onset and maintenance requires low arousal states, while anxiety is characterised by high states of arousal (Brown et.al., 2018; Mazzone et.al., 2018; Puzino et.al., 2018; Dahl, 1996). A consistent association between states of hyperarousal and sleep onset difficulties has been demonstrated in the literature in non-autistic children, autistic children, and adults experiencing anxiety disorders and insomnia (Brown et.al., 2018; Mayes & Calhoun, 2009; Mazzone et.al., 2018; Mazurek & Petroski, 2015; Puzino et.al., 2018). Cognitive hyper-arousal in particular has been evidenced as a contributing factor in sleep difficulties related to sleep onset delay, in non-autistic adolescent and adult populations, and in some studies within the autistic population (Brown et.al. 2018; Hollway et.al., 2013; Mazurek & Petroski 2015; Puzino et.al., 2018). The increased cognitive activity and associated physiological responses generated by anxious cognitions and perceptions of threat trigger heightened arousal, directly interfering with low arousal states required for initiating and maintaining sleep (Leahy & Gradisar, 2012; Mazurek & Petroski, 2015; Souders et.al., 2017). Results from some recent studies provide evidence of a correlation between severity of sleep disturbance, and pre-sleep cognitive arousal in young adults, with strong correlations between physiological hyperarousal states at sleep onset, and severity of self-reported anxiety (Mayes & Calhoun, 2009; Puzino et.al., 2018). These results are consistent with previous theories that suggest pre-sleep cognitive arousal and cortical hyper-arousal play a significant role in the onset and maintenance of sleep disturbances, particularly sleep onset delay. Evidence indicates ruminative, repetitive cognitions, including intrusive thoughts impact both physiological and emotional hyperarousal, contributing to difficulties getting to sleep (Brown et.al., 2018; Mazzone et.al., 2018).

Emotional Factors: Self-Regulation

Emotional awareness and regulation (interoception) involve the complex process of recognising, assessing, and adjusting emotional responses to emotional triggers, and is recognised as an area of challenge for autistic individuals (Tsypes et.al., 2013; Santomauro et.al., 2016). Difficulties with the process of emotion recognition and regulation; termed as Alexithymia by South & Rodgers (2017), is noted as a risk factor for internalising disorders such as anxiety or depression. This is in part, reflected in the higher prevalence rates of anxiety and depression in autistic individuals (Santomauro et.al., 2016; South & Rodgers, 2017; White et.al., 2014).

In addition to a number of shared risk factors for sleep disturbance and anxiety disorders in autistic children, Brown and colleagues (2018), suggest adolescent hormonal changes represent a contributing factor. Hormonal changes in the typical developmental pubescent phase increase the dysregulation of arousal and emotional states, which may exacerbate anxiety-related sleep disturbance during the adolescent period. Some studies framed an increase in adolescent sleep disturbance as a secondary symptom of the emotional dysregulation inherent in the experience of internalising disorders such as anxiety or depression (Alvaro et.al., 2017). However, research increasingly indicates the reciprocal nature of the relationship between sleep disturbance and emotional regulation (Brown et.al, 2018; Mazzone et.al., 2018; Puzino et.al., 2018). Vandekerchove & Cluydts (2010) framed the bidirectional association between sleep disturbance and emotion dysregulation in typical development, as pertaining to the restorative function REM sleep serves, in building emotional regulation capacity during the day. Emotional dysregulation experienced during the day can result in sleep deprivation, which limits the quantity and quality of REM sleep obtained during the night. Thus a bi-directional loop is created where sleep deprivation reduces capacity for emotional regulation, and vice versa. The potential impact of sleep

deprivation on emotion regulation and anxiety states in adolescents, was further explored in studies where reduced sleep resulted in self and parent reported increases in emotion regulation difficulties during the day (Baum et.al., 2014; Cousins et.al., 2011).

The bidirectional relationship between sleep disturbance and anxiety-related emotional dysregulation is reflected in literature that examines treatments for these conditions. Evidence indicates treatment of anxiety disorders can result in improvement of sleep (Alfano & Ginsburg, 2007; Chase & Pincus, 2011), and the treatment of sleep disturbance can reduce levels of anxiety (Brown et.al., 2018). These findings provide important evidence on the reciprocal influences at play in sleep disturbance and anxiety-related emotional dysregulation. However, more research is clearly needed to elucidate the relationship between anxiety, sleep disturbance, and autism characteristics. Rates of sleep disturbance and anxiety are higher in the autistic population than the non-autistic population. Therefore, such research has important implications for refining treatments which improve well-being and daily functional outcomes for youth with autism, and their families.

Behavioural Factors

For autistic individuals, intense physiological, sensory, and emotional manifestations of anxiety and sleep disturbance can result in overwhelming, unpleasant experiences, increasing the likelihood of distressed behavioural responses to everyday challenges (Hollway et.al., 2013; Samson et.al., 2015). Difficulties with emotion regulation, and limitations in capacity for cognitive reappraisal, increase their vulnerability towards expressing distressed behavioural responses, exacerbating emotional and behavioural difficulties such as sleep disturbance, and anxiety (Hollway et.al., 2013; Samson et.al., 2015). The development and maintenance of distressed behaviours related to sleep disturbance and anxiety always extend beyond the individual, involving their interactions with the people and environments around them (O'Nions et.al., 2018; Samson et.al., 2015; Souders et.al., 2017).

Environmental Factors and Behavioural Impacts in the Context of Autism

The interplay between anxiety and sleep disturbance factors (e.g. hyperarousal, emotional dysregulation) within an autistic child, and their resulting interactions with people and environments around them, can create a complex cycle of perpetuating influences. For example, sleep-related anxieties such as fear of the dark, or sensory sensitivity to light or texture tend to heighten arousal, and hinder sleep. If these factors are combined with other autism characteristics such as a child's need for routine, ritualistic behaviours, or support with transitions, the likelihood of the autistic child experiencing distress at bedtime is significant. This may well result in distress-related behaviors, motivated by a strong need to seek parental attention and support with bedtime routines and sleep initiation (Nadeau et.al., 2015; O'Nions et.al., 2018; Ozsivadjian et.al., 2012). Parents of both autistic and non-autistic children tend to respond naturally to their child's behavioral need for attention and support, by attempting to reassure their child, and acting to reduce behaviours which interfere with sleep. For non-autistic children, distress-related behaviours are more likely to be milder in expression, temporary, and more likely to resolve without intervention (Cervantes et.al., 2013; Nadeau et.al., 2017; Ozsivadjian et.al., 2012; O'Nions et.al., 2018; Owens & Moore, 2017). Conversely, autism-related characteristics tend to increase the intensity and duration of distress related behaviours in autistic children, with a greater reliance on external supports to manage distress and hyperarousal. These intense cycles of behaviour tend to increase stress levels, and reduce both parent and child well-being (Goldman et.al., 2012; Meltzer, 2011; Nadeau et.al., 2017; O'Nions et.al., 2018). Parents of autistic children caught in these behaviour cycles can feel a sense of despair. They may believe the present behaviour cycle is the only solution due to the intensity and endurance of the behaviours, and feel concerned their child with autism might not be capable of learning skills to achieve emotional regulation, and independent sleep. Fortunately, research and interventions based on

behavioural theory provide evidence, and thus hope for positive change in these situations. Evidence indicates these cycles of behaviour can be changed in autistic children. Independent self-regulation, sleep onset and maintenance can be learned, with the right treatment and support for both child, and caregivers (Nadeau et.al., 2017; O'Nions et.al., 2018).

Behavioural sleep insomnias can result in prolonged periods of disturbed sleep for autistic children and their parents throughout the childhood years if not addressed, and the impact of these problems on both child and family can be significant. Disturbed sleep is associated with increased risk of family difficulties such as child learning and behaviour difficulties, marital discord, poor parental health, or parent-child relationship difficulties (Wiggs & Stores; Kotagal et.al., 2010). Predictable, consistently established routines and bedtime practices, such as a set bedtime and relaxing pre-sleep activities can promote positive sleep behaviours. However, with autistic children, the factors driving and maintaining these behaviours can be complex, and frequently require specialised support and individualised intervention to alleviate sleep disturbances of this nature (Goldman et.al., 2012; Herrmann, 2016; Kotagal & Broomall, 2012; Wiggs, 2008; Wiggs & Stores, 2004; Owens & Moore, 2017).

Behavioural Treatments for Sleep Disturbance

Behavioural interventions apply the principles of learning theory to elicit change in a person's response to a particular stimulus (Owens et.al., 1999). Several antecedent, extinction-based, and reinforcement-based behavioral interventions have proven effective in treating sleep disturbance in non-autistic and autistic children, including faded-parental-presence (stimulus-fading), faded bedtime with or without response-cost, sleep restriction, planned ignoring, modified extinction, and the bed-time pass (Cuomo et.al., 2017; Jin et.al.,

2008; Leahy & Gradisar, 2012; Moon et.al., 2010; Turner & Johnson, 2012; Vriend et.al., 2011).

Antecedent-Based Procedures.

Sleep hygiene modifications

Sleep hygiene practices and bedtime routines are considered first line treatment for sleep disturbance, as other interventions are unlikely to be successful if these issues are not first addressed. This can include establishing a predictable bedtime and pre-bedtime routine, with a bedroom environment conducive to sleep, e.g. a dimly lit, quiet, warm room (Jan et.al., 2008). Research suggests sleep hygiene practices are insufficient as a stand-alone intervention, but are a necessary and effective component of all behavioral sleep interventions for non-autistic and autistic children and youth (Cortesi et.al., 2010; Jan et.al., 2008; Malow et.al., 2014; Vriend et.al., 2011).

Autism-specific Supports

The addition of visual supports and Social Stories (Gray & Garand, 1993) can also be useful supplementary components which support the delivery of behavioral interventions for autistic children. For example, visual schedules depicting the bedtime routine, or Gro-clocks that provide a discriminative stimulus for sleep and wake times, and social stories provide support for autistic children in understanding and accepting new routines and expectations (McLay et al., 2018; Souders et.al., 2017). Social stories are a visual support composed as an individualised story using language and images (often photographs), to increase understanding about specific social situations for autistic people (McLay et.al., 2018; Williams & Wright, 2017). Social stories have been found to increase motivation for change, and decrease anxiety for autistic children when introducing new, unfamiliar routines and behavioral expectations (Williams & Wright, 2017). Research demonstrates that visual

supports and social stories are useful, effective additions to multi-component behavioral sleep interventions (Moore, 2004; McLay et.al., 2018).

Faded Bedtime

Studies have shown evidence of success with faded bedtime interventions in autistic children (Christodulu & Durand 2004; Hunter et.al., 2021; Luiselli et.al., 2020; Moon et.al., 2010; Vriend et.al., 2011). This treatment identifies the typical time that a child falls asleep (not the time they go to bed), and delays their bedtime to within 15 minutes of that sleep onset time (Kodak & Piazza, 2008; Vriend et.al., 2011). A set morning waketime is included which initially restricts the amount of sleep the child gets, increasing their sleep drive for sleep onset in the evening. Once sleep onset is achieved within 10-15 minutes, the bedtime is moved earlier in gradual increments, until the target bedtime is reached (Kodak & Piazza, 2008). This procedure has been shown to be effective in addressing circadian dysregulation-based sleep difficulties (Carmassi, 2019; Hodge et.al., 2014). This procedure has demonstrated effectiveness in reducing SOD, NW's, and increasing duration of sleep in autistic children (Moon et.al., 2010; Kodak & Piazza, 2011).

Faded Bedtime with Response-cost

Faded-bedtime-with response-cost involves the same procedure as faded bedtime, but includes a consequence-based component. If the child does not fall asleep quickly when put to bed at the specified time, they are removed from bed, engaged in a quiet activity for a set amount of time, then returned to bed. Once the child is achieving rapid sleep onset, bedtime is incrementally moved forward in the same manner as above, until the target bedtime is reached (Vriend et.al., 2011).

Sleep Restriction Procedures

Sleep restriction procedures are similar to faded-bedtime-with-response-cost, but with a focus on overall duration of sleep and time spent in bed, not the time till sleep onset. The time a child spends in bed is limited to 90% of their baseline total sleep time. If there is a decrease in sleep disturbance, the child's bedtime is gradually adjusted until the desired bedtime is reached. The procedure may also involve restrictions on daytime sleep (Vriend et.al., 2011). Some studies suggest interventions which involve sleep restriction may be particularly suitable for treating anxiety-related sleep disturbance, and reducing hyper-arousal by increasing sleep pressure (Leahy & Gradisar, 2012; Vriend, et.al., 2011). Sleep restriction has demonstrated some success with autistic children (Durand & Christodulu, 2004; van Deurs et.al., 2019; Vriend et.al., 2011).

Consequence-Based Procedures – Behavioural Extinction

Planned Ignoring and Minimal Check

Extinction-based procedures have the greatest empirical support for treating sleep problems in both non-autistic and autistic children (Kodak & Piazza, 2008; Richdale & Wiggs, 2005; Vriend et.al., 2010). This intervention is sometimes referred to as 'planned ignoring', and involves the elimination or reduction of reinforcement for sleep interfering behaviours e.g., systematically ignoring children's bids for parental attention (Turner & Johnson, 2012). Modifications to extinction procedures that also have empirical support, include graduated extinction ('minimal check'), where parents enter the bedroom to check on the child, according to a pre-planned time schedule (e.g. every 5 minutes), rather than contingent on the child's behavior (Kodak & Piazza, 2008). This time interval is extended incrementally, essentially providing a gradual, rather than abrupt withdrawal of attention (Cuomo et.al., 2010; Turner & Johnson, 2012). This procedure has been shown as effective

in a limited number of studies involving autistic children or children with other developmental disabilities (Durand et.al., 1996; Moore, 2004).

Faded Parental Presence

Faded parental presence (sometimes called Camping Out) involves the gradual fading of a parent's presence while providing minimal interaction with the child. Treatment involves the parent remaining on a chair or separate mattress in the child's room. The parent remains with the child until they fall asleep, but provides minimal attention, with no verbal or physical interaction. If the child wakes in the night, the parent may return to the mattress or chair until the child returns to sleep. The distance between parent and child is gradually extended by moving the chair or mattress further away from the child over successive nights, until their presence is 'faded' out of the child's room, and the child is falling asleep independently (Cuomo et.al., 2017; France & Blampied, 2005; Vriend et.al., 2011). The parent's presence provides reassurance for the child, but minimal interaction eliminates reinforcement of attention-seeking behaviors. This reduces anxiety while gradually enabling the child to gain the skill of settling to sleep independently. Literature suggests this procedure may be particularly appropriate for children experiencing separation anxiety (Howlin, 1984; Sadeh, 1994). Some studies indicate faded parental presence can be effective in the treatment of sleep disturbance in both autistic, and non-autistic children (France & Blampied, 2005; McLay et.al., 2017; McLay et.al., 2019). Other studies have demonstrated effectiveness of this procedure when combined with other behavioural interventions (Howlin, 1984; McLay et.al., 2017; Souders et.al., 2017).

Consequence-based Procedures including a Reinforcement Component

Bedtime Pass

A 'bedtime pass' can be used to supplement extinction-based interventions. This involves both components of consequence, and reinforcement of target behaviours. The use of a physical 'pass card' entitles the child to momentarily leave the bedroom a prior agreed number of times, (e.g. 1-3 times) after being put to bed. For example, the child may use the pass to request items that do not compete with sleep such as a glass of water, or to use the bathroom (Freeman 2006). The child is rewarded in the morning if they get through the night without trading in their pass (Souders et.al., 2017). This can reduce anxiety during the intervention procedure, and has been shown to reduce the likelihood of an extinction burst (Fisher et.al., 1998; Freeman, 2006). A small body of literature demonstrated this strategy as effective with non-autistic children (Freeman, 2006; Friman et.al.,1999; Moore et.al., 2007) , but its effectiveness with autistic children has not yet been clearly established (Souders et.al., 2017)

Excuse-me Drill

A recently developed intervention which shows promise as a treatment for children experiencing sleep interfering anxiety is the *'excuse-me-drill',* which combines the complementary forces of extinction and reinforcement, in teaching a child the skill of falling asleep independently (Kuhn, 2010). This procedure includes multiple steps and commences during the bedtime routine once the child is settled into bed. The parent excuses themself for a short, pre-determined period of time, then returns and praises the child for tolerating their absence (Honaker & Meltzer, 2014). The 'excuse me' absences continue with the length of parent absence gradually increased. The eventual goal is for the child to fall asleep independently, without distress (Kuhn, 2010). Although this intervention can be time-

intensive in its initial stages, this procedure can minimise child and parent distress, while providing immediate reinforcement of acceptable replacement behaviors (Kuhn, 2010). A recent study demonstrated this procedure as effective in reducing disruptive bedtime behaviours, and achieving independent sleep onset in autistic children (Kuhn et.al., 2020).

Cognitive Behavioural Therapy (CBT) Targeting Anxiety

Cognitive behavioural therapy is a structured psychotherapeutic approach, utilised in both adult and paediatric populations to address a wide variety of mental health challenges, including anxiety. CBT explores connections between thoughts, emotions, and behaviour, supporting clients to shift unhelpful cognitions and behaviours, and replace them with helpful cognitions and behavioural coping strategies (Fenn & Byrne, 2013). The critical components of CBT treatment for childhood anxiety are founded on the basic elements of early behavioral, and cognitive theories (Gosch et.al., 2006). According to cognitive-behavioral theory, dysfunctional beliefs and behavior strategies underlie all common psychological disturbances, including anxiety. Methods can be implemented to modify thinking and belief systems, resulting in enduring emotional and behavioral change e.g., less anxiety and more adaptive behaviors (Beck, 2011; Dummett, 2006; Pearson, 2017).

CBT Core Components

The standard components of a CBT based therapy include psychoeducation, cognitive restructuring to manage and re-frame maladaptive thoughts, with graded exposure to feared situations, alongside emotional regulation strategies such as breathing and muscle relaxation (Danial & Wood, 2013; Dowell et.al., 2018; White et.al., 2010; Mellman, 2006; Souders, et.al., 2017). The majority of CBT interventions for children include both child, and parent components to improve generalisation, and consolidate maintenance of treatment affects (Danial & Wood, 2013; Dowell et.al., 2018; Kerns et.al, 2017).

Psychoeducation

Psychoeducation usually focuses on building understanding of anxiety, it's physical, emotional, and behavioural impact, and exploring the child's thoughts, feelings, and experiences of anxiety. Often the clinician will describe the cycle that maintains anxiety, and the client's own triggers, physiological, and behavioural responses will be identified. Parents are often involved in this step. Anxiety is also framed as a normal experience for all, where skills can be learned to reduce, and manage anxiety in daily life. Emotion regulation strategies such as diaphragmatic breathing and progressive muscle relaxation are often taught to reduce tension, and provide strategies to manage anxiety when implementing exposure tasks to feared stimuli (Kerns et.al., 2017; Read et.al., 2013). With children, information may be presented in association with their favourite interests or heroes, to increase engagement and motivation early in the therapeutic process.

Cognitive Restructuring

The link between thoughts, feelings and behaviour is revisited, with a specific emphasis on tackling unhelpful 'self-talk' (cognitions) as a way to reduce anxiety (Kerns et.al, 2017). Specific strategies are introduced to assist the child to recognise their 'worry thoughts', and reframe them with 'warrior' brave thoughts (Attwood, 2004; Huebner, 2006). This process promotes coping thoughts, and helps to reduce anxiety. However, this takes some practice, and is most suitable for children over 7 years of age, when metacognitive abilities are sufficiently developed (Read et.al., 2013).

Graded Exposure

Exposure is a core component of CBT, and essential in the effective treatment of anxiety, as it addresses a primary feature of anxiety disorders; avoidance (Chorpita & Daleiden, 2009; Peterman et.al., 2015). Exposure involves the systematic, graded experience

of fearful situations, with appropriate support. This provides the client with the opportunity to cope in a feared situation armed with new coping skills, without resorting to avoidance or escape (Chorpita & Daleiden, 2009). A hierarchy of the child's fears is created in collaboration with the child, with items graded from least, to most feared situations. Specific graded exposure tasks to face fears step by step, are planned with the child and parent.

CBT has generated a robust evidence base, and is currently considered the 'first line' treatment for anxiety within the general population, with the advantage of not presenting a risk of side effects, as with pharmacological interventions (Dowell et.al., 2018; Dummett, 2006; Keefer et.al., 2018; Moree & Davis III, 2010). Both brief, intensive duration, or longer-term duration of CBT intervention have proven effective (Dowell et.al., 2018). Evidence demonstrates CBT is an efficacious treatment delivered in a variety of formats including individual, group, classroom, and online interventions (Dowell et.al., 2018; Dugas & Koerner, 2005; Ebert et.al., 2015; Khanna & Kendall, 2010).

Modified CBT for Anxious Autistic Children

Although CBT has robust empirical support for its effectiveness as a child therapy for anxiety in the general population, there has been debate regarding whether it is suitable and effective for use with autistic youth, particularly the cognitive component (Moree & Davis III, 2010). Research suggests that with certain modifications, CBT can be highly effective in the treatment of anxiety in autistic youth with low support needs (Danial & Wood, 2013; Dowell et.al., 2018; Lang et.al., 2010; Moree & Davis III, 2010; Vasa et.al., 2014). Modifications to CBT therapies are frequently made to render them more suitable for autistic individuals. Evidence-based modifications which enhance the benefit of CBT for autistic youth include increased use of visual supports, social stories, illustrating emotions with concrete analogies and objects (scrapbooks and drawings describing emotions), and providing coping strategies that are less reliant on abstract language and concepts (Anderson

& Morris, 2006; Attwood, 2004; Gray, 1998; Hagopian & Jennett, 2008). Further suggestions which aim at increasing understanding and generalisation of therapeutic strategies include extending the duration and number of sessions, including a communication and social skills module, adjusting content to the developmental level of the child, greater involvement of caregivers, and incorporation of the child's special interests into the therapy process (Attwood 1999; Beebe & Risi 2003; Jassi, 2021; Moree & Davis III, 2010, Chorpita & Daleiden, 2009). Recent trends in CBT therapy include the incorporation of mindfulness-based techniques such as visualisation and meditation, for increasing emotional awareness and regulation, to reduce anxiety, challenging behaviours, and to improve sleep in both non-autistic, and autistic individuals (de Bruin et.al., 2015; Dykens et.al., 2014; Ekman & Hiltunen, 2015; Spek et.al., 2013).

A variety of therapeutic approaches have been applied in the management of anxiety for autistic children, including modified CBT, and psychosocial interventions, parent education, pharmacotherapy, and school-based social-skills programs (Delli et.al., 2018; Hagopian & Jennett, 2008; Rzepecka et.al., 2011). Treatments with the most empirical support in autistic children includes modified CBT, and pharmacological treatment with SSRI's (Danial & Wood, 2013; Delli et.al., 2008; Hofmann et.al., 2009; Munshi et.al., 2011; Vasa et.al., 2015). Recently a number of studies have indicated the efficacy of modified CBT in the treatment of anxiety in autistic children and adolescents (Ung et.al., 2015).

CBT Anxiety Treatment for Autistic Children

Group therapy

Some studies have demonstrated post-treatment reductions in anxious autistic youth through group treatment. In an early study, Sofronoff and colleagues (2005), provided modified group CBT delivered in six 2-hour sessions to 71 anxious youth aged 10-12 years.

Two treatment groups (one with a parent component and one without) were compared with a waitlist control group. Standard CBT treatment components were supplemented with additional visual aids, and social stories. Outcomes for the treatment groups demonstrated significant reductions in anxiety post-treatment, and increased adaptive coping skills according to parent reports, with some additional gains noted in the child and parent treatment group (Danial et.al., 2013). Chalfont and colleagues (2007) presented a 12 week group CBT program (without specific modifications to typical CBT components), to 47 autistic children with a mean age of 10 years. Parent, teacher, and self-report outcome measures on the Anxiety and Related Disorders Interview Schedule (ADIS-5) (Brown & Barlow, 2014), detected moderate decreases in anxiety in the treatment group, compared to waitlist-controls. Treatment fidelity was not assessed in either of these studies. A more recent study administered modified CBT for anxiety to 50 autistic adolescents aged 13-18 years. Participants were assigned to the CBT treatment group, or a Treatment as Usual (TAU) control group (medication and/or emotion-coaching or school-based social skills training). Key CBT elements in the 'Face Your Fears' Program were supplemented with the addition of parent/teen dyads in the group, social skills training, and video-modelling (Reaven et.al., 2012). Participants exhibited significant post-treatment reductions in anxiety compared to the TAU group, with 79% of treatment participants demonstrating positive treatment gains (Reaven et.al., 2012; Danial et.al., 2013; Kendall et.al., 2017).

Group therapy may possess some advantages, such as therapeutic reach in treating several clients simultaneously, and the potential for individuals to benefit from mutual support (Kendall et.al., 2017). However, individual therapy enables treatment to be more intensive and individualised, which may be particularly beneficial for autistic individuals (Kendall et.al., 2017). Literature suggests individual CBT can be highly effective in reducing anxiety in autistic children and adolescents (Kendall et.al., 2017).

Individual Therapy

A version of the Coping Cat program was modified to suit 22 autistic children with clinically significant anxiety (McNally-Keehn et.al., 2013). Participants were aged 8-14 and randomly assigned to 16 individual sessions of treatment, or a waitlist. Modifications to the program included additional visual materials and session reviews, and integration of children's special interests in content delivery. Following treatment, participants demonstrated significantly greater reductions on anxiety measures than those on the waitlist, and treatment gains were maintained at a 2-month follow up (McNally-Keehn et.al., 2013). Another manualised CBT program was utilised in 3 studies: Behavioural Interventions for Anxiety in Children with Autism (BIACA). Evidence-based autism appropriate modifications are embedded in this program, and include separate parent sessions (Ehrenreich-May, et.al., 2014). Ehrenreich-May and colleagues (2014) evaluated CBT treatment using BIACA with autistic adolescents. In this study, 20 adolescent (11-14yrs) autistic males with anxiety disorder participated in an open trial of the 16-week program. Significant reductions in parent and clinician-rated anxiety were evident from baseline to post-treatment, and parent-rated reductions in externalising symptoms were also noted (Ehrenreich-May et.al., 2014; Kendall et.al, 2017). Two later studies also indicated BIACA as effective in reducing anxiety in autistic youth. A 2015 study implemented the program with 33 adolescent participants, with positive treatment response indicated in 79% of treatment group participants post-treatment, compared to 28.6% treatment responders in the waitlist group (Wood et.al., 2015). A smaller study which used BIACA with 7 older adolescents aged 16-20 years also indicated significant reductions on clinician rated anxiety measures, following treatment (Wise et.al., 2019).

The literature reflects encouraging results regarding CBT treatment for anxiety in autistic youth, although the benefit of specific treatment components is yet to be identified,

and no direct comparisons on the effectiveness of group vs individual therapy have been studied to date (Wise et.al., 2019; Kendall et.al., 2017). However, evidence continues to emerge suggesting both individual and group CBT therapies can be effective in reducing anxiety in autistic children and adolescents, especially when individualised, and evidence-based autism-specific modifications are implemented (Danial et.al., 2013: Kendall et.al., 2017; Wood et.al, 2015).

Cognitive Behavioural Therapy for Insomnia (CBT-I)

Components of CBT-I

Baglioni et.al. (2019) suggests CBT-I represents a system of therapy which includes a family of evidence-based interventions, rather than a single therapy. Indeed, CBT-I contains several components which have been described in previous sections, such as sleep restriction, circadian therapy, stimulus control, sleep hygiene psychoeducation, and core elements of CBT for anxiety such as cognitive restructuring, and relaxation techniques. Third wave approaches, including mindfulness, visualisation and meditation techniques are increasingly being included in CBT-I treatment also (Baglioni et.al., 2019; van Deurs; 2020).

Primarily, CBT-I incorporates core components of CBT such as psychoeducation, cognitive restructuring, emotion regulation strategies, relaxation skills, and behaviour change, all within the context of sleep (Johnsen, 2021; Teo et.al., 2022). Factors which tend to maintain sleep disturbance over time such as sleep interfering behaviours and cognitions, and dysregulated sleep cycles are addressed through psychoeducation, sleep restriction and stimulus control therapy, and cognitive behavioural strategies (Baglioni et.al., 2019; Johnsen, 2019; Teo et.al., 2022). Psychoeducation promotes understanding about sleep hygiene practices, the circadian rhythm, and healthy sleep/wake cycles. Factors which are likely contributing to sleep disturbance (e.g. caffeine intake, device use in bed, daytime sleep) are

also explored (Paine & Gradisar, 2011; Teo et.al., 2022; Van Deurs, 2020). Cognitive restructuring focusses on identifying and challenging negative sleep-related thoughts and beliefs, and replacing them with helpful thoughts and attitudes (van Deurs, 2020; Johnsen, 2021). Stimulus control involves the elimination of discriminative stimuli associated with wakefulness such as device use in bed, or providing discriminative stimuli which act as a cue for sleep, such as putting on pyjamas and having a bedtime story. Stimulus control facilitates a stable sleep/wake cycle, the elimination of sleep-interfering behaviours and items, and promotes sleep conducive environments and behaviours. Stimulus control is an important behavioural element of CBT-I, for achieving the lowered states of arousal necessary for sleep (Baglioni et.al., 2022; van Deurs, 2020).

CBT-I Treatment

CBT-I for Non-autistic Adults and Youth

CBT for Insomnia (CBT-I) has been established in the literature as an effective treatment for individuals with insomnia in the adult population (Teo et.al., 2022; Johnsen, 2021). A smaller body of evidence indicates CBT-I demonstrates efficacy in the treatment of non-autistic children and adolescents (Aslund et.al., 2018; Johnsen, 2021; Schlarb et.al., 2010).

Schlarb et.al (2010) conducted a pilot study of insomnia treatment for non-autistic youth, implementing a CBT-I program called “JuSt”. The study involved 18 participants aged 11-16, and their parents. Six weekly sessions of age-oriented content were delivered face to face, including education about sleep cycles, sleep hygiene, stimulus control, cognitive strategies to manage sleep disturbing cognitions, and relaxation techniques. Post treatment data indicated reductions in SOL and NW, with participants reporting improved daytime energy, and fewer worries about their sleep (Schlarb et.al., 2010).

In 2015, de Bruin and colleagues investigated the efficacy of CBT-I in non-autistic adolescents through the delivery of digital, or face-to-face group CBT-I, compared to a waitlist group (de Bruin et.al., 2015). 116 adolescents aged 16-19 y/o were randomly assigned to the digital delivery, group therapy, or waitlist group. Six weekly sessions of CBT-I included core components of CBT-I; psychoeducation, sleep hygiene, sleep restriction, stimulus control, cognitive therapy, and relaxation strategies (de Bruin et.al., 2015). Both treatment groups demonstrated significant reductions in sleep disturbance following treatment compared to controls. Differences in treatment effect between digital and face-to face delivery was minimal, with medium to large effect sizes evident in both groups (de Bruin, 2015).

CBT-I for Autistic Youth

3 CBT-I studies with autistic youth have been undertaken to date. McCrae et.al (2020), provided CBT-I for autistic primary-school aged children. Components of CBT for insomnia included psychoeducation, cognitive therapy, and homework assignments, with reinforcement of target behaviours. Parents accompanied their children for the 8 session program, and an element of individualisation was incorporated to address primary presenting sleep problems for each participant. Protocols included evidence-based modifications for autistic youth such as visual supports, reduced abstract concepts, and incorporation of special interest themes (McCrae et.al., 2020). Post treatment outcomes demonstrated reductions in sleep onset, increases in overall nighttime sleep, and parent reported improvements in daytime functioning (McCrae et.al., 2020).

A 2021 study by van Deurs and colleagues undertook CBT-I with autistic youth aged 9-15 years of age, with an emphasis on including youth participants in treatment implementation (van Deurs et.al., 2021). Treatment was individualised and adapted for autism in collaboration with participants and families. Intervention components included

psychoeducation, faded parental presence, relaxation exercises, social stories, sleep-wake scheduling, and stimulus control (van Deurs et.al., 2021). Decreases in sleep disturbance were noted following treatment, including reduced SOL and NW's, and elimination of unwanted co-sleeping (van Deurs et.al., 2021).

A very recent study delivered CBT-I via internet, for adolescents with autism spectrum disorder, aged 13-17 years (Georen et.al., 2022). Content included core elements of CBT-I based on a manualised CBT-I treatment, modified for autistic individuals (Sanchez-Ortuno & Edinger, 2015). One module per week was digitally delivered for eight weeks via psychoeducational videos, including content on sleep hygiene, sleep restriction therapy, stimulus control, safety behaviour, cognitive strategies, and relaxation exercises (Georen et.al., 2022). Results indicated significant reductions in sleep disturbance symptoms including SOL, and total sleep time according to post-treatment quantitative data, with smaller gains evident at 6-month follow-up. Qualitative data suggested improvements in daytime functioning and well-being, and perceived sleep quality and quantity (Georen et.al., 2022).

Some recent studies of insomnia treatments for non-autistic youth have included evaluation of co-occurring psychological conditions such as depression, and anxiety. Evidence evaluating the effectiveness of CBT treatment of insomnia in non-autistic and autistic anxious youth, will be outlined, and discussed in the following section.

Cognitive Behavioural Therapy (CBT) for Anxiety-related Sleep Disturbance

Cognitive Behavioural Therapy (CBT) is an empirically supported intervention for the treatment of both anxiety and sleep disturbance (Peterman, et.al., 2016; Paine & Gradisar, 2011). CBT treatments for anxiety and sleep disturbance feature overlapping components, including psychoeducation, relaxation training, graduated exposure, and contingency

management (Belleville et.al., 2011; Clementi et.al., 2016; Danial & Wood, 2013). Early sleep research tends to pay significantly more attention to behavioural, rather than cognitive components of CBT, especially in treatments for younger children (Sadeh, 2005; Tikotsky & Sadeh, 2010). However, the importance of modifying parent's cognitions regarding their expectations, interpretations and emotions related to children's sleep behaviours, is noted as essential to the success of behavioural interventions (Malow et.al., 2014; Rigney et.al., 2018; Sadeh, 2005). The development of CBT-I treatment for adolescents has also increased emphasis on cognitive therapy to address sleep disturbing cognitions; particularly when sleep-disturbance involves other psychological conditions such as anxiety, or depression (Aslund, et.al., 2020; Zetterqvist et.al., 2021). Intervention research on older autistic children and adolescents with sleep disturbance and anxiety is limited, but overall, lends itself to the addition of more cognitive components such as relaxation strategies for reducing anxiety and pre-sleep cognitive arousal (Paine & Gradisar, 2011; Tikotsky & Sadeh, 2010; Zetterqvist, 2021). There are several empirically supported behavioural interventions for sleep problems in non-autistic children (Cuomo et.al., 2017; Vriend et.al., 2011) and to a lesser extent, autistic children (McLay et.al., 2018; Moon et.al., 2009; Turner & Johnson, 2012; Rigney et.al., 2018; Vriend et.al., 2011). However, few studies have investigated the effectiveness of interventions specifically for anxiety-related sleep disturbance (Hollway et.al., 2013; Souders et.al., 2017).

Although anxiety and sleep problems frequently co-occur, standard clinical practice still tends to involve separate treatment of each disorder (Clementi, 2017; Peterman et.al., 2016). Considering the prevalence of both sleep problems and co-occurring anxiety in the autistic population, and its impact on family health and well-being, there is a pressing need for further research which addresses the relationship between these two variables. Increasing understanding of the reciprocal relationship between anxiety and sleep disturbance has

prompted more interest in integrated interventions which address anxiety, and sleep problems simultaneously (Orchard et.al., 2020; Peterman et.al., 2016, Clementi et.al., 2016). However, research on interventions with anxiety as the primary treatment focus, but within the context of sleep disturbance is scarce, with only a few studies reporting on efficacy in non-autistic children (Clementi, 2017; Belleville et.al., 2011). Research that does exist has documented decreases in sleep-related problems including bedtime resistance, sleep anxiety, and pre-sleep arousal following CBT. This suggests anxiety-focused CBT utilised within sleep interventions may be effective in treating bedtime resistance and sleep onset delay (Clementi et.al., 2016; Peterman et.al., 2016). Evidence also demonstrates improvements through CBT treatment targeting nighttime fears in primary aged children, without the addition of targeted behavioural sleep treatment (Gordon et.al., 2007; Pincus et.al., 2012).

Clementi & Alfano (2014) highlighted implementation of a CBT anxiety treatment for non-autistic children, integrating it with targeted treatment of paediatric sleep disturbance. This targeted behavioural therapy included an individualised sleep intervention based on the child's sleep problems, using cognitive and behavioural strategies. For example, identifying children's 'sleep enemies', such as irregular bed/wake times, or fear of the dark, alongside graduated extinction procedures, in vivo exposures for anxiety, and relaxation training (Clementi & Alfano, 2014). Results demonstrated reduced sleep disturbing anxiety symptoms, and reduction in accompanying sleep difficulties in non-autistic children aged 7-13.

A case study involving an 8 year old autistic boy experiencing sleep-interfering anxiety, included an anxiety-management component, within a multi-component behavioral sleep treatment (Souders et.al., 2017). Anxiety-management strategies which promote relaxation and lowering of physiological arousal were added as part of the pre-sleep bedtime routine. For example, 'relaxation practice'; taught the child awareness of how their body feels when

they are relaxed, developing awareness of the feeling and pace of their own breath, as well as progressive muscle relaxation. Massage was used to provide deep pressure tactile input to serve sensory needs, and yoga exercises addressed proprioceptive needs. Environmental components included altering the bedroom environment to ensure a cool, dark room with a night light, and removal of sleep interfering toys from the bed. Positive evening routines were introduced, with visual supports to enhance learning. A faded bedtime protocol with a bedtime pass for night waking was the behavioural component targeting sleep onset and maintenance difficulties. Clinically significant improvements in both anxiety symptoms, pre-sleep arousal and SOL were evident post-treatment, in addition to amelioration of difficulties with bedtime resistance and NW's (Souders et.al., 2017).

Two recent studies have evaluated the effectiveness of CBT-I for non-autistic adolescents experiencing co-occurring anxiety or depression. A 2020 study provided CBT-I to 23 young people aged 13-17 presenting with clinical anxiety, and insomnia (Aslund et.al., 2020). A manualised program containing core elements of standard CBT-I (i.e. sleep hygiene, circadian therapy, sleep restriction, cognitive and behavioural change) was utilised. The manualised program named ySNOOZE originally designed for adults (Reimann et.al., 2017), was adapted for use with adolescents. Parents were informed about the treatment, but did not participate in therapy (Aslund et.al., 2020). According to pre-post treatment data comparisons, statistically significant improvements were indicated for all participants, with reductions in SOL, frequency of night waking (FNW), sleep efficiency, total sleep time, depression, and anxiety (Aslund et.al., 2020). This study suggests CBT-I can improve both sleep and anxiety in adolescents, however overall data needs to be interpreted with caution due to the small sample size, and the study did not include a control group (Aslund et.al., 2020).

The aim of a study by Zetterqvist and colleagues (2021) was to investigate the feasibility and effects of internet delivered CBT-I for adolescents diagnosed with anxiety or depression. 21 participants aged 13-17 years of age were recruited, and received seven weekly modules of internet delivered CBT-I, with therapist guidance and support available via email and text throughout treatment. Therapy included an emphasis on stimulus control particularly related to device use, and sleep restriction. Regular sleep and wake times, and limitations on time spent in bed were adjusted in collaboration with participants during the treatment phase. Sessions also included sleep hygiene advice, problem solving for removing obstacles to sleep conducive behaviour, psychoeducation about anxiety, time management and relaxation strategies, cognitive restructuring, and scheduled 'worry time'. Post treatment and 4-month follow up results on insomnia symptoms were positive, with large statistically significant improvements in SOL, sleep efficiency and sleep quality for over 90% of participants (Zetterqvist et.al., 2021). Small improvements on anxiety were also noted (.2 Cohen's *d*), although a general psychological measure (SCL-90) rather than an anxiety-specific measure was used (Zetterqvist et.al., 2021).

Additionally, evidence is emerging regarding the inclusion of 'third wave' therapies such as mindfulness and visualisation techniques for increasing emotional awareness and regulation to reduce anxiety, challenging behaviours, and to improve sleep in both non-autistic, and autistic individuals (de Bruin et.al, 2014; Dykens et.al., 2014; Spek et.al., 2013). Once again, evidence exploring the utility of interventions which simultaneously address both sleep disturbance and anxiety with these strategies is extremely sparse (Souders et.al., 2017).

In summary, a small number of studies have investigated the effects of CBT for insomnia, in both non-autistic and autistic individuals, and 2 recent studies evaluated the effectiveness of CBT-I for non-autistic individuals experiencing co-occurring anxiety or depression. CBT-I contains core treatment components which generally address anxiety, and

suggest cognitive and behavioural techniques for managing anxiety and hyperarousal in the context of sleep. However to date, no studies exist which specifically address and primarily evaluate treatment of anxiety-related sleep disturbance for autistic individuals.

Effective Assessment for Developing Individualised Treatment Plans.

Numerous variables contribute to the onset and maintenance of sleep disturbance in anxious autistic youth, and the presentation of anxiety related sleep disturbance in autistic children and youth can indeed be complex. Therefore, the importance of implementing assessment procedures which provide as much detail as possible on the nature, and function of an individual child's sleep difficulties within the context of their environment and family relationships cannot be overstated. Functional Behavioural Assessment (FBA) is an empirically based, comprehensive method for identifying the most likely relationship between variables, thus providing a bridge between assessment, and the development and implementation of treatment components for specific problem behaviours. (Dunlap & Kern, 2018).

Functional Behavioral Assessment

The variety of possible contributing factors and presentations of anxiety in sleep disturbance are diverse. An assessment which provides clarity on the likely function and reinforcement of specific behaviours contributing to sleep disturbance and anxiety, aids in the establishment of accurate treatment targets (Dunlap & Kern, 2018; McLay et.al., 2018; Scott et.al., 2010). Sleep disturbance tends to present as a complex and often unique combination of antecedents and consequences which are maintaining an individual's difficulties (Hanley 2016). Research indicates a treatment approach which effectively determines and addresses the most likely function of behaviours contributing to sleep disturbance, are more effective

than generic interventions (Hanley, 2016; Hanley et al., 2014; Brown et al., 2013; Didden & Sigafoos, 2001; Kodak & Piazza, 2008).

FBA is a specific method used for collecting information about antecedents, behaviours, and consequences, to identify the function of a behaviour i.e., why a behavior is occurring (Gresham et.al., 2001; McLay et.al., 2017; Scott et.al., 2010). Numerous studies have demonstrated the effectiveness of FBA in reducing problem behaviors in autistic and non-autistic children (Dunlap & Kern, 2018; McLay et.al., 2017). Evidence suggests interventions employing FBA are more effective than interventions that are not function-based, or which rely on an assessment of topography of behavior alone (Hurl et.al., 2016; Scott et.al., 2010).

The tapestry of variables contributing to the onset and maintenance of sleep interfering behaviors are unique to every child and family, and could include antecedents such as child anxiety about separating from a parent, or a child's drive to seek sensory stimulation. The consequence may be a parent providing their presence, or a child engaging in sleep interfering behaviors such as jumping on the bed. Establishing the function of sleep interfering behaviors through FBA, supports the development of individualised behavioral interventions which are a 'good fit' for families' needs, and feasible for implementation by parents in natural settings (Turner & Johnson, 2012; Moes & Frea, 2002). FBA is utilised in this study to identify the most appropriate, individualised treatments for each child and family.

Summary of Sleep Disturbance in Anxious Autistic Children, and Evidence of Effective Behavioural Treatments.

In reviewing the literature investigating the nature and presentation of anxiety, and its impact on sleep disturbance in autistic children, several things are clear. The incidence of

anxiety disorders, and sleep disturbance is elevated within the population of autistic youth (Cuomo et.al., 2017; Delli et.al., 2018). However the exact ways in which anxiety, autism characteristics, and sleep disturbance interact with each other, in the context of a child's environment and family relationships is still unclear (Lindor et.al., 2019; Nadeau et.al., 2015; Uhde et.al., 2009; Yang et.al., 2016). Research suggests the reciprocal influences between these variables is diverse, multi-faceted, and to some degree unique to each individual (Richdale & Schreck, 2009; Uhde et.al., 2009). Evidence is emerging which indicates specific neurobiological features within autistic individuals, contribute to specific vulnerabilities towards arousal and circadian dysregulation, representative of key factors in the manifestation and maintenance of both anxiety disorders, and sleep disturbance (Carmassi et.al., 2019; Mazzoni et.al., 2018; Uhde, et.al., 2009; Veatch et.al., 2015).

Empirical evidence has established Cognitive Behaviour Therapy (CBT) as an effective treatment for anxiety in non-autistic children, and to a lesser extent, autistic children (Danial & Wood, 2013; Dummett, 2006; Keefer et.al., 2018; Scarpa & Reyes; 2011). A number of modifications to traditional CBT components tends to increase treatment effectiveness for autistic individuals, such as less emphasis on cognitive challenge, use of visual supports, repetition, and incorporation of special interests (Dowell et.al., 2018; Lang et.al., 2009; Moskowitz et.al., 2017).

There is also robust evidence for the effectiveness of behavioural interventions in the treatment of a number of sleep difficulties in non-autistic children, with growing evidence of their effectiveness in ameliorating sleep disturbance in autistic children (Cuomo et.al., 2017; Leahy & Gradisar, 2012; McLay et.al., 2017; van Deurs et.al., 2020; Wiggs; 2008). Although investigation into understanding and effectively treating anxiety related sleep disturbance is still in its infancy, particularly in autistic children, some interesting and promising links are emerging. Studies suggest circadian and arousal dysfunction may play a

larger role in the incidence of sleep difficulties in autistic children than previously believed (Geoffray et.al., 2016; Mazzone et.al., 2018). Behavioural interventions which increase sleep pressure such as faded bedtime protocols and sleep restriction are indicating effectiveness in regulating circadian function and stabilising sleep/wake cycles (Carmassi et.al., 2019; Mazzone et.al., 2018). The bi-directional relationship between sleep and psychopathology suggest sleep interventions may reduce symptoms of anxiety and depression (Orchard et.al., 2020). Despite the relatively common co-occurrence of sleep disturbance and anxiety in both non autistic, and autistic individuals, these disorders tend to be clinically assessed, and treated separately, with little evidence of integrated treatments for anxiety related sleep disturbance (Clementi , 2017).

It is encouraging overall to see the utilisation of technology, and research which continues to positively contribute to the understanding and treatment of everyday challenges like sleep difficulties for families of autistic children, representing a greater potential to improve developmental outcomes, and family well-being. However, these studies are not without methodological limitations which may affect the overall robustness and generalisability of results. Systematic reviews can provide a useful and reliable perspective on the consistency and thematic qualities of evidence (Carmassi et.al., 2019), however many studies into sleep disturbance and anxiety in autistic children involve small sample sizes, and variation in the quality of research methods, and outcome measures (Carmassi et.al., 2019; Cuomo et.al., 2017). Articles are often lacking the addition of data on post-treatment and maintenance effects, which limits our ability to evaluate the stability and lasting impact of treatments (Diaz-Roman et.al., 2018). This study aims to include formal evaluation of post-treatment effects, and seeks to provide evidence on the maintenance of treatment effects by obtaining short, and long-term follow-up data. Continued commitment to the replication of studies, and a level of responsiveness to outcomes which increases reliability and

generalisability of data, will provide a valuable contribution to the future implementation of effective treatments for anxiety related sleep disturbance in autistic children. Given the prevalence of sleep disturbance and anxiety experienced by autistic children, and the negative impact this can have on them and their families, the paucity of research aimed at increasing our understanding of effective treatments is surprising, if not concerning.

The overall aim of this study is to explore the relationship between anxiety and sleep disturbance in autistic children, by evaluating the effects of behavioural sleep treatments in autistic children experiencing anxiety. Participants who appear to be experiencing anxiety according to psychometric evaluation, and self or parent report indicates anxiety may be interfering with sleep, will be included in this study. Previous research has primarily assessed, treated, and monitored anxiety and sleep disturbance separately (Nadeau et.al., 2015; Souders et.al., 2017;). This study aims to monitor anxiety and sleep disturbance at assessment, treatment, and post-treatment stages. Treatments will include sequenced components of behavioural sleep treatment and anxiety treatment as required for each participant, individualised according to FBA assessment. Therefore, the impact of treatment components on both anxiety and sleep disturbance can be evaluated at each phase of intervention.

Research Questions:

1.) Is behavioural sleep treatment effective for autistic youth experiencing anxiety?
2.) Are treatment effects maintained over time?
3.) What is the impact of improving sleep on young people's experience of anxiety, daytime behaviour, quality of life, parent sleep and well-being?
4.) How do parents/young people perceive the acceptability and effectiveness of behavioural sleep treatments for autistic youth experiencing anxiety?

Chapter 2: General Method

This research consists of two studies. The first is a single participant case study. The second study is a multiple baseline design study, across three participants. The methods outlined in this chapter describe the general methods that applied to both studies.

The Good Nights Programme

The current research is part of The Good Nights Programme; a nationwide research study which investigates the effectiveness of behavioral sleep interventions for autistic children and children with rare genetic neurodevelopmental disorders. The sleep research team is led by three senior academics, and includes a team of psychologists and intern psychologists, PhD, and Master's thesis students.

Ethics

This study has received ethical approval from the University of Canterbury Human Ethics Committee (#HEC 2014/150). The clinical treatment and data collection for the current study was undertaken within the context of the larger study, and no further approvals or consents were required. A telephone discussion was conducted initially with prospective participants, to explain what the study entailed and ensure parents and older children/adolescents fully understood what study participation involved. Written information was provided that described the study, before consent was obtained from both parents and young people.

Setting

Interventions were carried out in participants' homes, in their natural sleep setting. Treatment implementation involved varying levels of participation from parents and children, depending on developmental age, language ability, support needs, and specific sleep difficulties. Treatment components in the first study were implemented collaboratively by

the parent and participant. Treatment components in the second study were primarily implemented by the participants, with support from their parents as required. For example, reinforcement procedures implemented by parents were included when considered necessary, to encourage participants to engage in goal behaviours. All study participants were based in New Zealand. Meetings with families were carried out either in person within the family's home, via Zoom, or at the Pukemanu Centre located on campus at University of Canterbury. Treatment support, and daily contact during intervention was maintained by phone, over Zoom, in person at the Pukemanu Clinic, or during home visits.

Participants: Inclusion/Exclusion Criteria

Children or adolescents were eligible for inclusion in this study if they were aged between 2-18 years, and met the following criteria: 1) had a formal diagnosis of Autism, confirmed by a paediatrician, registered psychologist, or psychiatrist, 2) were experiencing parent or self-reported sleep disturbance, (further evidenced through parent/self-report, video surveillance, actigraphy and/or sleep diaries), 3) were experiencing anxiety which appeared to be interfering with their sleep; according to parent or self-report, and psychometric data. Participants were excluded from this study if they experienced a medical condition which put them at risk during intervention (e.g., seizure disorder), or if they were otherwise unable to follow procedures in this study (e.g. significant family or personal events impacted their ability to carry out treatment at the time of the study).

Recruitment

Participants were recruited through referral from organisations who provide services and support to autistic children and families, professional contacts, or through social media support groups. Interested families were provided with information about the purpose and processes involved in the study, and prompted to make direct contact with the research team

via email. An initial phone screening interview was then undertaken to ensure eligibility, and for families to make an informed decision about commitment to the study. Informed consent was then obtained following a family's choice to proceed with study participation.

Research Design

Research outcomes were evaluated using a case study (Study One) or single-case multiple baseline across participants design (Study Two). A single-case multiple baseline across participants design is appropriate for determining the effectiveness of an intervention with individual clients. Single case research is highly appropriate for use with autistic youth, given the heterogeneity of this population, as the effect of individualised treatments can be examined in detail both within and across participants (Carr, 2016; Kazdin, 2019).

Participant Characteristics

Four participants (two females and two males) met criteria for inclusion in this overall study. All were aged between 13-16 years (M = 13.75 years), and were formally diagnosed with Autism by registered Psychologists in New Zealand. Pseudonyms have been used to ensure confidentiality for all participants and their families. Two of the participants were NZ European, one of Asian descent, and one of Māori descent. None of the participants had been formally diagnosed with an anxiety disorder. However all participants' psychometric data revealed parent and/or self-reported clinically elevated anxiety scores, and all participants reported anxiety which interfered with their sleep – primarily intrusive thoughts which they could not control, and feeling tension or worry which impaired their sleep. A summary of participants' characteristics is presented in Table 1 below.

Table 1

Summary of Participant Characteristics

Name	*Age*	*Gender*	*Diagnosis*	*Sleep Problems*	*Medication*
Wynter	13yrs	Female	Autism	Co-sleeping, SOD, CC, NW	Melatonin
Gemma	16yrs	Female	Autism	SOD, NW	No meds
Oscar	13yrs	Male	Autism	SOD, NW	No Meds
Fletcher	13yrs	Male	Autism	SOD, NW	No Meds

*CC = Curtain Calls, NW = Night Waking, SOL= Sleep Onset Latency,

Measures

Sleep Diaries

Sleep diaries are commonly used for collecting information about sleep (Blampied, 2013; McLay & France, 2016). Sleep diaries provided a daily log of individuals' sleep patterns, and measured change in sleep and sleep-related behaviours across all study phases. All sleep diaries in both studies recorded information about what time participants went to bed, the duration of time till sleep onset, the frequency and duration of night awakenings, and what time they woke up for the day. Participants also noted other factors that disturbed their sleep, such as thoughts, or environmental disturbances. Study One participant's diary also included information on the frequency and nature of parental response to curtain calls, and the frequency and nature of co-sleeping occurrences. Participants in both Study One and Two were also provided with a rating scale for recording their level of worry or anxiety at sleep onset and night waking.

The participant in Study One shared responsibility for recording sleep diaries, and chose to collaborate closely with their parent. One participant in Study Two took sole

responsibility for recording all sleep diary data (Gemma). The remaining two participants in Study Two shared responsibility for data recording in sleep diaries, seeking collaboration with parents as they required. Sleep diary data was recorded daily throughout the study during baseline, intervention and follow-up phases.

Actigraphy

Actigraphy is a method of data collection through a miniaturised computerised device normally worn on the wrist like a watch, to monitor and store data generated by movement (Sadeh & Acebo, 2002). An actigraph can collect and store data for up to two weeks, and is accepted as a valid and accurate measure of sleep/wake patterns (Sadeh & Acebo, 2002). Actigraph data collection was used to triangulate sleep diary data with two participants in Study Two.

Videosomnography

Infrared video cameras are used to record objective data on children's bedtime and nighttime behavior. Videosomnography provides the opportunity to observe child behaviour in their natural setting during the sleep period, capturing sleep and wake periods throughout the night (Camerota et.al., 2018). Video recording equipment (Swann Advanced-Series DVR4-1200, D-Link HD Cloud, or TP-link Tapo camera) was used with one participant in Study Two. When used, cameras are positioned inconspicuously facing the child's bed, and recording commences from the time the child is put to bed, until they get up for the day. Video footage can provide additional information on children's sleep patterns and behaviour throughout the night, including parent-child interactions (Sadeh 2015). Video recording can also provide useful and effective interobserver agreement (IOA) data, and triangulation of data, providing a measure on reliability of sleep diary data. Only one participant, in Study Two consented to the use of video recording. In this case, video data was recorded for 25%

of the baseline phase, and treatment phase. No follow up data was recorded, as this participant retired from the study before the follow-up phase.

Sleep Assessment Measures

The following psychometric measures were used to gather pre-treatment assessment data for both Study One and Two.

Sleep Assessment Treatment Tool (SATT; Hanley, 2005)

The SATT is a functional assessment tool designed as an open-ended interview, to provide clarity around specific sleep interfering behaviors, and environmental variables which may be contributing to sleep disturbance (Jin et.al., 2013). Specific interview portions target information on child sleep history, specific sleep problems such as sleep onset delay or bedtime resistance, current sleep schedules and dependencies, antecedent and consequence conditions which may be related to the sleep disturbing behavior, and formulation of parent and participant sleep goals. The SATT is used as part of the FBA Assessment, to guide initial clinical interview questioning, and target relevant information to inform treatment planning, and has been widely used in research (Hanley, 2005; Jin et.al., 2013).

Sleep Outcome Measures

Child Sleep Habits Questionnaire (CSHQ; Owens et.al., 2000)

The CSHQ is a structured parent-report instrument which provides comprehensive screening of behavioral and physiological aspects of sleep. It contains 48 items, measuring sleep onset, bedtime resistance, sleep anxiety, night waking, parasomnias, sleep disordered breathing and daytime sleepiness. Items are scored on a 3-point Likert scale with a Total Score, and 8 sub-scale scores relating to key sleep domains. Higher scores reflect greater severity of sleep problems (Souders et.al., 2017), and the Total Score clinical cut-off is $\geq$41. The CSHQ has good psychometric properties (Hodge et.al., 2012), and acceptable internal

consistency for community and clinical samples (Owens et.al., 2000). The CSHQ demonstrates validity, with adequate sensitivity (0.80) and specificity (0.72) to distinguish between clinical and control samples (Owens et.al., 2000). The CSHQ is widely used in sleep research with non-autistic children, and commonly used in research with autistic children (Krakowiak et.al., 2008; Mazurek & Sohl, 2016). This screening tool was used to inform the FBA, and evaluate changes in sleep behavior from baseline to post-treatment.

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

The Adolescent Sleep Hygiene Scale is a 33 item self-report questionnaire which provides information on specific sleep hygiene behaviours and habits which can impact on adolescent sleep quality and quantity (Storfer-Isser et.al., 2013). The ASHS provides measurement according to nine subscale scores: Physiological factors, Behavioural Arousal factors, Cognitive factors, Emotional factors, Sleep Environment factors, Sleep Stability factors, Daytime Sleep factors, Substances factors, and Bedtime Routine factors. The mean of all nine subscales provides a Total Score. Higher scores indicate better sleep hygiene practices (LeBourgeois et.al., 2005). The ASHS demonstrates acceptable psychometric properties, and internal consistency (a =.80 – a = 0.84) (de Bruin, 2015; Storfer-Isser et.al., 2013). Data was collected through this measure to inform the FBA, develop appropriate treatment targets, and evaluate change post-treatment.

Child Anxiety and Well-being Measures

The measures described below were administered during baseline and post-treatment to assess pre and post-treatment adolescent behaviour, anxiety, and well-being.

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

The Pediatric Quality of Life Inventory is a 23-item parent and self-report questionnaire which measures an individual's health-related quality of life. The

questionnaire is scored on four core dimensions: Physical Functioning (eight items), Emotional Functioning (five items), Social Functioning (five items), and School Functioning (five items), generating three summary scores on Physical Health, Psychosocial Health, and an overall Total Score (Varni, 1999). Responses are rated on a 5 point Likert scale, with higher scores indicating a perceived higher quality of life.

The PedsQL has been utilised in research with autistic children, and demonstrates reliability as a measure in young people, with an internal consistency (Cronbach's alpha) of a=0.90 (Viecili & Weiss, 2015). This measure was completed pre- and post-treatment, to evaluate the effects of treatment on children's quality of life.

Child Behavior Checklist for Ages 6-18 (CBCL), and Youth Self-Report (YS-R; Achenbach & Rescorla, 2001)

The CBCL and YS-R provide a parent and young-person report of child and adolescent behavioural and psychosocial functioning (Achenbach & Rescorla, 2001). Responses to 100 questions about specific behaviours are based on a 3-point Likert scale (0=not true, 1=somewhat true, and 2=very true, or often true). Scores are grouped into nine syndrome scales: Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behaviour, Aggressive Behaviour, and Other Problems. Higher scores indicate worse problem behaviour. Subscale scores are categorised into Internalising Problems and Externalising problems, with an over-all Total Problem Behavior Score. Total (T) scores place the child's difficulties into the Normal (<60), Borderline (60-63) or Clinical Range (≥64). Adequate internal consistency (a =.63 - .79), and high test-re-test reliability (r =.90*)* has been demonstrated with the CBCL and YS-R (Achenbach & Rescorla, 2001). The Child Behaviour Checklist is well-established and widely used as a measure amongst non-autistic children and adolescents, and is accepted as a valid tool for assessing emotional and behavioural symptoms in autistic children and youth

also (Guerrera et.al., 2019; Pandolfi et.al., 2014) This measure was taken at assessment, and post-treatment to provide a measure of change in participants internalising and externalising behaviours post-intervention.

Parent Well-being Outcome Measures

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

The PSQI is a 19 item self-report measure which assesses sleep disturbances and overall sleep quality in adults over the previous month. Items are grouped into seven subscales, measuring Sleep Latency, Sleep Duration, Sleep Medications, Sleep Efficiency, Sleep Disturbances, Subjective Sleep Quality, and Daytime Functioning. Responses are rated according to frequency, and subscale scores are added to yield an overall PSQI score ranging between 0-21, with higher scores indicating more severe sleep impairment. The PSQI has been reported as possessing good psychometric properties, with internal reliability of .83, and test-re-test reliability of .85 (Buysse et.al., 1989). This measure has been used often in clinical research with autistic children, to evaluate sleep quality in parents and caregivers (Jellet & Porter, 2011: Hodge et.al., 2013; Meltzer, 2008). Parents completed this questionnaire prior to and following treatment, to evaluate changes in the quality of their sleep.

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

The DASS-21 is a 21 item self-report screening questionnaire covering symptoms related to stress, anxiety, and depression. This measurement can be administered as a clinical interview or filled out privately by parents before, and after the intervention process. Respondents indicate on a 4-point Likert scale the extent of their experience of Anxiety, Depression or Stress symptoms over the past week. Higher scores indicate increased severity. Although the DASS-21 is not a diagnostic instrument, it is a useful screening tool,

having demonstrated high internal consistency (*a* = .82 - .93*)* (Henry & Crawford, 2005). For both studies, the DASS-21 was completed by parents before and after treatment, then scores pre- and post-treatment were compared, to indicate change in parent mental health/well-being following treatment.

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

The TARF-R is a 20-item parent-report questionnaire, designed to provide a measure of the social validity of interventions. Responses are on a 7-point Likert scale, with 17 items addressing treatment acceptability, and 3 items evaluating severity of the problem, and parental understanding of the treatment process. Higher scores indicate greater acceptability (McLay, et.al., 2018). The TARF-R demonstrates acceptable reliability (*a=.92),* and has been used in various settings, including as a measure in autism research (McLay et.al., 2019; McLay et.al., 2021; Waddington et.al., 2020). The TARF-R was completed by families following treatment, to gain information about their perception of the acceptability of the intervention process, and satisfaction with treatment outcomes.

Study Procedures

Study One implemented a single case A (Baseline) B (intervention) design, with short-term and long-term follow up. Study Two employed a single case multiple baseline design across participants. Intervention plans and phases were individualised according to each participant's needs. Treatment phases are indicated on the graphed data with a phase change line. Treatment effects were evaluated by comparing treatment and follow up data with baseline data and assessing for change through visual analysis.

Assessment and FBA

Assessment included a comprehensive clinical interview with parents and participants. Further assessment data was gathered through the assessment tools outlined

previously. Clinical interviews were undertaken with parents and participants by registered intern psychologists, following the standard intake interview format used by Child and Family Psychologists at the Pukemanu Centre, under the supervision of a registered Psychologist. The interview process included confirmation of confidentiality, and participant consent. Interview questions elicited information about the participant's current sleep difficulties including the history and development of sleep disturbance, and environmental factors likely contributing to their current sleep behaviour. Interviews also allowed for the opportunity to observe family interactions and strengths, and develop rapport with the family, concluding with discussion around family needs and sleep goals. Data from the interview, SATT, QABF, sleep diary, and video recording were used to inform the FBA, and thus develop a comprehensive, individualised sleep treatment plan for each participant and their family.

Baseline

Participant families were randomly assigned to either a 1, 2- or 3-week baseline period where data was collected through sleep diaries, plus videosomnography or actigraphy. A start date convenient to the family was arranged to ensure the baseline phase was immediately followed by the onset of intervention. Baseline measures continued until a stable pattern was obtained, to facilitate reliable measurement of treatment effects (Kazdin,2019). Parents and participants were asked to continue their usual routines during this phase, to ensure information gathered represented a reliable reflection of their current sleep.

Treatment Procedures

Following the collection of baseline data, the sleep team formulated individualised treatment plans, based on information gathered through assessment and FBA. The proposed treatment plan was then shared and discussed with participants and their family, to reach

mutual agreement on timing and implementation of planned procedures, and to establish a collaborative, supportive approach to reaching the family's sleep goals (Jin et.al., 2013). Treatment plans included addressing sleep hygiene practices, in keeping with 'best practice' procedure for child sleep interventions (Vriend et.al., 2011). Treatment components were individualised according to FBA results, and the needs, strengths, and preferences of the family. Individual goals were established with each participant and their family prior to treatment. Attainment of sleep goals for each participant were evaluated according to the goals established prior to treatment, psychometric measures taken before and after treatment, and feedback from the participant and their family through a post-treatment interview.

Daily phone calls were made to families during the intervention phase, with parents and participants given the option of contacting the intern psychologist by email or phone for further support, as required. Daily communication enabled the intern psychologist to regularly document progress, provide guidance and encouragement to families, and address any difficulties the participant had promptly, to increase the potential for positive treatment effects. Active intervention continued until a stable, significant reduction of sleep disturbance was evident over a 10-14-day period, and/or until participants and families indicated they were satisfied with treatment outcomes, or chose to withdraw.

Maintenance

During the maintenance phase, no contact was made between the psychologist and the participant family for 4-6 weeks, to enable the participant to consolidate the learning they gained through treatment, and apply sleep strategies independently within their daily lives (Sanders & Burke, 2013). Post-treatment psychometrics were completed by the participant and family during this phase, and a post-treatment interview was undertaken with parents and participants to gain feedback on their experience of the intervention process. The interview provided information on their overall understanding of treatment purpose and procedures,

perceived strengths and challenges in implementing treatment, and their perspective on the impact of treatment outcomes on family life and well-being.

Short and Long-term Follow up

Short-term follow up included 1 week of data collection through sleep diaries and (in Study Two) videography or actigraphy immediately following the maintenance phase (6 weeks post-treatment), and again at 10-12 weeks post-treatment (long-term follow up). Data was then analysed to establish whether treatment effects were maintained over time.

Data Analysis

Visual analysis techniques are traditionally employed when analysing data in multiple baseline design studies (Ferron et.al., 2009). Data collected from the sleep diaries, video footage and actigraphy were examined, to evaluate dependent variables. A functional relationship between the independent and dependent variables is considered present when a clear, consistent change in target behaviors is demonstrated by systematic manipulation of the independent variable (the intervention) (Vannest & Ninci, 2014). Stable baseline data and at least three demonstrations of significant change co-occurring with treatment, is considered a valid indication of treatment effect (Kazdin,1981; Vannest & Ninci, 2014). Additionally, an important consideration in evaluating meaningful change, is a treatment effect which is significant enough to represent real-world impact for participants and their family. Pre and post outcome data from the CSHQ,ASHS, CBCL, PedsQL, PSQI, and DASS-21 assessments provided data to illustrate 'real-world' treatment effectiveness, if they reflected positive changes in sleep habits, anxiety, quality of life and well-being. Evaluation of TARF-R results and post-treatment interviews also provided feedback on participants' and families' perceptions regarding acceptability and social validity of the intervention.

Chapter 3: Case Study 'Wynter'

Wynter (pseudonym) was a 13 year old female with a recent diagnosis of 'Autism Spectrum Disorder Level 2' (according to DSM-V classification the individual 'requires substantial support') . Wynter lived at home with her mother. Wynter was referred to the Sleep Study by a private psychologist. Wynter had age appropriate expressive language, however, she found it difficult to follow some verbal instructions due to receptive language difficulties.

Presenting Concerns

Wynter was reported by her mother (Jenny - pseudonym) to have long-standing problems with sleep onset delay (SOD), night waking (NW), and a dependence on parental presence in order to fall asleep. Her mother slept in Wynter's room, going to bed at the same time as Wynter, and sleeping in her room for the whole night. Wynter followed a consistent bedtime routine. If this was completed before 8.20pm, she was rewarded with an interactive game of her choice before bedtime. Following the game, Wynter would take 0.5 mg melatonin, do a deep breathing exercise, and be in bed for lights out at 9pm. This routine had reduced bedtime resistance. Sleep onset took approximately 30 minutes. Wynter agreed she had a sleep problem, and reported she would wake 3 to 4 times per night for 5-35 minutes, up to 4 times per week. Wynter stated that 'thoughts' would sometimes keep her awake, and she felt safer with her mother in the room. On some occasions she would wake her mother saying she couldn't sleep, and her mother would get into Wynter's bed, which re-settled her quickly. The current co-sleeping arrangement was occurring every night. Wynter typically woke at 7-7.30am in the morning.

History of Sleep Disturbance

According to parent report, Wynter began having sleep difficulties at 6 weeks of age, being difficult to settle, and was awake for 'hours on end'. Following several years of intermittent sleep disturbance in early to mid-childhood, Wynter began co-sleeping with her mother.

Wynter struggled to articulate why falling asleep without her mother present was difficult, but noted exercise during the day made it easier to fall asleep because she was physically tired, and excitement or worries made it difficult to fall asleep because she was "all hyped up". Wynter said deep breathing exercises were "boring", but helped her feel more relaxed. She was not sure if they helped her to fall asleep faster. Wynter reported she wanted to improve her sleep, but expressed reluctance and uncertainty about her parent's goal for her to sleep in her own room independently. Previous efforts to improve Wynter's sleep included drinking tart cherry juice (which contains a minute amount of melatonin) and listening to various calming sounds such as waves on the beach during the sleep onset period. Wynter had also experimented with a weighted blanket which she reported had become annoying as it restricted her movement.

Goals for Sleep Treatment

Wynter's mother's goals for sleep were: 1) for Wynter to be able to fall asleep consistently within 20 minutes, and 2) to sleep independently (without parental presence) throughout the night in her own room. Her parent also noted it would be beneficial for Wynter to feel comfortable sleeping over at friend's houses, but that was not a significant priority for sleep treatment. Wynter's goals for her sleep were: 1) to fall asleep easily within 15-20 minutes, and 2) to sleep through the night without waking up.

Functional Behavioural Assessment

Various antecedent factors contributing to Wynter's sleep difficulties were identified through FBA. The achievement of a low state of arousal for a sufficient period of time is essential to achieving sleep onset. A predictable chain of sleep promoting behavioural and environmental conditions which act as reinforcing cues for sleep (discriminative stimuli), are important for enabling children to achieve sleep onset, or re-initiate sleep in the night. Inconsistent or inappropriate discriminative stimuli prior to sleep (inadequate stimulus control), contribute to the maintenance of sleep disturbance (Mindell et.al., 2009). For example, reliance on parent-provided discriminative stimuli such as a cuddle, or environmental factors which interfere with behavioural quietude such as a noisy, or overly warm sleep environment. Consistent calming pre-sleep routines, and a calm, comfortable sleep environment with the child in their own bed, promotes successful independent sleep onset (Blampied & France, 1993; Mindell et.al., 2009). Wynter was dependent upon her mother's presence, and specific environmental conditions to achieve sleep onset. Wynter's difficulties with self-soothing resulted in inconsistent sleep times, with her falling asleep at various times of the evening, and requiring the same discriminative stimuli to be present to re-initiate sleep if she woke in the night.

Overall, FBA indicated Wynter's sleep interfering behaviours served various functions. SOD and NW's were positively reinforced through social attention from her mother in the form of cuddles, or providing her with tangible items. Wynter was also able to avoid unpleasant feelings of worry and cognitive or physical hyperarousal from excited or anxious thoughts and feelings, through her mother's verbal reassurance and presence. It was hypothesised that the function of Wynter's sleep disturbance was access to social attention, and tangible items.

Baseline Phase

The baseline phase lasted for 14 days. During this time sleep data (i.e., CC's, SOL, FNW, DNW) were collected. Self-reported ratings of anxiety were recorded daily on a visual 'Worry Meter' scaled from one to five, with one being 'relaxed', 2 'a little worried', 3 'worried', 4 'very worried', and 5 'very worried, panic'. Anxiety ratings were self-reported by Wynter, while sleep diaries were completed primarily by her mother, with occasional input from Wynter.

Treatment Phase

Intervention consisted of 3 active treatment phases, which last 206 days (day 15-220) not including baseline. This was followed by a maintenance phase, with STFU and LTFU phases. Details of each phase are outlined below.

Treatment Phase One

Treatment Phase One lasted for 32 days (day 15-47), and involved practicing relaxation strategies every evening before sleep, and utilising a memory box. Wynter was rewarded with a preferred food item for practicing her relaxation strategies.

Relaxation Strategies. Relaxation strategies were implemented to support Wynter in developing self-calming skills to manage her sleep independently at sleep onset, and throughout the night. Three relaxation strategies were introduced to Wynter in one session with the researcher, (accompanied by an intern psychologist) prior to commencing treatment. The three strategies facilitated a low arousal state required for sleep by slowing down breathing, relaxing the body, and calming the mind (reducing physiological, emotional, and cognitive arousal). Strategy one involved a re-introduction of deep breathing. Wynter was invited to imagine a batch of warm cookies coming out of the oven, slowly breathing in to smell the cookies, and slowly breathing out to cool them down, while placing her hand on her

stomach to assist with drawing her breath from the diaphragm area (Lowenstein, 2016). Strategy two introduced visualisation. This researcher encouraged Wynter to choose her own calm environment to visualise. Wynter was guided by a narrative from the researcher to focus on the sounds, feelings and peaceful sensations to establish her experience of visualisation. Strategy three was a Progressive Muscle Relaxation procedure (PMR). Wynter was guided by the researcher to lie down and mentally focus on specific parts of her body by tensing, then relaxing her muscles in a sequential manner, starting with her toes, feet, and legs, and finishing with her neck and face (Lowenstein, 2016). Wynter identified deep breathing and visualisation as her preferred strategies, and agreed to practice these strategies daily as part of her pre-sleep routine. Wynter was also encouraged to practice her strategies as regularly as possible during the day.

Memory Box. Wynter had expressed a worry that she would forget special memories. She reported this was interfering with her sleep, as she would spend time mentally rehearsing her memories at night to ensure she could recall them. Therefore, Wynter was also introduced to the 'Memory Box' during the pre-treatment session. Wynter decorated her own memory box to store written and visual records of special memories. Wynter was encouraged to look through her memory box in the early evening, before carrying out her other bedtime routines.

The first weeks of treatment occurred during the Christmas break, and the family were on holiday for some of that time. Wynter's goals were set to accommodate for this change in normal routine. Her goal for treatment weeks one, two and three while on holiday was to practice relaxation strategies before bed, and use her strategies to re-initiate sleep if she woke in the night, rather than waking her mother. Wynter was supported with gradual preparation towards treatment Phase Two, by regularly discussing the steps planned for the next phase of

treatment in the weeks prior to implementation. Phase Two began once the family were back home.

Treatment Phase Two

Faded Parental Presence: Co-sleeping and Sleep Onset. Phase Two introduced faded parental presence, and began on Day 48, lasting till day 157. Steps towards the goal of independent sleep were shaped by Wynter and her mother, in collaboration with an intern psychologist. Wynter and her mother expressed a preference for small and gradual steps towards independent sleep. Step one involved Wynter falling asleep at bedtime without her mother in the room 2 times per week, and planned to be slowly built up to independent all night sleep 7 times per week in collaboration with the family, and the intern psychologist. Although a mattress remained in Wynter's room, her mother planned to go to bed in her own room once Wynter was asleep.

Reward for falling asleep independently. Wynter received a small reward in the morning if she managed to fall asleep without her mother being present, and also received a marble in a jar, with an initial quota of three marbles earning the reward of an activity or small item of her choice on the weekend. A list of appropriate weekend rewards had been agreed upon prior to commencing this phase. Rewards were faded out as Wynter progressed towards the goal of independent sleep. The frequency of rewarding her in the morning was reduced from every day to every second day, then every third day, and so on. Treatment Phase Two continued for 110 days, ending on day 157. Several treatment alterations were made throughout this phase, and are described below.

Planned Co-sleeping and Independent-sleep Nights. Steps towards eliminating co-sleeping during the night were more firmly structured from day 91, as co-sleeping had been occurring intermittently in response to Wynter requesting her mother to sleep in her room.

The re-defined goal was for Wynter to sleep independently 5 nights of the week, with her mother sleeping in Wynter's room two nights per week; on Thursdays and Sundays.

Planned Worry Time. The March 2020 five-week Covid-19 Lockdown commenced on Day 118 of treatment. Wynter began to resist carrying out bedtime routines, and getting to bed on time. She began to raise health-related concerns when her mother came to say goodnight. A time to discuss her worries during the day outside of the sleep environment, was planned and implemented from day 123. Reinforcement for compliance with bedtime routines was also re-introduced. Wynter was rewarded with one marble in the jar for starting her bedtime routine on time, and another marble for being in bed and not discussing worries when her mother came to bid her goodnight.

Planned Co-sleep Nights Phased Out. On Day 136, planned co-sleep nights (co-sleeping on Thursdays and Sundays) were eliminated(shifting to independent sleep 7 nights per week) with both Wynter and her mother's agreement.

Treatment Phase Three

Re-structured Fading of Parental Presence at Sleep Onset. Following the elimination of co-sleeping, Wynter's SOL, and CC's at sleep onset began to increase again, and Wynter's mother's responses to these bids for attention were variable. Jenny would sometimes respond to Wynter's bid for attention immediately and briefly, or delay responding to Wynter's bids for attention at times, or lie with Wynter until she fell asleep at other times. On Day 158 of intervention, Wynter and Jenny agreed to a consistent plan in which her mother would sit with her for 12 minutes every night when she went to bed. This was faded to 7 minutes on day 193, and then eliminated on day 200 of treatment. Wynter's mother was encouraged to provide a small reward for Wynter each time she fell asleep and slept through the night independently. Treatment concluded on day 220 as Wynter and her

mother expressed satisfaction with treatment outcomes, and a confidence to continue with sleep routines independently.

Short Term Follow Up (STFU) and Long Term Follow Up (LTFU).

1 week of sleep diaries was collected at 6 weeks (STFU) and 12 weeks (LTFU) post-treatment for short- and long-term follow-up, respectively.

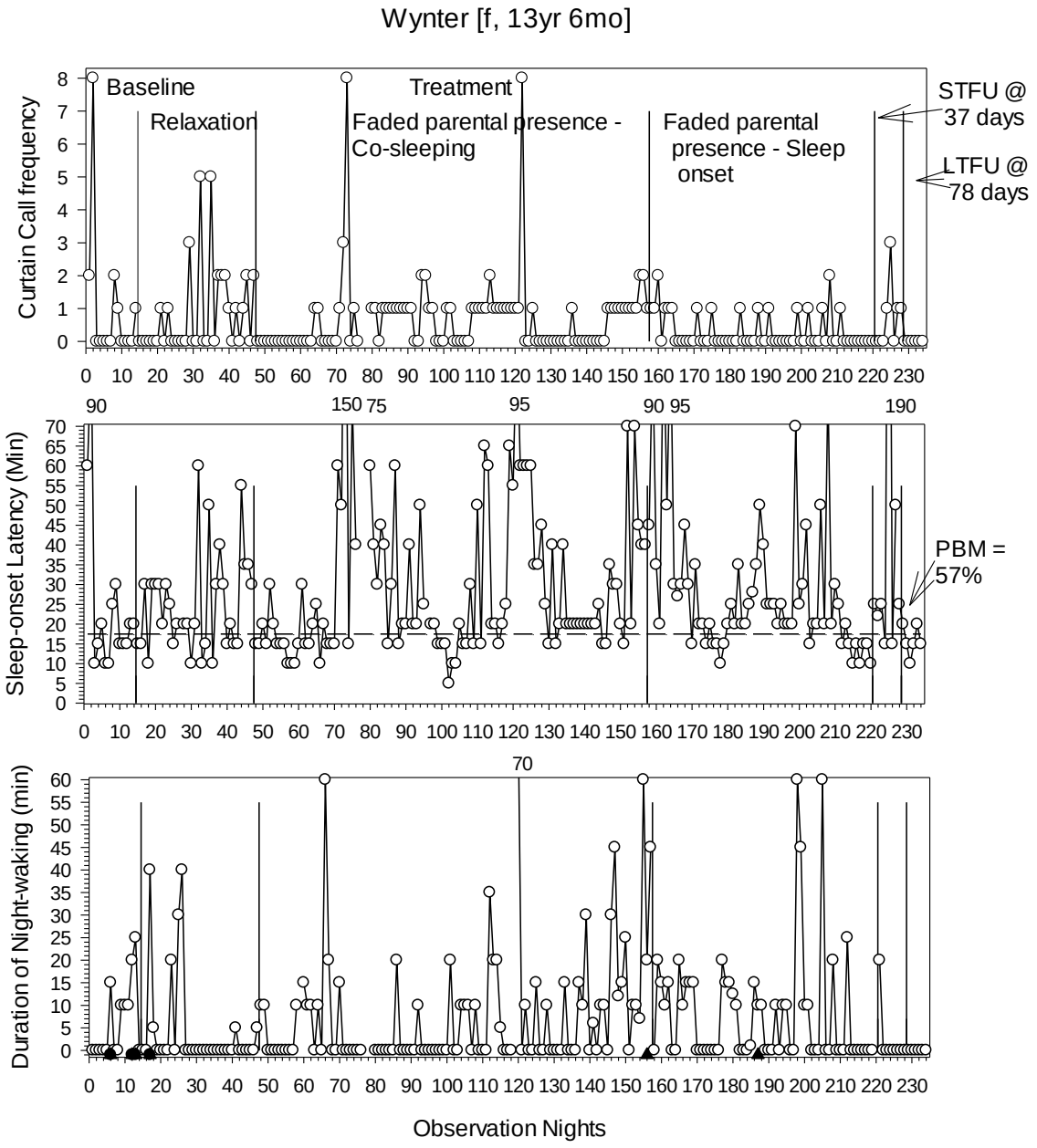
Results

Sleep Outcomes

Wynter's treatment lasted a total of 206 days (not including baseline). Sleep outcome data is presented in Figure 1, for SOL, CC's, FNW's, and DNW's. Treatment Phase Two occurred during the Covid-19 Lockdown in NZ. Sleep diary data is missing for nights 77-79, due to Wynter's mother being away on holiday. Anxiety ratings were not provided by the family on nights 3, 7,8, 13,15, 28,29,35,78,79,80,82,83,87,88, 115, and 186-192. All results from this study are discussed in detail in Chapter 6.

Figure 1

Curtain Call (CC) Frequency, Sleep Onset Latency (SOL), Frequency (FNW) and Duration of Night Wakings (DNW) for Wynter across Sleep Phases



Notes

1. For the SOL and DNW graphs, the numbers along the top of the graph state the duration of the out-of-scale latency or night waking respectively (in minutes).
2. For the DNW graph, the filled triangles along the x-axis mark nights of more than 1 night waking. In *every* case this was 2 wakes except for nights 156 and 187 where there were 3 wakes. All other non-zero wakes were from 1 wake.
3. STFU = Short-term Follow-up. LTFU = Long-term Follow-up.

Curtain Calls (CC's)

CC's ranged between 0-8 during Baseline (*M* =1) . CC's reduced slightly during Phase One, ranging from 0-5 (*M* = 0.84). Bursts of increased curtain calls were evident on nights 32 and 35 during a family holiday. On days 37-39, 3 consecutive nights of 2 CC's occurred as the family re-settled into home routines. CC's steadily decreased over nights 40-47, followed by evidence of possible treatment effect, with a settled period of only 2 nights of single curtain calls occurring during nights 48-70.

The CC's average score again reduced very slightly in Phase Two (M =.66). CC's ranged from 0-3, with two outliers of 8 CC's on night 73, and night 122. Higher CC's correlated with specific stressful life events reported in sleep diaries, such as Wynter's mother being away from home (night's 73-82), and the beginning of Covid-19 Lockdown in NZ (night 122). A sustained period of 1 CC per night for 16 nights occurred on nights 109-125. It was noted in sleep diaries that Jenny provided her presence when requested (in response to the CC), and remained in the room until Wynter fell asleep on these nights.

During treatment Phase Three there was a stable reduction in CC's, with a range of 0-2 (*M* = 0.28). A significant treatment effect is evident towards the end of treatment, with parental presence eliminated on day 200, and 0 CC's recorded for the final 10 days of treatment. Improvements were largely maintained at follow up, with CC frequency ranging from 0-3 (*M* = 0.71) at STFU. Treatment gains increased at LTFU with CC's ranging from 0-1 (*M* = 0.14), indicating a stable drop in CC behaviour compared to baseline.

Sleep Onset Latency (SOL)

Wynter's SOL was variable throughout baseline and treatment phases. A slight reduction in SOL was noted towards the end of treatment Phase Three, with improvements partially sustained at STFU and increased at LTFU. Bursts of extended SOL tended to

correlate with life stressors reported in sleep diaries, such as the Covid Lockdown, commencement of the school term, or mother's absence.

SOL ranged from 10-90 minutes during baseline, with a median score of 17.5 minutes. Sleep onset remained variable throughout treatment Phase One (day15-47), ranging from 10-60 minutes, with 34.3% of nights falling below the Baseline median (PBM) score. Wynter's longest SOL during treatment is recorded during her mother's absence from home, on night 73 of treatment (150min). Variability in SOL continued through treatment Phase Two (day 48-157). Nights of extended SOL correlated with stressful life events recorded in sleep diaries. Sleep onset ranged from 5-150 minutes in Phase Two, with 33% PBM. Indication of treatment effect appears minimal overall at treatment Phase Three, with a range of 10-95 mins, and 23.8% PBM, however a notable reduction and stabilisation in SOL occurred late in this treatment phase, with a range of 10-15min, and PBM score at 100% during the final week of treatment. Evidence of treatment effect is largely sustained at short-term follow up, with the exception of one outlier of 190 minutes, and increased at LTFU. Scores ranged from 10-25 minutes, with a PBM score of 57.1% at LTFU. Follow-up data indicated a modest improvement in SOL when compared to baseline. Overall, SOL is highly variable, and statistically represents evidence of minimal treatment effect.

Frequency of Night Waking (NW)

Frequency of night waking was problematic for Wynter at baseline, with a range of 0-2 wakings, and NW's occurring on 42.8% of nights during this phase. In Treatment Phase One, NW's occurred on 18.1% of nights, and ranged from 0-3. A consistent reduction in NW's is seen in the second half of treatment Phase One with no NW's from day 27- 47. No occurrence of NW's continued into Phase Two, with 0 NW's from day 48-57 during a family holiday with friends. NW's re-occurred upon return home with the re-commencement of faded parental presence procedures. The cluster of night wakings on consecutive nights from

day 58-62 possibly represent a post-extinction burst in response to re-initiation of fading parental presence. Sleep diary data indicates Wynter's mother co-slept through the rest of the night (following wakings) at Wynter's request on these nights (58-62). NW's occur intermittently throughout the rest of treatment Phase Two. NW's ranged from 0-3 in treatment Phase Two, with wakings occurring on 30.9% of nights during this phase. A reduction in NW's occurred in treatment Phase Three, with NW's occurring on only 12.6% of nights overall during this phase, and a range of 0-3. NW's were consistently absent for the last 3 weeks of treatment. STFU and LTFU reveal a continuation of treatments gains, with just one night waking recorded during STFU, and no night wakings throughout the LTFU phase.

Duration of Night Waking

The duration of night wakings ranged from 10-25 minutes at Baseline, (*M* =15), and 5-40 minutes (*M* =20.7) during Phase One of treatment. Similar DNW's occur across all treatment phases. During Phase Two DNW's ranged from 5-70 minutes (*M* =21) and Phase Three ranged from 10-60 minutes (*M* =20). Longer DNW's appeared to correlate with specific events recorded in sleep diaries, such as Christmas, her mother going on holiday, or the Covid-19 lockdown. Night wakings are eliminated by the final week of treatment. Frequency and duration of NW's remained significantly lower than baseline at STFU with one NW of 20 minutes. NW's did not occur at LTFU. Overall, treatment resulted in a significant and stable reduction in the frequency and duration of NW's from baseline to follow-up.

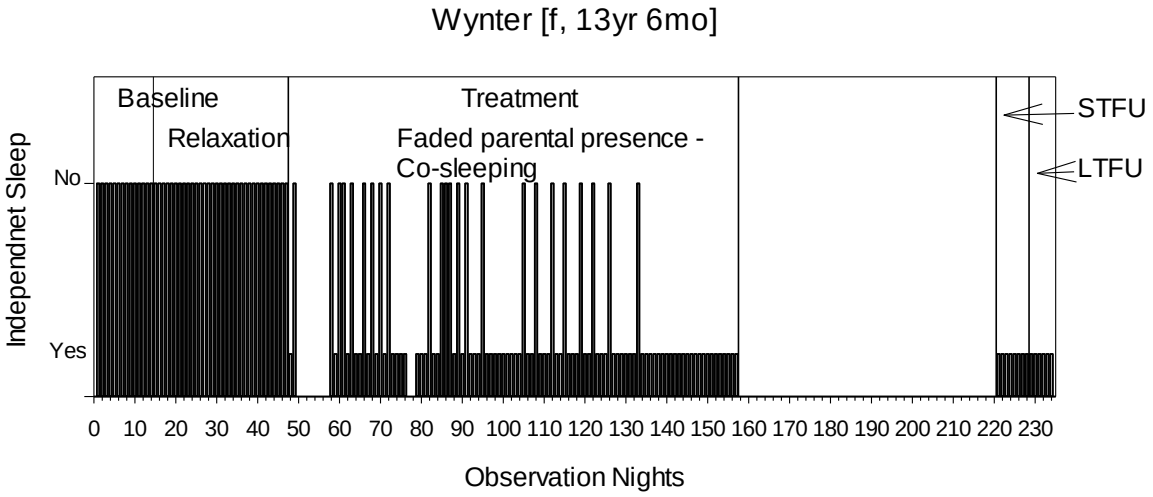
Independent Sleep/Elimination of Co-sleeping

Independent sleep was defined as sleeping the whole night without the presence of a parent – from bedtime, until getting up in the morning. Some spontaneous behaviour change regarding independent sleep at *sleep onset* was evident towards the end of baseline, where Wynter fell asleep independently, before her mother came to co-sleep on nights 11-14 (this behaviour had not occurred previously).

The occurrence of independent all night sleep is represented in Figure 2. Rewards for independent all night sleep began on night 48, when Wynter achieved her first ever full night of independent sleep. Co-sleeping occurred intermittently from nights 49-90, with Wynter's mother sometimes agreeing to Wynter's request for co-sleeping. On day 91 of treatment, the structured plan for restricting co-sleeping to 2 nights per week commenced. Wynter consistently achieved this goal from nights 91-136. On night 136, co-sleeping was eliminated, with independent sleep commencing for 7 nights per week. Wynter achieved this goal consistently, and was declared 'no longer co-sleeping' on night 158 of treatment, following 3 weeks of independent sleep every night. STFU and LTFU data indicates independent sleep was maintained consistently, with no episodes of co-sleeping reported during follow up phases. Overall, independent all night sleep was clearly and consistently achieved as a result of treatment.

Figure 2

Occurrence of Independent Sleep Throughout all Treatment Phases for Wynter

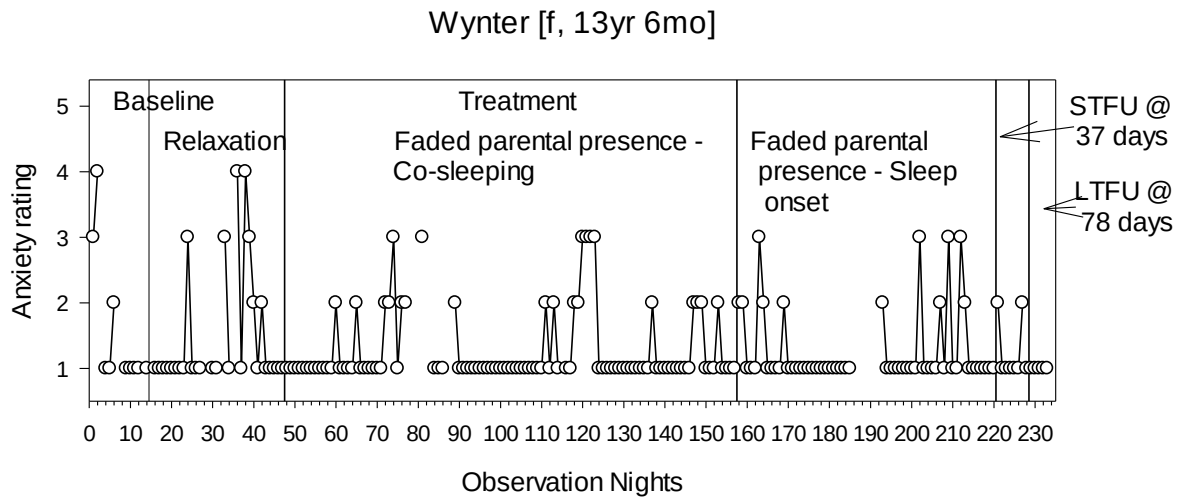


Anxiety Ratings

Anxiety ratings were self-reported by Wynter. During the baseline phase, ratings ranged between 1-4 (*M* =1.6). Data was not provided by the family on 4 of the 14 nights of the baseline phase. During Phase One ratings ranged between 1-4 (*M* =1.5). Treatment Phase Two ranged from 1-3 (*M* =1.5), with no further ratings above 3 recorded throughout further treatment and follow up phases. In treatment Phase Three ratings ranged from 1-3 with a slight reduction in mean score (*M* =1.2). Higher ratings correlated with higher SOL and NW scores. Higher anxiety ratings in the latter part of Phase Three and STFU phases are recorded in sleep diaries as feelings of excitement, rather than anxiety, for example – 'feeling excited about my favourite music' or 'excited about lizard species'. Anxiety ratings reduced slightly at STFU, with a range of 1-2 (*M* =1.2), and LTFU indicated a modest reduction in Anxiety, with all scores at 1. Overall, anxiety ratings reduced in intensity (range 1-4 at Baseline, and 1 at LTFU) and stabilised following treatment (*M* =1.6 at Baseline, *M* = 1.2 at LTFU). Statistically however, a minimal treatment effect was evident overall. Figure 3 presents anxiety ratings across all treatment phases for Wynter.

Figure 3

Anxiety Ratings Self-reported by Wynter across Treatment Phases



Psychometric Results

Child Sleep Habits Questionnaire (CSHQ) Scores

Wynter's total sleep difficulties score reduced from 56 to 49 from baseline to post-treatment. This indicates a meaningful reduction in real-world sleep difficulties overall, although does not represent a clinically significant change, as scores remained above the clinical cut-off of 41 for sleep difficulties . CSHQ item scores are illustrated in Table 2. Sleep variable scores indicate Wynter's perceived difficulties with bedtime resistance and sleep anxiety clearly reduced following treatment, while sleep onset remained the same, and night time awakening slightly increased.

Table 2

Pre and Post Treatment Comparison of CSHQ Subscale Scores

Dependent Variable	Pre-Treatment	Post Treatment
Bedtime Resistance	11	6
Sleep Onset	1	1
Night Awakening	4	5
Sleep Anxiety	11	6

Child Behaviour Checklist - ASEBA Youth Self Report (Y S-R)

CBCL-YS-R outcomes are presented in Table 3. Pre to post-treatment comparison of Wynter's scores on this measure demonstrate a reduction in both internalising and externalising problems. Post-treatment reductions were noted on the Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, and Aggressive Behaviour subscales, with scores remaining the same on the Rule-breaking and Other Problems subscales. Overall scores indicate a clinically significant reduction on Internalising Problems, lowering from the Clinical Range to the Normal Range, and scores on the Anxious/Depressed subscale falling from the Borderline Range to the Normal Range, with the Total Score also reducing from the Clinical Range to the Normal Range.

Table 3

Pre and Post-treatment Syndrome Scale and Total Scores on the CBCL YS-R

Subscale	Pre-treatment Score	Post-treatment Score
Anxious/Depressed	10	**6**
Withdrawn/Depressed	6	**3**
Somatic Complaints	7	**5**
Social Problems	8	**3**
Thought Problems	9	**6**
Attention Problems	9	**3**
Rule-Breaking Behaviour.	2	2
Aggressive Behaviour.	13	**6**
Other Problems	6	6
Total Internalising Behaviours	63	**55**
Total Externalising Behaviours	58	**49**
Total Scores	**70**	**40**

*Numerals in bold highlight a post-treatment reduction. T Score 60-63= Borderline Clinical, above 63 = Clinical Range

Anxiety subscale – 7-10 Borderline Range, above 10 Clinical Range

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

Wynter completed the self-report version of the PedsQL, and her mother completed the parent-report PedsQL at baseline and post-treatment. PedsQL data is presented in Table 4

below. Increased scores on PedsQL indicate improvement in quality of life. Wynter reported an increase in quality of life on all subscales with a Total Score increase from 64.1-76.1. Parent report data indicated her mother perceived an improved quality of life for Wynter in Emotional Functioning, Social Functioning and School Functioning following treatment. Parent report data scored Physical Functioning as unchanged, with a Total Score improvement from 59.7 pre-treatment to 71.7 post-treatment.

Table 4

Pre and Post-treatment PedsQL Scores

PedsQL (Self-report)	**Pre Treatment**	**Post Treatment**
Physical Functioning	84.4	90.6
Emotional Functioning	40	60
Social Functioning	60	65
School Functioning	60	80
Total Score	64.1	76.1
PedsQL (Parent Report)	**Pre Treatment**	**Post Treatment**
Physical Functioning	87.5	87.5
Emotional Functioning	45	65
Social Functioning	55	60
School Functioning	35	65
Total Score	59.7	71.7

Parent Well-being – Depression-Anxiety-Stress Scale (DASS-21)

Each scale on the DASS-21 (Lovibond, S.H., Lovibond P.F. 1995) has a minimum score of 0 in the Normal Range, to a maximum score of 42, in the Extremely Severe Range. For Wynter's mother, all scores resided in the Normal Range at pre-treatment and post-treatment. Scores on Depression rose from 1 at pre-treatment to 4 at post-treatment. Anxiety

scores reduced following treatment, from 6 pre-treatment to 5 post-treatment. The Stress scale score also reduced after treatment from 12 pre-treatment to 8 post-treatment.

Pittsburgh Sleep Quality Index (PSQI)

Questions on the PSQI relate to typical sleep habits over the past month, with a global score denoting overall sleep quality. A lower score indicates better sleep quality. Overall, parent's PSQI scores rose following treatment, indicating Jenny perceived her own sleep quality as having slightly worsened compared to the pre-treatment score.

Treatment Acceptability Rating Form (TARF-R)

The TARF-R (Reimers et.al., 1992) rates parents' perceptions of overall treatment acceptability post intervention. Parent ratings indicate Wynter's mother experienced the intervention process as acceptable overall, clear to understand, and she was very confident the treatment was effective. Wynter's mother indicated that although treatment required a lot of time to carry out, there were no disadvantages to following the treatment, and treatment was likely to result in permanent improvements in her child's behaviour. Parent's Total Acceptability rating was 102/119.

Post Treatment Feedback

Post-treatment feedback was obtained from the family through a semi-structured interview with the researcher.

Wynter. Wynter reported she liked the intervention process overall, and that rewards had really helped her to follow the treatment plan. She noted that filling out sleep diaries and practising relaxation strategies was challenging at times. Wynter stated visualisation was good at first, but sometimes difficult to do when she felt really stressed. Wynter reported that deep breathing had made the biggest difference for her. She stated positive changes as a

result of the treatment included getting more sleep, feeling “less grumpy”, and having more friends.

Wynter’s mother. Wynter’s mother described the intervention process as very effective, and stated she felt well supported along the way. She reported the sleep team’s way of relating to and including Wynter, so she was engaged and motivated was particularly valuable, and not always easy to achieve. Wynter’s mother noted the workload of diary entries was challenging at times, as was having to wait to implement new treatment steps while procedures were planned in conjunction with wider sleep team and supervisor consultation. She felt the realisation that she was uncomfortable leaving Wynter when she was distressed was a significant shift for her, and the knowledge that it was not ‘bad parenting’ to say “you can do this on your own, you don’t need me” was a revelation, and a real breakthrough for her.

She also felt recognising the importance of Wynter being self-motivated, and playing a role in planning and pacing of treatment steps made a significant contribution to effective treatment outcomes. She said learning how to talk to Wynter in a way which promoted success such as reminding her of progress already made, and the reward she would gain from reaching her goals were things that also made a difference in being able to adhere to the treatment process. Wynter’s mother felt the effectiveness of the relaxation strategies themselves was minimal, and the key to their effectiveness was that they were independent strategies that ‘weren’t Mum’. She stated that for Wynter, the realisation she had other strategies available to her and she didn’t need her mother was a key point of value regarding the relaxation strategies she learnt.

Wynter and her mother both reported feeling less anxious, and happier following the intervention. Significant positive changes for Wynter’s mother included having more time to herself in the evening, and worrying significantly less about Wynter’s sleep. She had

previously believed the sleep difficulties were impossible to change, as they had been present in varying degrees for almost all of Wynter's life. Overall, Wynter and her mother reported that treatment was a significant commitment, but positive outcomes meant treatment was well worth the time and effort, and the family felt well supported by clinicians throughout the treatment process.

Reliability

Inter-observer Agreement and Treatment Fidelity

IOA was not possible to calculate, as instrumental observation measures (Videosomnography or Actigraphy) were not consented for use by the family. Treatment fidelity was calculated for 25% of nights for Phase Two and Three. Implementation of relaxation strategies in Phase One were not recorded, therefore direct measurement of treatment fidelity was not calculated for this phase. Treatment Fidelity was 89.2% in Phase Two, and 93.7% in Phase Three.

Chapter 4: Study Two Method

Research Design

Study Two used a single-case multiple baseline A-B design. This design incorporated baseline (A), treatment (B), short-term follow-up, and long-term follow-up phases to evaluate intervention effects, with each single participant acting as their own control (Kazdin, 2019). Single-case designs allow causal inferences to be drawn through repeated collection of data on primary dependent variables within, and across phases (Blampied, 2013; Kazdin, 2019). This allows treatment effects to be detected through changes in outcome variables when making a comparison between stable baseline data, and data obtained in the active treatment phase (Kazdin, 2019; Shadish & Sullivan, 2011). A multiple baseline enables behaviour change to be measured across several participants with behaviours and treatments which are similar, whilst ensuring the complexities inherent within a heterogeneous (e.g., autistic) population are incorporated into the entire research process of assessment, treatment, and data analysis (Blampied, 2013; Shadish & Sullivan, 2011; Kazdin, 2019). Additionally, multiple baseline single case design enables real-world clinical change to be identified within individuals, e.g., alleviation of distress, improvement in daily functioning, and attainment of goals related to quality of life (Blampied, 2013).

Additional Outcome Measures

Psychometric measures described and utilised previously in Study One were implemented with participants in Study Two. An additional anxiety measure (MASC-2) and an additional psychometric sleep outcome measure (SSR) were included in this study, both described below.

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

The MASC-2 includes a parent, and optional child self-report measure of anxiety for youth aged 8-19 years. Items are rated on a four-point Likert scale, with 39 items in total. The Likert scale is allocated from 0-3, and described as 0 = never true about my child, to 3 = often true about my child on the parent questionnaire, and 0 = never, to 3 = often, on the child-report measure. There are 6 item domains which elicit data on Physical/Somatic symptoms, Social Anxiety (humiliation/rejection/social performance fears), Generalised Anxiety, Obsessions and Compulsions, Harm Avoidance (perfectionism/anxious coping), and Separation Anxiety (fear of being away from parents/home/family). Total Scores and individual subscale scores can be calculated (March et.al., 1997; Muris et.al, 2001). A higher subscale score, or Total Score indicates higher probability of Problematic Anxiety. The Total Score is used to gauge an overall Anxiety Probability Score with 0 representing Low Probability of Problematic Anxiety, 1 = Borderline Probability, 2 = High Probability, and 3 = Very High Probability. The MASC-2 reflects acceptable reliability (a = 0.92) , and strong test-re-test reliability (.80-.94) (Fraccaro et.al., 2015). The MASC-2 is accepted as appropriate for use with the verbal autistic population, with acceptable internal consistency for use with autistic children and youth (Lecavalier et.al., 2014; Van Schalkwyk et.al., 2017). MASC-2 was administered to participants in Study Two before and after intervention, to assess pre-treatment anxiety levels and to evaluate collateral effects of sleep treatment on participants' overall experience of anxiety.

Sleep Self Report (SSR: Owens et.al., 2000)

The SSR was developed to obtain self-report data relating to the frequency of sleep-problem behaviours occurring in children over the previous week. The SSR is a 26-item questionnaire designed for children aged 7-12, and closely corresponds to CSHQ subscales. The SSR contains six subscales: Bedtime Resistance, Sleep Onset Delay, Sleep Duration,

Sleep Anxiety, Night Wakings, and Daytime Sleepiness. Respondents answer "Yes" or 'No" questions related to their sleep behaviour, and also rate items on a three-point scale, with 0-1 rated as "rarely occurs", 2-4 "sometimes occurs", and 5-7 "usually occurs". In addition to the six subscale scores, the sum of all item scores provides a Total SSR Score. Scores have a potential range from 23-69, with higher scores indicating more severe sleep difficulties. The SSR has demonstrated internal validity and test-re-test reliability (0.76-0.88; Owens et.al., 2000), and has been used with non-autistic and autistic children in research settings (Richdale & Baglin, 2015).

Study Phases

The study involved five distinct study phases: assessment, baseline, treatment, maintenance, and follow-up.

Assessment

Assessment of the child and family was completed through clinical interview, pre-treatment psychometric measures such as the SATT, CSHQ, CBCL, PSQI, and functional behavioural assessment (FBA), as described in Chapter 2.

Baseline

The baseline phase followed the initial assessment and recruitment period. Each participant was assigned randomly to a one, two, or three-week baseline phase, using an online random number generator (Random.org). During the baseline phase, participants and families are asked to maintain their typical sleep routines and behaviours, and engage in recording data in sleep diaries, and videosomnography or actigraph if consented. The treatment phase is introduced immediately once stable baseline data is obtained, to ensure changes can be attributed to the introduction of treatment.

Treatment

The treatment phase was commenced with families as soon as possible after completion of the baseline phase. Treatment goals were established in response to assessment, FBA data, and discussion with the family. The intervention process was outlined in detail prior to commencing the treatment phase, to ensure participants and their family clearly understood the treatment steps, and to confirm treatment was a feasible fit for the family. Regular contact with families was maintained throughout treatment (daily phone, text, or email contact) to support the family with treatment adherence, and to respond quickly to any problems. Participant progress was discussed weekly with the sleep team, and treatment steps adjusted if required. The treatment phase continued until sleep difficulties were reduced or eliminated, treatment goals were met, and the family expressed satisfaction with treatment progress.

Maintenance

The maintenance phase began immediately following treatment. Aside from post-treatment interviews and post-treatment assessments, no contact was made with the family to allow participants to consolidate independent sleep skills into their daily routines without support from clinicians.

Follow up

Short-term follow up (STFU) was completed 4-6 weeks after treatment, and long-term-follow-up (LTFU) was carried out 10-12 weeks after treatment. Follow-up included the daily completion of sleep diaries for one week, and video or actigraph data collection if consented by the family.

Data Analysis

Sleep outcome data was obtained through sleep diaries recorded throughout baseline, treatment, short- and long-term follow-up phases. Dependent variable data for each participant was graphed for visual analysis. Dependent variables graphed for participants in Study Two included Sleep Onset Latency (SOL), Duration and Frequency of Night awakenings (DNW, FNW). Participants were encouraged to record daily ratings of anxiety on a scale of one to five, with five being the highest rating of anxiety, and one being the lowest. Only one participant (Gemma) provided consistent recordings of anxiety ratings in Study Two, and this participant's anxiety rating data is presented in graph form.

Systematic visual inspection of graphed data was the primary method of data analysis in Study Two. Visual analysis is commonly indicated as the optimal method of analysis in single-case research, as it enables real-time analysis of behaviour change, and allows researchers to identify changes that most likely occur as a result of treatment effect (Blampied, 2013; Cohen et.al., 2014). Treatment effect was evaluated according to data trend, level, variability, latency, and consistency of change (Cohen et.al., 2014). Additionally, the percentage of data exceeding the baseline median (PEM: Ma, 2006; Parker et.al., 2011) was calculated as an effect size, to determine treatment effect on dependent sleep outcome variables (SOL, DNW, FNW), and anxiety ratings for one participant (Gemma). Where treatment effect is indicated by a decrease in behaviour, the percentage of data points below the baseline median (PBM) were calculated, and interpreted as follows: a PBM score of below 50%, indicated ineffective treatment, 50-69% mild treatment effect, 70-89% moderate treatment effect, and 90% or above represented a high treatment effect (Ma, 2006). Data from pre and post treatment psychometric measures were analysed to assess for treatment affects within and across participants, and qualitative data from post treatment

interviews were also examined to obtain further evidence on participants' overall treatment outcomes, attainment of goals and perceived acceptability of treatment.

Participants

Gemma

Gemma was a 16-year-old female, diagnosed with Autism. Gemma had strong verbal ability. Gemma stated she experienced high levels of anxiety, but had not sought a formal diagnosis of an anxiety disorder. She lived at home with family. Gemma typically shared a bedroom with another sibling, but had recently begun sharing a double bed with her mother, as she found her own bed uncomfortable.

Presenting Concerns

Gemma was referred to the sleep study following assessment by a paediatric specialist who suggested her sleep difficulties may be related to 'obsessive thoughts'. During assessment, Gemma reported difficulty falling asleep every night of the week, and she experienced intrusive thoughts which interfered with falling, and staying asleep. This was corroborated by her mother who noted that it took her 2-3 hours to fall asleep most nights. Gemma also noted long periods of wakefulness during the night, 2-3 times a night, up to 6 nights per week. Gemma reported problems with sleep were impacting her ability to function during the day, with low energy and poor concentration affecting her on most days of the week.

She noted barriers which impacted her ability to get to bed at a specific time included regular social and family events in the evening, and environmental factors such as streetlights, and noise from the household interfered with her ability to sleep. Gemma's mother stated Gemma had experienced sleep difficulties since she was about 2 years of age, primarily with getting to sleep at the beginning of the night, and sleep difficulties had become

progressively worse over the past two years. The family had tried various nutritional supplements to aid Gemma's sleep with minimal effect, and she did not take any medications or supplements prior to or during the sleep study treatment.

Functional Behaviour Assessment

Information from the FBA indicated various factors were playing a role in the onset and maintenance of Gemma's sleep difficulties. This included inconsistent bedtime and waketimes contributing to a lack of physiological sleep pressure and dysregulation of her circadian rhythm. This interfered with Gemma's ability to fall asleep and remain asleep all night. Insufficient sleep pressure at bedtime appeared to increase vulnerability towards the occurrence of sleep interfering cognitions (i.e. intrusive repetitive thoughts and worries) which exacerbated high levels of arousal, incompatible with the low level of arousal required for sleep. A number of antecedent variables were also contributing to sleep disturbance at sleep onset, and throughout the night. A busy family environment (e.g. noise from other family members, and ambient light) prior to sleep, and regular social/family events were interfering with Gemma's ability to establish a sleep conducive environment, and consistent bedtime routine. The subsequent inconsistent sleep and waketimes resulted in insufficient sleep pressure and reduced motivation for Gemma to achieve sleep at a regular time each night. In addition to insufficient sleep pressure, FBA indicated social attention was a primary function of Gemma's sleep incompatible behaviours.

Sleep Goals

Gemma's sleep goals were 1) to fall asleep in less than one hour 2) to achieve a good quantity and quality of sleep each night, so she would have the energy to learn well at school and join in with her friends during social activities. Gemma's mother's goal was for Gemma to fall asleep in under one hour, and to stay asleep throughout the night. She stated her ideal

sleep/wake schedule for Gemma would be 9pm - 7am. Gemma indicated her preference was to complete sleep diaries and carry out intervention steps independently, with her mother's support only if required.

Intervention

Sleep/wake rescheduling. Following 2 weeks of baseline measurement, Phase One of treatment commenced with establishing a regular sleep/wake schedule of 10pm bedtime, and 6.30am morning waketime. Gemma was provided with psychoeducation from the intern psychologist to explain the treatment plan. This included psychoeducation about the circadian rhythm and the importance of a regular bed and wake time to stabilise the sleep-wake cycle. Sleep hygiene practices such as ensuring the sleep environment was cool and dark, and being sufficiently hydrated were also discussed. Sleep interfering cognitions were monitored during this phase with Gemma recording her thoughts in sleep diaries, in preparation for the introduction of relaxation strategies if required.

Procedural Modifications. On day 55, Gemma's sleep and wake time were adjusted to 10.30pm-7am, to better fit in with family routines and seek further improvement in SOL and reduction in NW's .

The introduction of relaxation strategies was planned as the next phase of treatment, but Gemma and her family felt satisfied with treatment results and the family was moved to maintenance phase before relaxation strategies were implemented. Gemma indicated that the act of writing down her thoughts for the purpose of data recording had a therapeutic effect for her, where thoughts were not so intrusive at sleep onset. Intervention lasted for 88 days.

Oscar

Oscar was a 13-year-old boy, diagnosed with Autism one year prior to engaging with the sleep study. Oscar had used melatonin for the past year, but had recently stopped taking

it as he felt it was not effective, and he did not take medication or supplements during sleep treatment.

Presenting Concerns

Oscar's mother stated Oscar's sleep disturbance had been lifelong, with ongoing difficulties getting to sleep. She was unsure if he woke in the night as he did not disturb his parents, but she stated they were aware he would lie awake and 'worry'. Oscar and his mother reported he would take 30-120 minutes to fall asleep currently, with sleep onset being up to 3 hours at its worst. Oscar said he would usually lie in bed trying to fall asleep, but would not leave his room, or seek his parents' attention. Oscar stated he found it difficult to stop thinking and 'switch off'. Oscar reported he would wake up at 12 am on 5 out of 7 nights, and would take 30 minutes or more to fall back to sleep. On weekends, Oscar would often catch up on sleep. In the past he used to sleep until 12 or 1pm but recently, his parents had been waking him at 10am. Oscar said he often felt very tired, and had difficulty getting up in the morning. His parents noticed that Oscar would find it hard to concentrate, and could be grumpy when he had not had sufficient sleep.

During infancy, Oscar was only able to sleep for 45 minutes at a time. Instead of long sleeps, Oscar would 'nap' over the 24-hour day-night cycle, until he was 2 years of age, when he began to sleep more deeply and for longer periods through the night. Oscar's parents had tried a number of strategies to improve his sleep over the years, including using blackout curtains and installing a ceiling fan, and medications such as quetiapine, and melatonin. Oscar and his parents felt these strategies had minimal impact on his sleep. Oscar stated being active during the day, and thinking about favourite things had sometimes proven helpful in getting to sleep, but this had not consistently improved his sleep.

Functional Behaviour Assessment

Oscar's sleep disturbance included SOD and NW's. Oscar had an inconsistent sleep wake schedule, where his weekday sleep/wake routine contrasted significantly to his weekend sleep/wake routine. Late bedtimes and consistent early rising for school during the week, contrasted with late bedtimes and delayed waketimes (sleeping in) on the weekends. An inconsistent sleep/wake schedule resulted in a dysregulated circadian rhythm which decreased sleep pressure at appropriate times for sleep. Sleep interfering thoughts at bedtime also appeared to contribute to high states of arousal, interfering with achievement of lower states of arousal required for sleep. Decreased sleep pressure, and cognitive hyperarousal affected Oscar's motivation to go to bed and fall asleep at appropriate times, and he tended to engage in other sleep interfering behaviours such as reading in bed when he was not able to fall asleep. In addition to insufficient sleep pressure, FBA information indicated the function of Oscar's sleep interfering behaviours was access to preferred activities.

Sleep Goals

Oscar's sleep goals were 1) to fall asleep within 20-30 minutes, 2) to manage his sleep independently without medication, and 3) to wake up feeling refreshed. His parent's goals were 1) for Oscar to fall asleep within 30 minutes, and 2) to consistently get a good night's sleep.

Intervention

Faded Bedtime/Sleep/wake Rescheduling. Following a 2-week baseline phase, a faded bedtime was implemented during Phase One of intervention. This consisted of establishing the average time that Oscar fell asleep, and setting his bedtime at this time. His bedtime was then gradually shifted earlier when his sleep onset fell within acceptable and stable limits (e.g., 15 mins). The addition of a consistent morning wake time was also

implemented to assist in regulating Oscar's sleep/wake cycle. This was done to facilitate sufficient sleep pressure in the evening. Based on assessment and baseline data, Oscar's recommended sleep time and wake times were set at 10pm, and 7.30 am at the start of treatment.

Stimulus control. Stimulus control can improve sleep by eliminating discriminative stimuli which tend to interfere with sleep (e.g. removing stimuli associated with wakefulness such as device use in bed) or providing discriminative stimuli which act as cues for sleep onset (e.g. stimuli consistently associated with sleep onset such as putting on pyjamas and a bedtime story). Individuals experiencing sleep disturbance tend to spend large periods of time awake in bed, feeling frustrated or anxious about not getting to sleep. Therefore, bed and the sleep environment can inadvertently become a cue for high arousal, rather than the low arousal state required for sleep (Richdale et.al., 2014). Oscar was advised to use his bed only for sleep, and avoid lying on his bed during the day, to reinforce his bed and sleep environment as a cue for sleep onset.

Procedural Modifications. Oscar's bedtime was adjusted to 9.45pm, with morning waketime 7.30am on day 61 of treatment, as sleep onset was stable, and his parents preferred the earlier time to ensure he gained sufficient overall sleep, and Oscar agreed to this. Oscar's treatment concluded on day 84, as he and his family were satisfied that their sleep goals had been achieved.

Fletcher

Fletcher was a 14-year-old boy who was diagnosed with Autism at 12 years of age. He lived at home with his parents, and siblings.

Presenting Concerns

Fletcher's mother contacted the sleep team in response to her concerns with Fletcher's sleep difficulties, which were impacting on his school attendance. His mother described Fletcher's sleep as a long-standing issue, and he was currently refusing to go to school if he did not get sufficient sleep. Fletcher's parents reported that Fletcher worried about things excessively, with a tendency to impose rigid routines and control to combat feelings of anxiety. His parents stated that Fletcher was 'not aware' of this anxiety himself, due to his autism.

Fletcher and his family reported his primary sleep difficulty related to SOD, and intermittent NW's. Fletcher typically got to bed at 12am, and took between 1-3 hours to fall asleep. At its worst, Fletcher had experienced nights where he did not get any sleep at all. His parents stated they often had trouble waking him up in the morning. Fletcher said he woke up about 4 times a night, and would take 20-30 minutes to fall back asleep. Sometimes he woke to use the toilet or get a drink, other times he was disturbed by slight noises, or light shining in the window. Fletcher's parents reported sleep disturbance had been a lifelong challenge for him, and sleep difficulties had worsened from the age of 10. Fletcher had recently tried melatonin which appeared to have little effect, and Fletcher felt it made him drowsy the next day, so chose not to take it.

Functional Behaviour Assessment

According to FBA information, several factors were maintaining Fletcher's sleep difficulties. This included extended pre-sleep routines which delayed his bedtime, and likely contributed to a state of hyperarousal. Fletcher's device use, long shower rituals, intermittent consumption of a full meal just before bed, and an entrained late sleep time where interfering with his ability to get to bed in a timely manner, and to fall asleep quickly. FBA also

identified environmental factors such as noises and lights were interfering with sleep onset, and disturbing night sleep. A significant factor contributing to SOD appeared to be lack of sleep pressure due to significant circadian dysregulation. It was hypothesised that inadequate sleep pressure due to circadian dysregulation, and access to preferred items and activities were maintaining Fletcher's sleep interfering behaviours.

Sleep Goals

Fletcher's sleep goal was to get 8.5 to 9 hours of sleep every night, and to wake up at the same time every morning. His parent's goals were for Fletcher to get to bed earlier, and fall asleep at a set time, to enable him to wake easily in the morning, and get to school. His mother stated she would like him to learn sleep conducive strategies he could use independently, to help him self-manage his sleep successfully.

Intervention

Phase One: Faded Bedtime/Sleep/wake Rescheduling. The first phase of treatment began following one week of baseline monitoring. Phase One involved establishing a regular bedtime that approximated Fletcher's average sleep onset time, and a regular wake time in the morning, to regulate his sleep/wake cycle. Fletcher's bedtime was set for 12am, with a morning waketime of 8.30am. Fletcher was advised to wake up at 8.30am in the morning regardless of the quantity or quality of sleep he managed in the night. Fletcher was provided with psychoeducation about the importance of a regulated circadian rhythm to enable sufficient sleep, and stimulation of growth hormones. Information explaining regular sleep and waketimes for resetting your circadian 'body clock' and establishing the regulation of sleep/wake hormones, was provided for Fletcher and his parents. Treatment Phase One lasted for 46 days.

Phase Two: Sleep Hygiene. In Phase Two of treatment, Fletcher was given the choice of fading his bedtime to an earlier time in 15-minute increments, (with some evidence of sleep consolidation during Phase One), or improving sleep hygiene practices. Fletcher chose to improve sleep hygiene. External/environmental factors interfering with Fletcher's sleep were thus addressed, with a 'good sleep hygiene' routine agreed and implemented. This included co-creation of a before-bed checklist that outlined steps in Fletcher's bedtime routine, including completing exercise routines before 5pm, completing device/social media use one hour before bed, and engaging in a quiet activity before bed such as reading, listening to audiobooks, or a warm bath. The checklist was also intended as a visual support to aid Fletcher in following his bedtime and morning waketime consistently, as he had found it difficult to do this during Phase One. Fletcher disclosed at this time, that he had very recently begun consuming caffeinated workout supplements to support his exercise routines. He was advised to ensure these were taken before 5pm, and this was added to his sleep hygiene checklist. Fletcher was encouraged to get up out of bed when he woke in the morning, and to avoid lying on his bed except when he was going to sleep for the night. His parents purchased a bean bag to support him with this change. Other environmental modifications designed to reduce sleep disturbance included the purchase of a larger bed for Fletcher, as there was some suggestion the smaller size of his current bed may be causing discomfort. Blackout curtains were also placed over windows to reduce light pollution. Phase Two was carried out for three weeks, from day 55, to day 76 of treatment. During the course of treatment, Fletcher became increasingly focussed on his interest in nutrition and exercise, and he reported that he found it increasingly challenging to prioritise sleep treatment protocols if he perceived them as interfering with his nutrition and exercise priorities.

Phase Three: Adjustment of morning waketime. On day 76 of treatment, Fletcher's morning waketime was adjusted from 8.30am to 8am. For the 4 days prior to

Phase Three, Fletcher had been having difficulty following through with his sleep hygiene routines.

Withdrawal from Study. Phase Three was carried out for 6 days. Following this, Fletcher and his family decided to withdraw from the sleep study after 82 days of treatment. Fletcher had commenced an alternative model of schooling and he felt that sleep treatment was no longer necessary as he was able to sleep in.

Chapter 5: Study 2 Results

This chapter presents sleep outcome data from sleep diaries and psychometric measures for all participants in Study Two. Pre-and post-treatment measures of participant sleep quality, quantity and sleep hygiene, anxiety, daytime behaviour, and quality of life are presented. Parent data on well-being and sleep quality is also included. Finally, post-treatment feedback and treatment acceptability data are reported.

Quality of Data

Missing or incomplete sleep diary data were noted for all three participants. Treatment length in days, details about missing data, and variations in psychometric measures administered across participants are outlined below.

Treatment Duration and Missing Sleep Diary Data

For two of the three participants, treatment continued until the families' sleep goals had been met. Sleep diaries were not completed by families on various days during treatment, for unspecified reasons. For Gemma, treatment lasted for 88 days, with sleep diary data missing on days' 4, 8, 9, and 11 (all during baseline phase). For Oscar, treatment duration was 84 days, with sleep diary data missing on days 1, 2,11,52,53, and 56. Fletcher's treatment continued for 83 days before withdrawal from the study. Sleep diary data were missing on days 26-28 due to Fletcher sleeping elsewhere that night. Sleep data is also missing for days 34,36,37,38, 51,65,67,68,71, and 73-76 . Fletcher reported he was not following the treatment plan on days 73-76 and the family withdrew from the study on day 83, before the final intervention phase was completed. Self-reported daily ratings of anxiety were only consistently recorded by one participant, Gemma.

Psychometric Measures

There was some variation in psychometric measures completed by participants pre- and post-treatment, with different lead clinicians overseeing casework. Gemma was not administered the CSHQ. The ASHS was not administered with Fletcher. Post-treatment data on the MASC-2 measure was not obtained from Fletcher due to the family withdrawing from treatment. Post-treatment data on the MASC-2 measure was not obtained from Oscar's family for unspecified reasons. All other measures were applied consistently across participants.

Actigraph and Videosomnography Data

Technological issues arose with actigraphy data, therefore only three nights (3%) of usable data were available for Oscar, and two nights (2%) for Gemma. This limited the possibility of calculating IOA. IOA was calculated via videosomnography data for Fletcher on 23 % of nights across baseline and treatment phases.

Summary of Participants Treatment Phases

Families were randomly assigned to a one, two, or three week baseline phase, using the True Random Number Generator (Random.Org). Gemma and Oscar were assigned a two-week baseline phase, and Fletcher was assigned a one-week baseline phase.

Fletcher - Treatment Phase One

Faded Bedtime. Modification of the sleep/wake cycle, establishing a regular sleep and waketime (12am – 8.30am). Phase One continued from day 8-54.

Treatment Phase Two

Sleep Hygiene. Changes to sleep-hygiene routine implemented, including environmental modifications and stimulus control. Phase Two continued from day 55-76.

Treatment Phase Three

Adjustment of morning waketime. Bedtime 12am, morning waketime shifted to 8am on day 77. Family withdrew from treatment on day 83.

Gemma - Treatment Phase One

Faded Bedtime. Modification of sleep/wake cycle, establishing regular sleep and morning waketime (10pm-6.30am).

Treatment Modification. Bedtime was adjusted on day 55 of treatment (10.30pm-7am). Gemma and family were satisfied with goal attainment, and moved to maintenance phase after 88 days of treatment. (An additional phase of relaxation strategies was not implemented).

Short (STFU) and Long Term Follow Up (LTFU)

One week of short-term follow up (STFU) data was collected five weeks post-treatment. One week of long-term follow up (LTFU) data was obtained ten weeks after treatment was completed.

Oscar - Treatment Phase One

Faded Bedtime. Modification of the sleep/wake cycle, establishing a regular sleep and morning waketime (10pm-7.30am).

Treatment Modification. Sleep/waketimes were adjusted to 9.45pm bedtime, and morning waketime 7.30am, on day 61 of treatment. Treatment was completed on day 84.

Short term Follow-up (STFU)

One week of STFU data was collected six weeks after treatment was completed. No LTFU data was collected, as the family were not contactable at the time.

Sleep Outcome Data

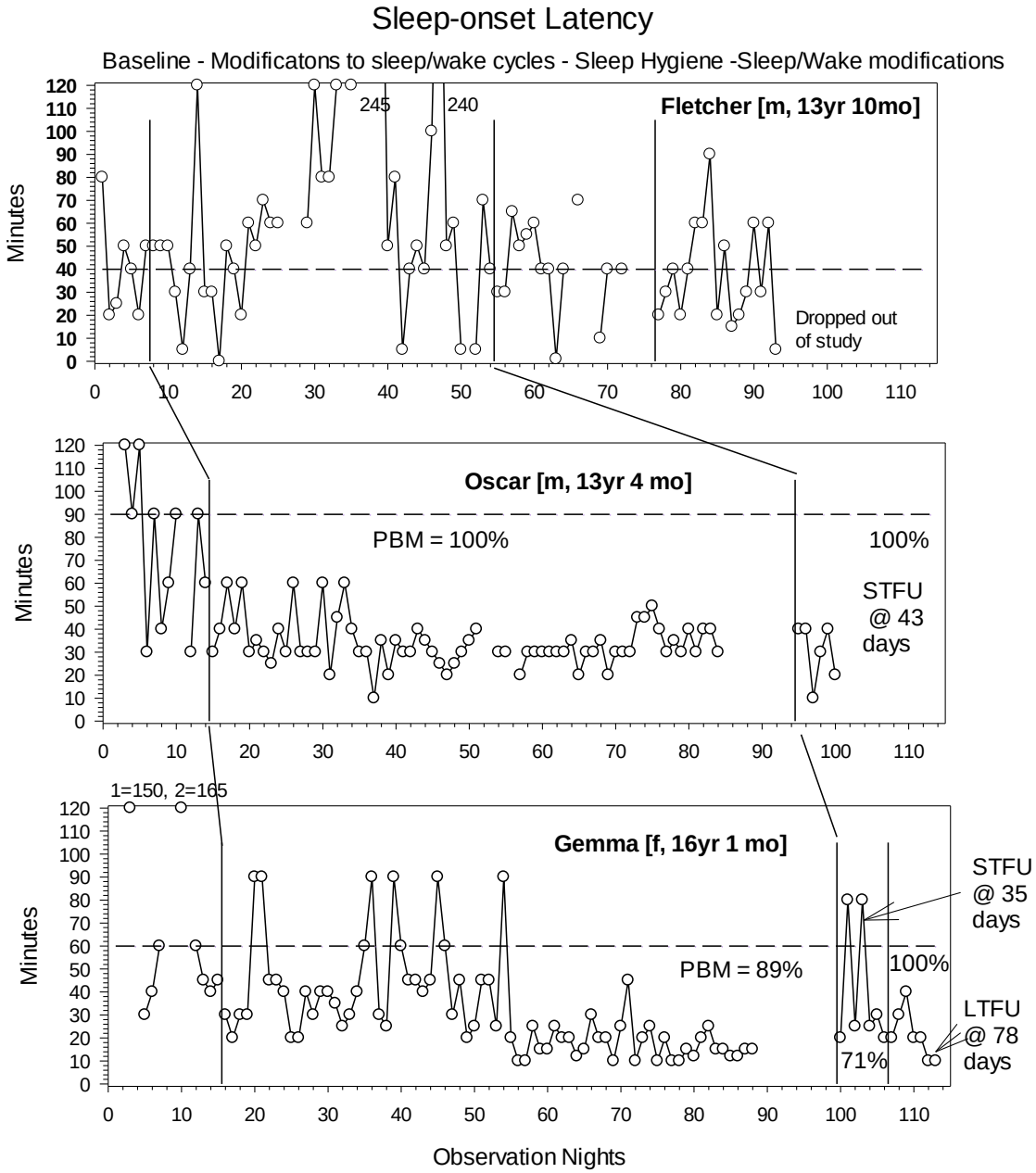
Sleep outcome data obtained through sleep diaries is presented in graph form, and written details report on all dependent variables including SOL, FNW, and DNW for all participants across treatment phases. Daily anxiety ratings from sleep diaries from one participant (Gemma) are presented as a graph, and described in written detail below.

Sleep Onset Latency (SOL)

Figure 4 presents duration of SOL in minutes, for each of the three participants across study phases. All participants demonstrated highly variable SOL during the baseline phase. For Oscar and Gemma, a reduction in SOL is seen when treatment begins, and a treatment effect is evident overall throughout treatment and follow up phases. Fletcher's data indicates a pattern of continued variability throughout treatment, with minimal evidence of a reduction in SOL overall.

Figure 4

Duration of Sleep Onset Latency in Minutes across Treatment Phases for Fletcher, Oscar, and Gemma



Notes: 1. Phase labels are shown above the top border of the figure. Only Fletcher implemented Sleep Hygiene based modifications. .

2. Numbers just below the top border of Fletcher's figure indicate the out-of-scale durations of SOL (min). Abbreviations are interpreted as m = male, f=female, yr.=years, mo.=months, PBM=Percentage Below the Mean. STFU=Short Term Follow-Up, LTFU=Long Term Follow-Up.

Fletcher

Sleep onset is highly variable and above the clinical cut off (15 mins) for Fletcher at baseline (range of 20-80 min; *M* = 41). PBM scores were not applicable to Fletcher's outcome data due to the high frequency of floor scores of 0. Although there is a rise from floor scores for Fletcher at treatment Phase One (sleep/wake cycle modification), data indicates no evidence of treatment effect when compared to baseline. SOL ranges from 0-245 minutes *(M* = 52.6) in Phase One. Phase Two (sleep hygiene), saw a minimal reduction in SOL, ranging from 1-70 minutes (*M* = 40.7). SOL variability remained throughout treatment Phase Three (sleep/wake modification), with a range of 5-90 minutes *(M* =38). Overall, SOL remained variable, problematic and above the clinical cut off for Fletcher, with minimal evidence of significant or sustained treatment effect prior to his withdrawal from treatment.

Oscar

SOL was highly variable at baseline, and rested above the clinical cut off (15mins), ranging from 30-120 minutes (*M* =74.5). SOL reduced and stabilised on commencement of treatment Phase One (sleep/wake cycle modification), ranging from 30 to 60 minutes during the first 33 days of treatment, followed by a further reduction in SOL, ranging from 10-50 minutes (*M* =30) for the remainder of treatment. Statistically, data indicated treatment was highly effective at 100% PBM throughout the treatment phase. STFU indicated continuation of treatment gains, with SOL ranging from 10-40 minutes *(M* =30), and 100% PBM. Overall, data indicated treatment was effective, with a clinically significant reduction in SOL, with Mean and Range scores falling below clinical cut-off following treatment.

Gemma

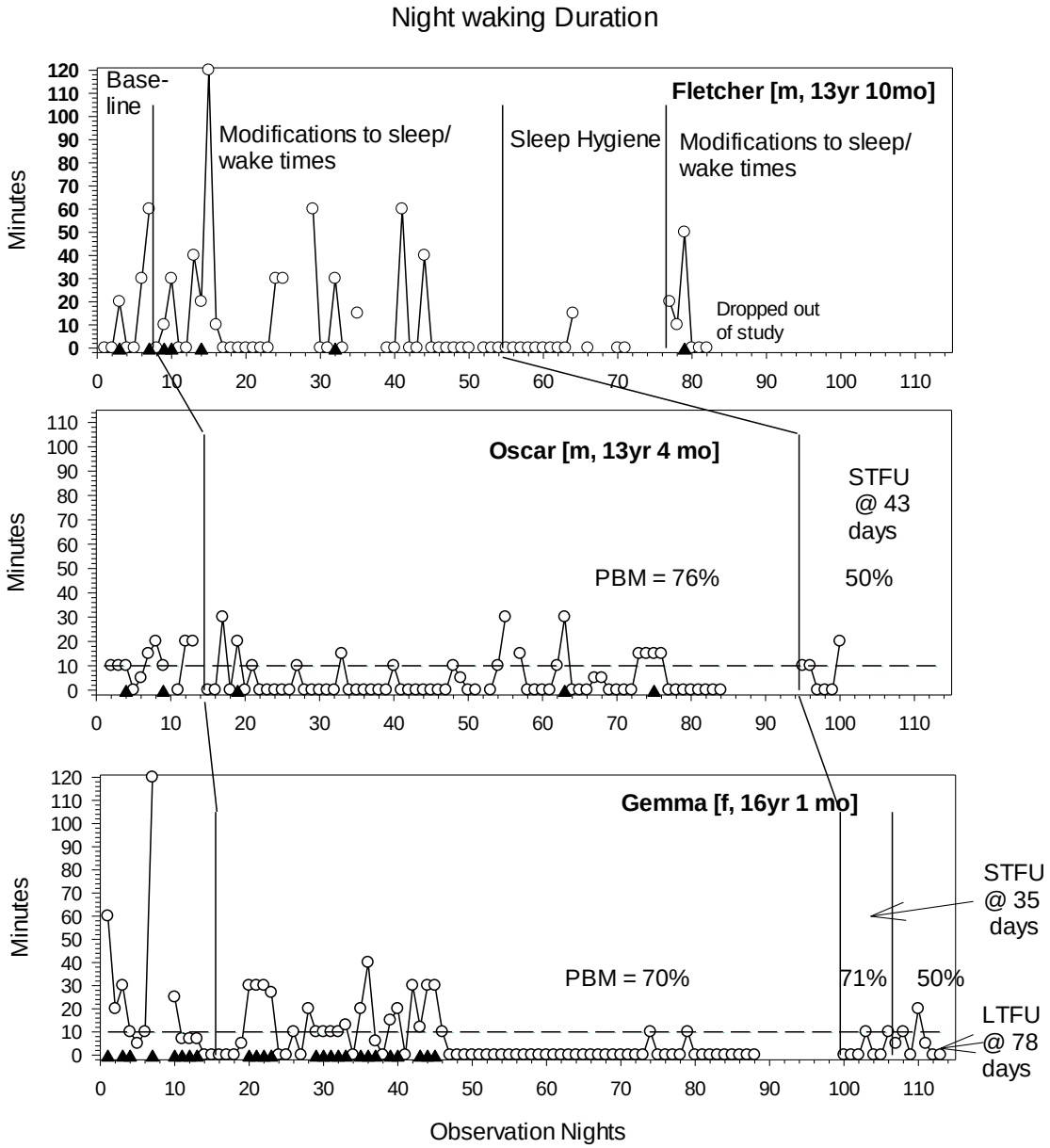
SOL was problematic for Gemma at baseline, ranging between 30-165 minutes *(M* = 80). Improvement is seen as Phase One (sleep/wake cycle modification) commenced, but peaks of extended SOL continue during the first 54 days of treatment with SOL ranging from 20-90 minutes. A significant and stable drop in SOL occurs from day 55, aligning with the additional modification to the sleep/wake schedule. Duration of SOL ranges from 10-30 minutes for the rest of the treatment phase, with one outlier score of 45 minutes on day 71 of treatment. A moderate treatment effect was indicated through this phase, at 89% PBM. A slight reduction in treatment effect is seen at STFU, with SOL ranging from 20-80 minutes *(M=* 40), and 79% PBM. Treatment gains increase at LTFU, with SOL ranging between 10-40 minutes *(M*=21.4) and 100% PBM. Overall a significant reduction in SOL was evident following treatment compared to baseline, and treatment gains were maintained at follow up for Gemma.

Frequency and Duration of Night Wakings

Figure 5 presents the FNW and DNW in minutes for all three participants across treatment phases, and is reported in detail below. No follow up data is presented for Fletcher due to his withdrawal from the study. Evidence of a reduction in FNW and DNW was indicated to varying degrees for all participants as a result of treatment. Moderate treatment effects were indicated for Gemma and Oscar during treatment, with modest evidence of treatment gains being sustained at follow up.

Figure 5

FNW's and DNW's in Minutes Across Study Phases for Fletcher, Oscar, and Gemma



Notes: Filled triangles indicate nights when there was more than 1 wake. For Fletcher & Oscar the range was 2-3, and for Gemma 2 – 4 wakes. Balance of non-zero wakes = 1 wake. Abbreviations are interpreted as m = male, f = female, mo =months, yr. =years. PBM = Percentage Below the Mean. Treatment Phases are labelled within the image (top of Fletcher's graph) , and Phase lines continued downward through each data graph.

Frequency of Night Wakings (FNW)

Fletcher. FNW ranged from 0-2 at baseline (*M* = 0.7) for Fletcher. FNW's reduced midway through Treatment Phase One, and across treatment Phase Two, with a period of 0 NW's occurring for 3 consecutive weeks, and only one NW occurring in Phase Two. Three consecutive nights of waking occurred immediately prior to the family withdrawing from treatment early in treatment Phase Three. This coincided with a period of low treatment fidelity reported in sleep diaries. Overall a modest reduction in night wakings was indicated for Fletcher. FNW's ranged from 0-3 in Phase One (*M* = 0.5), and 0-1 in Phase Two (*M* = 0.07).

Oscar. Baseline data demonstrates variability in NW's for Oscar, ranging from 0-3 wakings (*M* =1.4). A stable reduction in NW is evident following the first week of treatment Phase One. FNW's remains low throughout treatment compared to baseline, ranging from 1-3, with average FNW's falling below the baseline average (*M* = 0.32). Treatment gains reduced slightly at STFU, although remained below the baseline average (0.57). Overall, data indicated treatment was moderately effective in reducing FNW's for Oscar.

Gemma. For Gemma, night wakings were problematic at baseline, and ranged from 0-3 (*M* =1.4). FNW's remain at baseline levels until midway through treatment Phase One. A clear and consistent reduction in NW's emerges from day 47, with only two further NW's occurring throughout the rest of treatment. Treatment gain is largely maintained at follow up, with two single NW's occurring at STFU (*M* = 0.2), and remaining low relative to baseline at LTFU (*M* = 0.7). Overall a clear treatment effect is indicated for Gemma who experienced the highest frequency of NW's across all participants at baseline. These were significantly reduced as a result of treatment, and remained below baseline average at follow-up.

Duration of Night Wakings (DNW)

Fletcher. Baseline data revealed DNW's ranged from 0-60 minutes *(M* = 24). At treatment Phase One, night waking duration continued to be variable and ranged from 0 to 120 minutes with a subtle reduction in average duration *(M* =12.6). The final week of treatment Phase One suggested some evidence of sleep consolidation, with 0 NW recorded. Some indication of treatment effect continues into treatment Phase Two, and remains largely stable with only one night waking recorded in this phase. DNW's range between 0-15 minutes *(M* =1) although there are gaps in data that are noted toward the end of this phase. During Phase Three there is an increase in the variability of DNW, ranging from 0-50 minutes. This phase lasts for 6 days before the family's withdrawal from treatment. PBM calculations were not applicable to Fletcher's DNW data due to a 0 floor scores effect.

Gemma. Baseline data exhibits the longest DNW's for Gemma, ranging from 0-120 minutes *(M* =23). Improvement is seen at commencement of treatment, but DNW remains variable until clear evidence of a treatment effect emerges 7 weeks into treatment. An abrupt and consistent drop in the frequency, and duration of NW's is seen from day 47, just prior to an adjustment of Gemma's sleep wake schedule on day 55. Although statistically, the overall treatment effect is moderate with a PBM of 70% across Phase One, a significant and stable reduction in DNW is evident from day 47, and this remains low and stable through to the end of treatment. Treatment gains are maintained at STFU with a PBM of 71%, (moderate treatment effect) and is partially maintained at LTFU with a PBM of 51% (mild treatment effect).

Oscar. Oscar's DNW's during baseline ranges from 0-20 minutes *(M* =10). DNW variability continues through the first week of treatment, followed by indications of sleep consolidation with the first stretch of 0 NW from day 22-26. DNW ranged from 0-30 minutes overall through treatment Phase One, with 76% PBM indicating a moderate

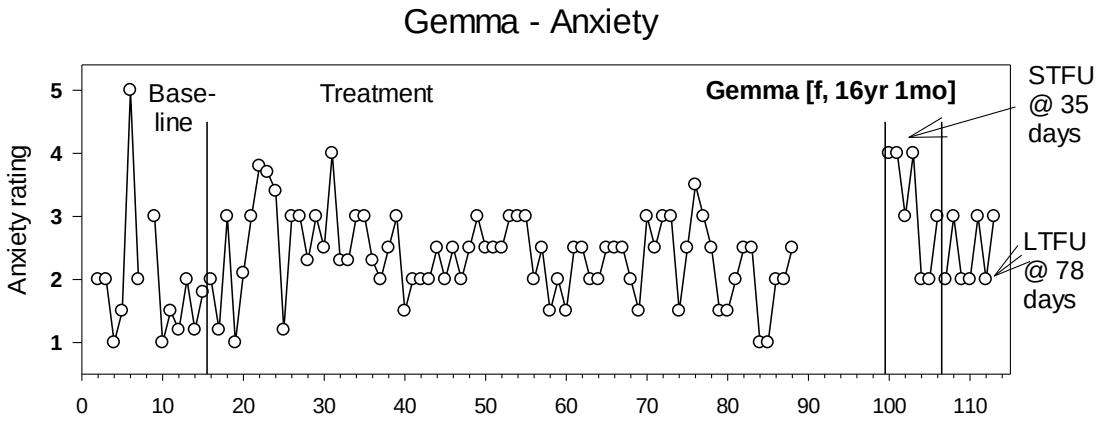
treatment effect. By the final week of treatment, DNW had reduced consistently to 0. Treatment effect was partially maintained at STFU, with DNW ranging from 0-20 minutes (*M* = 6), and 50% PBM. LTFU data were not provided by the family.

Anxiety Ratings.

Anxiety ratings were only consistently recorded by Gemma. Data on Gemma's daily ratings of anxiety on a 1-5 scale are presented visually in Figure 6, and summarised in detail below.

Figure 6

Daily Ratings of Anxiety for Gemma Across Treatment Phases



Gemma. Gemma's highest anxiety rating was recorded in the baseline phase, where ratings ranged from 1-5 *(M* = 2). Gemma included fraction ratings in her reporting of anxiety data (e.g. 1.8). Ratings of anxiety reported by Gemma remained variable throughout treatment Phase One, ranging from 1-3.8 *(M* = 2.3). STFU ratings ranged from 2-4 (M = 3.1), and LTFU ratings ranged from 2-3 *(M* = 2.4). No evidence of sustained treatment effect was indicated, with overall anxiety rising slightly in the treatment phase and follow up phases, relative to baseline data.

Psychometric Measures – Sleep Outcomes

Table 5 presents the psychometric sleep outcome data obtained from participants on the Children's Sleep Habits Questionnaire (CSHQ), Adolescent Sleep Hygiene Scale (ASHS), and Sleep Self-Report (SSR). Dashes in the table indicate data was not obtained.

Table 5

Pre and Post-Treatment Psychometric Total Scores for Participants' Sleep

Participant	CSHQ		ASHS		SSR	
	Pre	**Post**	**Pre**	**Post**	**Pre**	**Post**
Gemma	-	-	4.7	4.7	-	-
Oscar	56	60	4.83	5.49	41	31
Fletcher	46	-	-	-	41	-

Child Sleep Habits Questionnaire (CSHQ) (Owens et.al., 2000)

The CSHQ was completed by Oscar before and after treatment, and before treatment by Fletcher. Higher scores indicate a greater degree of sleep difficulty than lower scores. Scores were in the clinical range (≥41) for both Oscar and Fletcher at baseline. Oscar's CSHQ score increased from 56-60 post-treatment, and remained in the clinical range indicating Oscar reported his sleep habits as worsening after treatment according to the CSHQ scores. Post-treatment data was not obtained for Fletcher.

Adolescent Sleep Hygiene Scale (ASHS)

ASHS data were collected before and after treatment for Oscar and Gemma. Clinically significant change cannot be determined on this measure as no clinical cut-off score is indicated. Both Gemma and Oscar scored below 'good' sleep hygiene pre-treatment,

with scores of 4.7, and 4.8 respectively. No data was collected for Fletcher on the ASHS questionnaire. Post-treatment scores indicated Gemma had maintained similar sleep hygiene habits, with an identical score of 4.7 Oscar indicated improved sleep hygiene, with a post-treatment score of 5.49.

Sleep Self Report (SSR)

Oscar and Fletcher completed the Sleep Self Report at baseline, and Oscar provided post-treatment scores also. The SSR does not have a representative clinical cut off score, however reductions in scores indicate perceived improvement in sleep. Oscar's post-treatment score reduced from 41 to 31, reflecting perceived improvements in sleep.

Anxiety, Children's Daytime Behaviour and Well-being

Multidimensional Anxiety Scale for Children 2nd Edition (MASC-2)

Pre and post-treatment scores obtained from participant and parent-reports are presented in Table 6. Parents completed the MASC-2 parent-report and participants completed the MASC-2 self-report prior to treatment. Post-treatment MASC-2 data were completed by Gemma and family, but no post-treatment data were obtained from Oscar and Fletcher.

Table 6

Pre and Post-Treatment Sub-Scale and Total Scores for MASC-2 (Parent/Self-report)

Subscale	Gemma		Oscar		Fletcher	
(Parent/Self report)	**Pre**	**Post**	**Pre**	**Post**	**Pre**	**Post**
Separation Anxiety (S/P)	90/76	82/63	50/45	-	55/40	-
Generalised Anxiety (GAD)	90/80	78/68	60/60	-	69/42	-
Social Anxiety (S/A)	82/71	61/65	64/60	-	57/40	-
Obsessions & Compulsions (OC)	90/75	77/55	42/46	-	90/59	-
Physical Symptoms (P/S)	90/80	90/58	47/54	-	59/46	-
Harm Avoidance (H/A)	64/57	45/47	66/54	-	56/40	-
TOTAL SCORE	**90/85**	**82/62**	**59/52**	**-**	**69/42**	**-**
Probability Rating	Very High/ Very High	Very High/ Very High	High/ High	- -	Borderline/ Low	- -

*65-69= Borderline Clinical, 70 or above=Clinical Range

Gemma. Pre-treatment parent scores placed Gemma in the Clinical Range on all subscales except 'Harm Avoidance', which scored in the Slightly Elevated Range. Parent Total Score was in the Clinical Range and indicated a Very High Probability of problematic anxiety. Self-report scores also lay in the Clinical Range on all subscales except for Harm Avoidance, with Total Score in the Clinical Range, and a Very High Probability of problematic anxiety. Post-treatment parent scores indicated a reduction on all subscale measures except for Physical Symptoms, with a reduction in Total Score from 90-82, though this remained in the Clinical Range. Gemma's self-rated scores also reduced on all subscales. Although probability rating remained in the Very High Range, Gemma's Total Score reduced from 85-62, indicating a clinically substantive reduction in anxiety overall, shifting from the Clinical Range to the Slightly Elevated Range.

Oscar. Parent-report and Self-report subscale pre-treatment scores both placed Oscar in the High Probability Range for problematic anxiety, with elevated Social Anxiety and Generalised Anxiety Scores. Total Score on the parent-report was in the High Average Range, and Self-report Total Score sat in the Average Range.

Fletcher. Fletcher's parent reported a pre-treatment elevated score on the Generalised Anxiety scale, and a score in the Clinical Range on the Obsessions and Compulsions subscale, indicating a Borderline Probability of problematic anxiety for Fletcher. Parent-reported Total Score was in the Slightly Elevated range. Fletcher's pre-treatment self-report indicated scores in the Average Range on all subscales and Total Score, with a Low Probability of problematic anxiety.

Child Behavior Checklist 6-18 (CBCL) and Youth Self-Report (YS-R)

Parents completed the CBCL at pre-treatment and post-treatment for Gemma and Oscar, with pre-treatment obtained by Fletcher's parents. Gemma and Oscar completed the Y-SR at pre and post-treatment, and Fletcher completed this at pre-treatment. CBCL data is presented in Table 7.

Table 7

Pre and Post-Treatment Subscale Scores and Total Scores on the CBCL and YS-R

CBCL(Parent) / YSR(Child)	**Gemma**		**Oscar**		**Fletcher**
Subscale	**Pre**	**Post**	**Pre**	**Post**	**Pre**
Anxious/Depressed	7/13	9/13	3/5	8/7	6/5
Withdrawn/Depressed	10/12	6/12	10/6	10/7	6/7
Somatic Complaints	10/7	9/6	0/4	4/4	4/5
Social Problems/Emotionally Reactive	6/10	9/7	4/0	2/1	3/5
Thought Problems (Sleep problems)	6/17	6/12	6/4	6/2	7/8
Attention Problems	11/15	10/12	8/4	5/5	5/10
Rule Breaking Behaviour	4/5	2/4	0/0	0/0	4/4
Aggressive Behaviour	9/12	9/9	0/1	4/0	12/7
Internalising Problems	73/71	71/70	63/61	70/64	67/63
Externalising Problems	61/61	59/56	34/34	48/37	62/53
TOTAL Score	**68/70**	**68/67**	**56/48**	**59/49**	**63/60**

T Score 60-63= Borderline Clinical, above 63 = Clinical Range

Anxiety subscale – Parent R - above 8=Borderline, above 12 Clinical. YSR 7-10 Borderline, above 10 Clinical

Gemma. Pre and post-treatment scores remained similar on Internalising Problems and Total Scores on the parent-report and self-report. Post-treatment Externalising Problems reduced from the Clinical Range to the Borderline Clinical Range on both parent (61 to 59) and self-report (61 to 56). Post-treatment Internalising Score on the Parent-report (73 to 71) and Self-report (71 to 70), and Total Scores on the Parent (68 to 68) and Self-report (70 to 67) remained in the Clinical Range following treatment, indicating little change in Internalising Problems overall.

Oscar. Pre-treatment scores on Internalising Problems were in the Borderline Clinical Range on both parent and self-report. Internalising scores rose slightly post-treatment, with both parent score (63 to 70) and self-report score (61 to 64) shifting from the Borderline into the Clinical Range. Externalising Problem scores also increased following treatment on both parent (34 to 48), and self-report (34 to 37). Total Scores on the parent

report (56 to 59) and self-report (48 to 49) also increased, but remained in the Normal Range. Overall, these scores are indicative of a slight increase of internalising and externalising symptoms following treatment.

Fletcher. Pre-treatment scores indicated Internalising Scores in the Clinical Range on parent (67) and self-report (63), with parent-report pre-treatment Externalising Score in the Borderline Range (62), and self-report score in the Normal Range. Pre-treatment Total Scores were in the Borderline Clinical Range on the parent-report (63), and the self-report (60). No post-treatment data was available.

Paediatric Quality of Life (PedsQL)

Participants completed the self-report version of the PedsQL before and after treatment, and parents also completed pre and post-treatment PedsQL questionnaires. Post-treatment measures were not obtained for Fletcher. Higher scores reflect improvement in quality of life. A standardised clinical cut-off score is not indicated for the PedsQL.

Table 8

Pre and Post-Treatment Psychometric Scores on the Pediatric Quality of Life (PedsQL)

PedsQL (Self-Report)	**Gemma**		**Oscar**		**Fletcher**	
Subscale	**Pre**	**Post**	**Pre**	**Post**	**Pre**	**Post**
Physical Functioning	59.3	68.7	56.3	68.8	68.75	
Emotional Functioning	40.0	40.0	70.0	65.0	60.0	
Social Functioning	50.0	70.0	75.0	90.0	80	
School Functioning	60	65.0	65.0	75.0	50	
Total Score	**48.5**	**60.9**	**65.2**	**73.9**	**64.6**	
PedsQL (Parent Report)						
Physical Functioning	37.5	59.3	81.2	81.3	50	
Emotional Functioning	40.0	40.0	85.0	65.0	45	
Social Functioning	35.0	35.0	50.0	70.0	60	
School Functioning	30.0	45.0	65.0	90.0	55	
Total Score	**35.6**	**44.8**	**71.7**	**77.2**	**52.5**	

An improvement in overall quality of life was reported by participants, with Total Scores increasing post-treatment for Gemma (48.5-60.9), and Oscar (65.2-73.9). Parent scores also reflected an overall increase in quality of life for Gemma (35.6-44.8), and Oscar (71.7-77.2). Scores on self-reported Emotional Functioning decreased for Oscar (70.0-65.0), and remained unchanged for Gemma (40.0 – 40.0). This was the case on parent-report also where scores were unchanged for Gemma (40.0 -40.0), and decreased for Oscar (85.0-65.0). Parent-reported Social Function score remained the same for Gemma (35.0 – 35.0), while Gemma's self-reported score on Social Functioning improved after treatment (50.0-70.0). Oscar also self-reported improvement in Social Functioning post-treatment (75.0-90.0), as did his parent (50.0-70.0). Improvements in School functioning were also reported by Gemma (60.0-75.0), and Oscar (65.0-75.0), and by their parents, with Gemma's parent score increasing from 30.0-45.0, and Oscar's parent score increasing from 65.0-90.0. Physical Functioning reflected improvement on self-reported scores (Gemma 59.3-68.7, Oscar 56.3-

68.8), and parent-reported scores (Gemma's parent 37.5-59.3, Oscar's parent 56.3-68.8). Overall, an increase in quality of life was indicated for Gemma and Oscar, except in the area of emotional functioning for both participants.

Parent Sleep Quality and Well-being

Depression-Anxiety-Stress-Scale (DASS-21)

Five parents completed the DASS-21 prior to treatment. All parents scored in the Normal Range (Clinical cut-off score ≥70), on the Depression and Stress scales at pre-treatment. Four of five parents scored in the Normal Range on the Anxiety Scale prior to treatment, with one pre-treatment parent score in the Mild Range. Post-treatment parent data on the DASS-21 measure was only obtained from Oscar's family. Of the two parents who completed post-treatment data, improvements were seen overall with reduced post-treatment scores on mother-reported Depression (1-0) and Stress (3-1) subscales, and Anxiety (1-1) subscale score unchanged. Father-reported scores decreased on the Depression (2-0) and Anxiety (1-0) scales post-treatment, and increased on the Stress scale (1-5) post-treatment.

Pittsburgh Sleep Quality Index (PSQI)

Gemma's mother, and Oscar's mother and father completed the PSQI before and after treatment. Of the three parents who completed pre and post-treatment measures of sleep quality, one parent's Global PSQI pre-treatment and post-treatment score was the same, (Gloria's parent 5-5) indicating sleep quality remained the same after treatment, in the "good" sleep range, and two parents reported slightly higher Global PSQI scores after treatment, indicating their sleep quality worsened slightly after treatment (Oscar's father 5-6, Oscar's mother 12-13), both within the "poor" sleep range.

Treatment Acceptability

Treatment Acceptability Rating Form-Revised (TARF-R)

Three of five parents (2 mothers, 1 father), and one participant completed treatment acceptability ratings after treatment. Parent and participant ratings are presented in Table 9.

Table 9

Treatment Acceptability Rating Form-Revised (TARF-R)

Subscale	**Gemma**		**Oscar**	
	Mother	**Self**	**Mother**	**Father**
Reasonableness	17/21	15/21	20/21	18/21
Effectiveness	16.5/21	16/21	17/21	18/21
Side Effects	13.5/21	16/21	17/21	19/21
Disruptive/Time Consuming	16.5/21	16/21	12/21	18/21
Cost	14/14	14/14	14/14	14/14
Willingness	18/21	16/21	20/20	21/21
Problem Severity*	8 /14	8/14	7/14	9/14
Understanding*	6/7	7/7	6/7	6 /7
Total Acceptability	**95.5/119**	**93/119**	**100/119**	**108/119**

NB: *Subscales not included in Total Score. Maximum Total Acceptability Score 119

Parent Total Acceptability Scores ranged from 95.5-108, and participant Gemma rated a Total Acceptability score of 93. Scores indicate parents and the participant found the treatment cost effective with all Cost subscale scores at 14/14. Reasonableness scores were also high, with scores ranging from 15-20/21. Scores also indicate families found the treatment easy to understand (score range 6-7/7), and families were willing to engage in the treatment (score range 16-21/21). Gemma's family noted some side effects (score range 13.5-16/21), and both families indicated treatment could be disruptive or time consuming

(score range 12-18/21). Total Acceptability ratings were high, suggesting families found the treatment process acceptable overall.

Post-treatment Feedback

Post-treatment feedback was obtained from Oscar's mother, and from Gemma. Themes were noted in feedback from both families. Challenges were experienced with adhering to early morning waketimes, and daytime tiredness occurred in the early stages of treatment. Oscar's family found providing daily feedback and writing up diaries a struggle at times. Gemma expressed that recording her thoughts daily in sleep diaries was the most helpful aspect of treatment. Both families reported a strength of the treatment was the kindness, patience and problem solving support of the intern psychologist. Overall the treatment was reported by both families as a significant commitment, but highly effective, and worth the time and effort.

Reliability

Inter-observer Agreement (IOA)

IOA data was calculated for 3% of nights with Oscar (SOL), and 2% of nights for Gemma (SOL). Mean IOA was 95% for Gemma, and 88.3% for Oscar. For Fletcher, IOA was calculated for 23% of nights across baseline and treatment phases (IOA M = 95.3%).

Treatment Fidelity

For two participants (Gemma and Oscar), treatment fidelity was calculated for 25% of nights across treatment, based on adherence to sleep/wake schedules. Treatment fidelity was 80% for Gemma, and 83% for Oscar. For Fletcher, treatment fidelity was calculated for 25% of nights across Phase One with a Mean of 33%. Due to the low Phase One treatment fidelity rating, this was also calculated across 25% of Phase Two nights based on adherence to sleep hygiene checklist. Fletcher's Mean score for Phase Two was 80%.

Chapter 6: Discussion

Research Aims and Treatment Goals

The overall aims of this research were: 1) to evaluate the effectiveness of an FBA-informed behavioural sleep intervention in autistic youth experiencing anxiety, 2) to evaluate the maintenance of treatment effects over time, 3) to examine the effect of improving sleep on adolescents' experience of anxiety, daytime behaviour, quality of life, and parent well-being, and 4) to evaluate the parents' and young person's perceptions of the acceptability and effectiveness of behavioural sleep treatment. Study One describes a case study of a 13-year-old girl who experienced sleep disturbance and anxiety, particularly in relation to separating from her mother for night time sleep. Study Two used a single-case multiple-baseline AB design across three participants, to investigate the effects of FBA-informed behavioural sleep treatment for autistic youth experiencing sleep disturbance and anxiety.

This chapter will first outline key findings from Study One, followed by key findings from Study Two. This is followed by a discussion of overall findings related to the research aims, highlighting points of interest about the relationship between anxiety and sleep disturbance in autistic youth. Study strengths and limitations are noted, as are suggestions for future research, clinical implications, and final conclusions.

Key Findings: Study One

This case study involved a thirteen-year-old girl (Wynter) with a long history of sleep disturbance, involving difficulties with sleep onset delay, curtain calls, night wakings, and dependence on parental presence to achieve and maintain sleep throughout the night. According to parent report and psychometric assessment, Wynter was experiencing anxiety relating to various aspects of daily activities, including being separated from her mother at

night. Treatment involved the utilisation of relaxation strategies, and fading of parental presence.

The Effectiveness of FBA-informed Behavioural Sleep Treatment

Following treatment, Wynter and her family experienced improvements in her sleep, including a reduction in the frequency and duration of NW's, and CC's. These gains were maintained at 6 and 10 weeks post-treatment. SOL demonstrated improvement in the treatment phase, but follow-up results indicated a minimal treatment effect overall for SOL. The most significant positive outcome reported by Wynter's family and evident from the data was the achievement of independent sleep onset and maintenance. Data indicated that treatment effects were maintained up to 10 weeks post intervention. It is possible that an additional treatment component targeting Wynter's sleep/wake schedule may have served to further consolidate sleep, and elicit greater improvements in SOL. Wynter and her mother expressed satisfaction with treatment outcomes by the end of treatment Phase Three. The family reported that treatment had required a significant commitment of time and effort, but this was more than worthwhile due to the achievement of independent sleep which had a marked positive impact on the family. They also noted that Wynter gaining a sense of agency and independence through her involvement in the intervention was an important factor in treatment success.

A recent study suggested participant's active involvement in intervention steps was a significant factor in treatment effectiveness for autistic youth (van Deurs, 2021). Within this current case study, individualised FBA-informed intervention phases, and a high level of child participation and parent support appeared to have a strong influence on treatment success, particularly in achieving independent sleep. Personalised, highly structured, and gradual treatment steps for fading parental presence at sleep onset and night wakings were designed in collaboration with Wynter and her mother, and reinforced by a reward schedule

negotiated between them. The structured and gradual nature of treatment steps were developed largely in response to Wynter and her mother's self-reported need for tailored support in building their tolerance for distress. Highly structured, predictable plans, and close collaboration over the nature and pace of treatment appeared to be vital for the family, in maintaining their commitment and capacity to adhere to treatment. The implementation and success of treatment in this case study, reflects the utility of FBA in developing sleep interventions which are individualised, and uniquely responsive to the needs of the child and family. This is particularly pertinent in the context of autism, where autism-specific modifications to non-autistic treatment designs tend to improve intervention effectiveness for autistic individuals (Danial & Wood, 2013; Dowell et.al., 2018; Lang et.al., 2010; Moree & Davis III, 2010; Vasa et.al., 2014).

Study One Effects of Sleep Treatment on Anxiety

Overall, data suggested a reduction in Wynter's experience of anxiety following treatment, with treatment gains maintained at follow up. Psychometric data also indicated improvements in anxiety, with a reduction in sleep anxiety reflected in the CSHQ. Scores on the CBCL-YSR indicate a clinically substantive reduction in Internalising Problems and Total Problem Score, shifting from the Clinical Range to the Normal Range, including a shift from the Borderline Clinical Range into the Normal Range on the Anxious/Depressed subscale. Qualitative data from sleep diaries and post-treatment feedback indicate Wynter found the acquisition of relaxation strategies enabled her to develop a sense of agency over managing mild to moderate feelings of anxiety, particularly utilising diaphragmatic breathing to self soothe. Data suggests her ability to self-regulate feelings of anxiety developed over time. Sleep diary anxiety ratings did not reduce when initially introduced in Phase One, and increases in anxiety ratings clearly correlated with stressful life events and peaks in disturbed sleep (SOL, CC's, NW's) through treatment Phase One, Two, and the first weeks of Phase

Three. Although no significant life stressors are reported in Phase Three, it is possible that Wynter may have become less reactive to external stressors during this phase, and more able to manage anxious feelings independently, resulting in a reduction of anxious feelings noted in post-treatment psychometric data.

Wynter also reported she found it challenging to apply relaxation strategies when experiencing intense feelings of anxiety. According to both parent and participant report, Wynter utilised relaxation strategies intermittently, stating they helped 'sometimes' when falling asleep or waking in the night, and only when she was experiencing mild to moderate states of hyperarousal. Wynter noted when she was feeling very anxious or excited, she was unable to engage in self-management strategies of visualisation or deep breathing, as she was "too stressed", or "over excited". It is unclear whether strategies were regularly practiced at times when Wynter was not feeling anxious. It is possible that greater adherence to treatment, i.e. consistent daily practice of relaxation strategies at times of lower arousal, may have increased her capacity to utilise these strategies when faced with more intense states of anxiety or hyperarousal.

There are other factors noted in the literature which may be linked to limitations in Wynter's capacity to apply relaxation strategies in the face of higher states of anxiety or distress (Daughters et.al., 2014; Doan et.al., 2018). Distress tolerance can be defined behaviourally, as the ability to perform goal-directed behaviour while experiencing physical or psychological distress (Daughters et.al., 2014). For Wynter, goal-directed self-management strategies targeting reduction of physiological and cognitive arousal, were only utilised and effective for her when she was experiencing mild to moderate anxiety (distress). This suggests Wynter had a limited capacity for tolerating distress – where she was unable to carry out an independent, goal-directed behaviour such as relaxation strategies, in the face of anything beyond a mild state of anxiety or distress-related hyperarousal. During times when

her state of arousal or distress was beyond what she experienced as manageable, she was less able to manage independently, seeking out her mother's presence to calm her and return to sleep.

Additionally, Wynter's mother's responses at times during treatment appear to have been influenced by her own struggle to tolerate Wynter's distress, over and above adherence to treatment guidelines. She reportedly struggled to adhere to treatment guidelines around withholding parental attention in situations when she perceived Wynter as unable to manage her feelings of distress independently. Research indicates links between maternal regulation of distress, and distress tolerance in children (Doan et.al., 2018), and maternal response to adolescent distress, and distress tolerance in adolescent daughters (Daughters et.al., et.al., 2014). In this case study, both the participant and parent's capacity to tolerate distress developed significantly throughout the treatment process.

Study One Conclusions

This study highlights the importance of collaborating with autistic youth and their family in formulating and individualising treatment steps which are acceptable, feasible, and manageable in demand and pace. The level of trust and collaboration the participant maintained with the intern psychologist was reported as a significant factor in overall successful treatment outcomes, and their maintenance. This researcher was not the primary clinician in this case, and face-to face involvement with the family was limited. It is possible the limited amount of active engagement between researcher and participant may have influenced anxiety-related outcomes. One treatment component (relaxation strategies and psychoeducation on anxiety) was introduced directly to the participant by the researcher (accompanied by the intern psychologist), through one face–to-face session, and opportunity for the development of participant-researcher rapport and trust was minimal. In line with established research, rapport and trust between therapist and client can have a significant

impact on treatment outcomes for non-autistic (Clark, 2013), and autistic youth (Kerns et.al., 2017). Literature suggests that a warm interpersonal connection, and a trusting, collaborative therapeutic relationship enhances clients' willingness to learn and acquire new ways of thinking and behaving, directly influencing treatment outcomes (Clark, 2013; Kerns et.al., 2017). It is possible that minimal personal contact between researcher and participant resulted in a lack of therapeutic alliance. This may have influenced the participant's level of understanding about rating anxiety, and their intermittent engagement in relaxation strategies, and this may have impacted treatment outcomes.

Findings from this case study demonstrate a relationship between anxiety and sleep, where external stressors which elicit anxiety, and reported hyperarousal states clearly correlated with increased sleep disturbance, as evidenced in previous literature (Carpenter et.al., 2019; Hollway et.al., 2013; Mazureck et.al., 2015; Nadeau et.al., 2015). Additionally, psychometric and qualitative data suggest improvements in sleep may reduce anxiety, which has been inferred in previous studies (Brown et.al., 2018; Souders et.al., 2017). Treatment clearly resulted in positive outcomes for this family with improvements in sleep, quality of life, daytime behaviour, and well-being. However, clinical findings from this case study are subtle, and not clear enough to make robust conclusions, or provide clarity on the exact nature of the relationship between sleep and anxiety in autistic individuals.

Effective sleep treatment clearly impacted this participant's experience, and self-management of anxiety within the context of sleep, with indications of positive effects on anxiety experienced outside of the sleep context also. During the process of treatment and follow up in Study One, several points of interest emerged, raising questions about the nature of the relationship between anxiety and sleep in autistic youth. For example, can improvements in sleep significantly reduce anxiety experienced outside the context of sleep? Do strategies used to manage anxiety within the sleep context generalise to other contexts

where anxiety is experienced? Participants selected for Study Two appeared to be experiencing anxiety related sleep disturbance, and reported experiences of anxiety in other contexts of their daily life. This presentation of anxiety and sleep disturbance enabled further exploration of the complex bi-directional relationship between anxiety and sleep.

Key Findings: Study Two

All participants in Study Two were autistic adolescents experiencing sleep disturbance, and self or parent reported anxiety within the context of sleep, and other contexts. Participants in Study Two received FBA-informed behavioural sleep treatment to address sleep disturbance. Overall, those who completed treatment (2 of the 3 participants) demonstrated evidence of significant improvement in SOL, FNW, and DNW, and attainment of sleep goals. Evidence on the impact of sleep improvements on anxiety was variable.

Effectiveness of FBA-informed Behavioural Sleep Treatment for Anxious Autistic Youth

In Study Two, all participants commenced treatment with adjustment to sleep/wake schedules. For one participant (Fletcher), intervention also included an additional phase with modifications to sleep hygiene, though this participant withdrew from the study prior to treatment completion. Sleep/wake schedule adjustments were individualised for each participant, according to data obtained through assessment and baseline monitoring indicating the time of night when they typically fell asleep, and woke in the morning. A morning waketime was set, according to recommended age-related sleep quantity, and family routines. Sleep and waketimes were adjusted as needed during the treatment phase until families' sleep goals were reached. One participant (Gemma) also recorded her thoughts in sleep diaries for data monitoring, in preparation for further anxiety treatment as required.

Improvement in SOL was evident for the two participants (Gemma and Oscar) who completed sleep treatment. Sustained reductions in FNW and DNW were also seen during,

and at the completion of treatment for Gemma and Oscar. For the other participant, Fletcher, SOL remained variable throughout treatment. This variability is likely linked to challenges with treatment adherence, with treatment fidelity related to sleep/wake schedules low, at 33% in Phase One of treatment. Fletcher also experienced periods of stable reduction in FNW and DNW during treatment Phase Two (sleep hygiene), which aligned with greater adherence to treatment protocols during this phase (80%). Reductions in sleep problems (SSR), and improvement in sleep hygiene practices (ASHS) were reported by treatment completers following treatment. One parent reported a perceived worsening of sleep problems at post-treatment on the CSHQ.

Overall, behavioural sleep treatment was effective in reducing sleep disturbance for the two participants who completed treatment, and treatment effects were maintained to varying degrees 6 -10 weeks following treatment. For treatment completers, adjustment of sleep/wake schedules alone was sufficient to reduce sleep disturbance and meet sleep treatment goals, and further treatment targeting anxiety/hyperarousal was not necessary.

These findings reflect evidence from previous literature demonstrating the effectiveness of behavioural sleep treatment for addressing problems with sleep onset and maintenance in autistic children and youth (Cuomo et.al., 2017; Rigney et.al., 2018, Souders et.al, 2017; van Deurs et.al. 2019). In particular, previous evidence has suggested adjustment of sleep/wake schedules (faded bedtime) alone can be effective in ameliorating sleep disturbance in autistic children (Carmassi, 2019; Ford et.al., 2021; Hodge et.al., 2014; Hunter et.al., 2021; Luiselli et.al., 2020; Moon et.al., 2010). Other evidence has indicated additional treatment components such as sleep hygiene, (Jan et.al., 2008;Vriend et.al., 2011), or relaxation strategies (Souders et.al., 2017; van Deurs et.al., 2021) may be required.

Study Two: Effects of Sleep Treatment on Anxiety

Limited data were available from Study Two participants, and reflected mixed findings related to participants' experience of anxiety following sleep treatment. Overall anxiety outcomes were variable across participants, and measures. MASC-2 data indicated clinically substantive improvements in parent and self-reported anxiety following treatment, while CBCL and YS-R ratings on Anxiety and Internalising Problems remained unchanged, or slightly worse following treatment for the two treatment completers. Daily self-rated anxiety completed by one participant remained similar during the treatment phase, and increased at follow-up, compared to baseline data. The variable results evident in this study align with current literature. Some research demonstrates a reduction in anxiety following behavioural sleep treatment for autistic children (Brown et.al., 2018; Loring et.al., 2018). Other literature has produced mixed or inconclusive results, which perhaps highlights the complexity and heterogeneity of reciprocating influences between sleep and anxiety in autistic individuals (Brown et.al., 2018: Hollway et.al., 2011b; Nadeau et.al., 2015).

It is interesting to note that although adjustment to sleep/wake schedule alone was sufficient treatment for Study Two participants to attain sleep goals, one participant reported a data monitoring process not directly related to sleep/wake scheduling, as a contributor to treatment success. Gemma reported that the act of writing down her thoughts in sleep diaries prior to bedtime was the "most helpful" aspect of treatment. Although the recording of thoughts in isolation does not constitute the full therapeutic component of cognitive restructuring in evidence-based CBT therapy for anxiety, identifying thoughts which may contribute to anxious feelings and arousal is considered the first step in cognitive restructuring (Attwood, 2004; Huebner, 2006; Kerns et.al., 2017). In CBT treatment for anxiety, this step is typically followed by the challenging of maladaptive thinking patterns, and replacing anxious cognitions with more adaptive cognitions, or 'helpful self-talk'

(Attwood, 2004; Huebner, 2006; Kerns et.al., 2017. Literature indicates these steps utilised together as part of a comprehensive CBT treatment package, demonstrate evidence of reducing anxiety in non-autistic youth (Chorpita, 2007; Davis et.al., 2011b; Read et.al., 2013), and autistic youth (Danial & Wood, 2013; van Steensel & Bogels, 2015). It is possible therefore that the procedure of recognising and recording thoughts possessed some therapeutic benefit for the participant in this current study, reducing cognitive hyperarousal and promoting sleep, but it is not possible to determine the extent to which this contributed to improvements in sleep outcomes. The small number of participants in this study means findings must be interpreted with caution. However several points of interest arise from Study Two regarding the nature of the anxiety-sleep relationship, and this is discussed further in the Overall Findings section.

Overall Findings – Study One and Two

This current research suggests FBA-informed behavioural sleep treatment can be effective in reducing a variety of sleep disturbances for anxious autistic youth, including problems with SOL, NW's, CC's, and achieving independent sleep onset and maintenance. Treatment effects were largely maintained at 6, and 10 weeks after treatment. Overall, evidence of improvements in anxiety following sleep treatment was variable within, and across participants.

Study One and Two Maintenance of Treatment Effects

Improvements in sleep were largely maintained across studies, for participants who completed treatment. These findings align with previous research indicating treatment effects can be maintained over time for autistic youth, following behavioural treatment for sleep disturbance (Georen et.al., 2022; van Deurs et.al., 2019). Evidence indicating maintenance of treatment effects is significant, considering the prevalence and pervasiveness of sleep

difficulties in the autistic population. Sleep disturbances are known to increase the risk of internalising disorders such as anxiety or depression (Mazureck et.al., 2015; Nadeau et.al., 2015; Rzepecka et.al., 2011), and thus increase the risk of internalising disorders affecting autistic individuals into adulthood if sleep is poorly managed (Forde et.al., 2022; Gustemps et.al., 2021). Effective treatment which endures is vital for this adolescent population, in supporting their successful transition into the independence of young adulthood. Treatments which enable autistic individuals to sustain healthy sleep habits into adulthood increases the potential for positive future outcomes; supporting their capacity to participate fully in community, employment, or education settings, without being impaired by sleep disturbance (van Deurs et.al., 2020).

Study One and Two Treatment Fidelity

The current findings suggest that greater treatment adherence was associated with increased treatment effect. Previous research indicates overall treatment effectiveness is linked to treatment adherence where higher levels of adherence are associated with greater improvements in sleep (Riedel & Lichstein, 2001; Vincent & Hameed, 2010). Essentially, individuals cannot benefit from interventions which they have not been able to engage in consistently (Miller & Rollnick, 2014). It appears treatment adherence likely influenced treatment outcomes in this current study, where higher levels of treatment adherence are associated with positive treatment effects, and vice versa. In Study One, lower adherence to treatment guidelines was associated with reduced treatment effects at specific points during treatment. However, challenges with treatment adherence were acknowledged, and addressed collaboratively with this family during the intervention, and adjustments to treatment protocols resulted in greater adherence to treatment, and improvements in outcomes, with successful attainment of sleep goals achieved by the end of treatment. This highlights the effectiveness of a responsive, flexible, FBA-informed treatment process,

resulting in successful treatment outcomes. For one participant in Study Two (Fletcher), low treatment adherence appeared to have a significant influence on treatment, where low adherence to treatment guidelines in Phase One was associated with low treatment effect, and greater treatment adherence reflected greater treatment response in Phase Two of treatment. This aligns with the current literature, where variable treatment outcomes may be more reflective of non-adherence, rather than non-response to treatment (Eicher et.al., 2019; Vincent & Hameed, 2010). A number of factors may influence treatment adherence, including social/family support, ease of assimilation (of treatment procedures) into daily life, the presence of psychological factors such as internalising disorders, and perceived relevance, or effectiveness of treatment (Matthews et.al., 2013; Vincent & Hameed, 2010). For autistic individuals engaging in treatment, other treatment adherence factors related to autism-specific characteristics may also come into play, such as social communication differences (Malow et.al., 2014; van Deurs, 2021), or differences in social emotional motivation and understanding (Nadeau et.al., 2015; Souders et.al., 2017). It appears likely that differences in social/emotional motivation (strong engagement in a special interest) may have interfered with Fletcher's ability to adhere to sleep treatment procedures at times. His perception of the relevance of treatment shifted during the course of the intervention. This appeared to influence the family's decision to withdraw from treatment, where Fletcher noted treatment was "no longer necessary" once he transferred to flexible online learning, rather than attending scheduled morning classes. Additionally, some evidence indicates treatment adherence may be influenced by the presence of internalising problems such as anxiety or depression (Matthews et al, 2013; Vincent & Hameed, 2010). Parent and self-reported scores on the CBCL and YSR placed Fletcher in the Borderline Clinical Range for Internalising Problems, therefore it is possible this may also have influenced his engagement and adherence to treatment at the time of the intervention.

Study One and Two Treatment Acceptability

Overall findings from post treatment interviews and the TARF-R reflected similar themes regarding therapeutic process, and effectiveness of the treatment overall. Themes emerged which indicated parents and participants found treatment was time consuming, but worthwhile due to the benefits of effective treatment, and families felt well supported by the clinicians.

One specific challenge noted by all parents was that daily completion of sleep diaries was a significant commitment, and at times difficult to accomplish consistently. Participant families indicated treatment could be disruptive and time consuming, with one parent reporting side effects. All families reported that the consistent, individualised, and responsive support from their primary interventionist (intern psychologist) was valuable and essential in being able to fully engage in the treatment process.

Another consistent theme reported by participants and parents was that treatment was effective, and resulted in improvements to their child's sleep, and positive outcomes for the whole family. Families also emphasised that the effectiveness of the treatment overall and the consequent positive impacts made the time commitment and disruption well worth their time and effort. Parents and participants reported that treatment was cost effective, easy to understand, reasonable, and overall highly acceptable. Previous literature on treatment acceptability in behavioural sleep treatment reflects similar themes to those indicated in this current research, with indications of treatment being time consuming, reasonable, understandable, effective, and acceptable overall (McLay et.al., 2019; McLay et.al., 2021; van Deurs et.al., 2021). Findings from this study, and evidence from previous research demonstrate that although behavioural treatments for sleep disturbance may require a significant commitment from families, it is perceived as well worth the time and effort, as treatments are effective, and acceptable overall.

Study One and Two: Effects of Sleep Treatment on Children's Daytime Behaviour, and Quality of Life

Research findings across studies suggest that those that completed treatment experienced varying degrees of positive change in their daytime behaviour and quality of life when compared to pre-treatment data.

Daytime Behaviour. Links between reduced sleep disturbance and improvements in daytime behaviour have been demonstrated in previous studies (Cohen et.al., 2014; Goldman et.al., 2011; Hunter et.al., 2020; Meltzer, 2008), and are largely reflected in overall outcomes from this study. Two of the three families who completed treatment reported improvements in overall daytime behaviour, including a clinically significant reduction in externalising behaviour, and improvements in attention, social problems, and aggressive behavior. Conversely, one treatment completer indicated a slight worsening of overall Externalising Behaviour on the CBCL and YSR with an increase in Aggressive Behaviour reported. The small number of participants in this study prevents robust conclusions, but overall findings reflect previous findings, with an association between reductions in sleep disturbance, and improvement in daytime behaviour. Brown and colleagues (2018) noted that hormonal fluctuations during the adolescent period may exacerbate emotional dysregulation and increase externalising behaviour difficulties. It is possible given the age of the participants in this current research, that hormonal fluctuations may have influenced the daytime behaviour of the participant who did not experience improvements, however this must be interpreted with caution, due to the small number of participants represented.

Quality of Life. Findings from the PedsQL indicate all participants experienced an overall improvement in quality of life following behavioural sleep treatment. All participants reported improvements in all areas except for Emotional Functioning, and overall improvements were reflected in increased Total Scores for all participants. One participant

(Oscar) reported a deterioration in Emotional Functioning. Parent Scores reflected the self-reports, with improvements reported in all areas of functioning except for a deterioration in Emotional Functioning reported by Oscar's parents.

Overall findings from the current study align with previous research which indicates improvements in sleep are associated with improvement in quality of life for autistic children (Delahaye et.al., 2014; Malow et.al., 2014; McLay et.al., 2021 Papadopoulos et.al., 2019). Poorer emotional functioning following improvements in sleep contrasts with existing literature. Previous literature has tended to incorporate the adolescent population into either child or adult groupings, and findings relating specifically to the adolescent population is sparse (Lawson et.al., 2020). It is possible there are mechanisms at play which are a distinct feature of the adolescent period, and influence quality of life during this developmental stage, particularly for autistic individuals (Brown et.al., 2018; Lawson et.al., 2020). Further investigation into the relationship between sleep, daytime functioning, and quality of life during the adolescent period is warranted (Lawson et.al., 2020).

Study One and Two Effects of Sleep Treatment on Parent Well-being and Sleep Quality

Study data contained some variation, but reflected overall improvement in well-being for parents following their child's behavioural sleep treatment. All parents reported a reduction or stable anxiety score; 2 of 3 parents reported a reduction in stress, and 2 of 3 parents noted a reduction in depression according to DASS-21 scores. Links between child sleep and parent well-being has been demonstrated in previous research, (Hiscock et.al., 2008; Hodge et.al., 2013; Mindell et.al., 2009), and improvements in parent mental health following improvements in non-autistic children's sleep has been demonstrated in the literature (Hauck et.al., 2012; Mindell et.al., 2009). Evidence regarding the impact of sleep improvements on parental mental health with autistic children is sparse (McLay et.al., 2021). This sits within the context of evidence which indicates that parenting an autistic child can be

significantly more challenging than parenting of non-autistic children, or children with other developmental difficulties (O'Nions et.al., 2018), and is associated with increased risk of mental health challenges for parents of autistic children (Bonis, 2016; Hodge et.al., 2013). This current study included some variation in parental mental health and well-being following children's sleep improvements, including an increase in depression for one parent, and an increase in stress for another parent, although all scores remained within the Normal Range. A number of variables not necessarily captured within this study, and unrelated to adolescents' sleep, may have influenced parent wellbeing such as personal relationship factors, and health or financial stressors. Therefore it is not entirely unexpected that the relationship between adolescent sleep treatment outcomes and parent well-being is not clear in this case. Overall, this study supports emerging evidence that improvements in autistic children's sleep is linked to improvements in parent mental health and well-being (McLay et.al., 2021; Papadopoulos et.al., 2019).

Parent Sleep Quality. Study findings indicated one parent's sleep quality remained the same after treatment, while three parents reported a deterioration in sleep quality. Previous research has indicated that sleep disturbance in children adversely affects parent sleep (Hodge et.al., 2013; Lopez-Wagner et.al., 2008). Very little research has investigated the impact of improvements in child sleep on parent sleep quality. A study by McCrae and colleagues (2019), demonstrated parents experienced improvement in their sleep following child sleep treatment. Participants in that study were all children under the age of 12, and parents played a prominent role in the child's treatment. Adolescent sleep habits and behaviours may present quite differently to those of children in early to middle childhood, with shifts in sleep patterns, and more autonomy around bedtime routines and behaviours (McLay et.al., 2021). In light of this, it is possible that the relationship between parent and adolescent sleep habits differs from that of parent and child sleep habits. Therefore the sleep

quality of parents in this current study may not be strongly linked to the sleep habits of their adolescent child, and deterioration in parent sleep following their adolescent's treatment may be influenced by factors unrelated to study outcomes. However, one parent who reported a worsening in sleep quality was strongly influenced by her adolescent's sleep, which contrasts with previous literature findings. Wynter's parent was significantly affected by her adolescent's sleep disturbance, and heavily involved in treatment. It is worth noting in this case that sleep quality measures were collected immediately following treatment, and it is possible that parent's sleep patterns may take time to be re-established following improvements in their child's sleep. It is important also to be cautious in drawing conclusions from findings related to one participant. In future research, additional data on parent sleep quality during follow-up phases may provide more robust conclusions regarding parent sleep quality after adolescent sleep treatment.

Study One and Two Overall Effects of Sleep Improvements on Anxiety in Autistic Youth

Research suggests the relationship between sleep disturbance and anxiety in autistic children is complex and bidirectional, with autism and anxiety impacting children's sleep in numerous ways (Nadeau et.al., 2015; Souders et.al., 2017). This current study appears to be no exception, with data suggesting diverse influences at play. Previous literature suggests factors related to paediatric sleep disturbance and anxiety are diverse, but also share common physiological, emotional, and behavioural factors (Cox & Olatunji, 2016; Leahy & Gradisar, 2012). Both conditions involve states of physiological and cognitive hyperarousal, emotional dysregulation, and a tendency to employ unhelpful coping strategies (Brown et.al., 2018; Hollway et.al., 2013; Paine & Gradisar, 2011). Recent research has also suggested links between circadian dysregulation and internalising disorders such as depression and anxiety (Carmassi et.al., 2019; Fang et.al, 2019). Certain autism characteristics also tend to exacerbate sleep difficulties and anxiety in autistic children, such as sensory sensitivities, and

social-emotional processing differences (Hollway et.al., 2013; Kerns et.al., 2017). Although these various factors present as diverse, a common theme is their link to arousal regulation. The manifestation of both anxiety and sleep disturbance involves states of physiological, emotional, and cognitive hyperarousal, which tend to be exacerbated by circadian or sensory dysregulation, and are commonly experienced by autistic individuals ((Brown et.al., 2018; Mazzone et.al., 2018; Puzino et.al., 2018).

It appears both anxiety and sleep are significantly influenced by arousal states, where heightened, dysregulated states of arousal can result in anxiety, and sleep difficulties, and a regulated arousal state promotes the lowered states of arousal required for sleep, and management of anxiety. The individual's interactions with, and responses to particular behavioural and environmental stimuli plays a key role in the regulation (or dysregulation) of arousal states, (Hollway et.al., 2013; O'Nions et.al., 2018; Samson et.al., 2015; Souders et.al., 2017) as demonstrated in this current study. Study outcomes are variable, yet findings bring to light a number of possible considerations regarding the relationship between anxiety, sleep, and autism.

This current study demonstrated substantial improvements in sleep for all participants who completed treatment. Findings related to anxiety were inconsistent, with some indications of reduced anxiety, and some indication of increased anxiety following treatment. Previous research has suggested the possibility that neurobiological influences in anxiety and sleep may rest along the same continuum, and in fact represent the same neurobiological features. According to the literature, a shared aetiology would be indicated if treatment of either component (sleep, or anxiety) resulted in stable, significant improvement of *both* conditions – sleep disturbance, and anxiety (Carpenter et.al., 2019; Tsypes et.al., 2013; Uhde, 2009). Therefore this current study does not support the hypothesis of shared aetiology. Study findings suggest that although sleep and anxiety appear to share similar, reciprocating

influences, (Alfano & Ginsburg, 2007; Hollway et.al., 2013; Taylor et.al., 2012), they may also possess differing neurobiological features and aetiological origins. An alternative hypothesis is that sleep disturbance and anxiety may be synergistically influenced by a third factor, such as autism characteristics (Hollway et.al., 2011b; Hollway et.al., 2013; Nadeau et.al., 2015; Uhde et.al., 2009; Mazzone et.al., 2018). Hollway and colleagues (2011b) hypothesised that autism characteristics such as differences in sensory processing, communication and social understanding may represent significant vulnerability factors which predispose children to sleep disturbance and anxiety. Environmental stressors may trigger or exacerbate both conditions, and are often maintained by subsequent maladaptive coping strategies. This current study supports evidence regarding the influence of certain vulnerability factors on sleep disturbance and anxiety. Two participants (Gemma and Wynter) reported in sleep diaries that external stressors such as school assignments, Covid-19, and social worries exacerbated their sleep disturbance, and increased feelings of anxiety. It appears that aspects of the sleep treatment (related to anxiety management) facilitated a greater degree of adaptive coping skills for managing anxiety within the sleep context (relaxation strategies for Wynter, and writing down thoughts for Gemma). This correlated with improvements in sleep, but overall reductions in anxiety outside of the context of sleep were not consistent. Difficulties with generalising skills learned within a specific context can be a challenge commonly experienced by autistic individuals (Kerns et.al., 2017). This may account for anxiety strategies learned within the context of sleep not generalising to other contexts. Additional treatment components specifically targeting anxiety in contexts outside of sleep may have improved anxiety outcomes for participants. An additional vulnerability factor to consider in light of the age of study participants, is the influence of puberty related hormonal fluctuations. Some evidence suggests hormonal changes may be a contributing factor in the exacerbation of anxiety (and sleep disturbance) for autistic individuals during the

adolescent period (Alvaro et.al., 2017; Brown et.al., 2018). It is possible that given the age of study participants, hormonal changes may have exacerbated their experience of anxiety and internalising problems, reflected in outcomes during the treatment and follow up phases of this study.

Another possibility to consider, is the role of exposure as a mechanism in the treatment of anxiety. For participants in Study Two, behavioural sleep treatment alleviated sleep disturbance, but did not appear to result in significant reduction of anxiety outside the context of sleep. It is well established in the literature that graded exposure to feared situations is an essential component of anxiety treatment, where facing feared situations and learning to manage anxious responses reduces anxiety, while avoiding anxious situations and feelings tends to increase anxiety over time (Chorpita & Daleiden, 2009; Danial & Wood, 2013; Dowell et.al., 2018; Peterman et.al., 2015). The process of graded exposure also results in a regulated state of arousal in situations which previously elicited a heightened state of arousal (Chorpita & Daleiden, 2009; Kerns et.al., 2017). Sleep treatment for Study One participant Wynter involved the gradual removal of parental presence from the room, to achieve independent sleep. This process intrinsically involves an element of graded exposure to her feared stimulus (separation from her mother in the context of sleep), and this resulted in significant behaviour change (achieving independent sleep), and evidence of a reduction in her overall perceived experience of anxiety. Participants in Study Two did not receive direct treatment targeting anxiety, and the nature of their treatment did not directly involve exposure to anxiety provoking situations. It is possible the absence of this exposure component may have minimised reductions in overall anxiety for these participants. Contrasts in treatment components and outcomes between Study One and Two also suggest another possibility regarding the role of arousal regulation in the relationship between anxiety, and sleep.

A heightened state of arousal interferes with sleep, regardless of its origins (e.g. anxiety and/or circadian dysregulation), and a lowered arousal state is essential for achieving sleep (Brown et.al., 2018; Papadimitriou et.al., 2005). It appears behavioural sleep treatment resulted in participants being able to regulate their arousal state sufficiently to ameliorate sleep disturbance, but this capacity to regulate arousal states did not translate to heightened arousal (anxiety) experienced in other contexts, particularly for participants in Study Two. This suggests the likelihood of sleep pressure being the primary mechanism for Study Two participants' achieving the lowered arousal state sufficient for sleep onset. Sleep drive was increased sufficiently (through regulating the sleep/wake cycle), to override other sleep interfering factors such as anxiety-related cognitive or physiological arousal. Previous literature has demonstrated that increases in sleep pressure can effectively reduce pre-sleep arousal states, improving sleep (Leahy & Gradisar, 2012; Vriend, et.al., 2011). If sufficient sleep drive moderated the lower state of arousal required for sleep, this could explain why arousal regulation was not experienced by Study Two participants in other contexts during the day, when sleep pressure is appropriately low, and not influencing arousal states.

Another factor which may have impacted anxiety related data in this study, is the possibility of individual autism characteristics influencing participants' reporting of anxiety data. Previous research has noted autism characteristics such as diminished awareness of internal emotional states (limited interoception), challenges in expressing abstract concepts, or reduced motivation to report on these symptoms may impact the accuracy of existing diagnostic instruments in detecting anxiety, particularly those which rely on self-report data (Wood & Gadow, 2010, South & Rodgers, 2017). It is possible this represents a contributing factor in variable self-reported anxiety data in this current study, where core features of autism such as diminished awareness of emotional states or reduced motivation to report

symptoms, may have elicited inaccurate representations of the nature and extent of participants' experience of anxiety.

Findings from this current study overall suggest that sleep treatment alone may not be sufficient to reduce anxiety experienced outside of the sleep context, in autistic youth. Significant improvements in sleep and achievement of sleep goals were attained by participants. This suggests behavioural sleep treatment can be effective in ameliorating anxiety which interferes with sleep (cognitive and physiological hyperarousal), but reductions in anxiety experienced outside of the sleep context were not clearly evidenced. Further treatment specifically targeting anxiety may be required for autistic individuals experiencing co-occurring anxiety and sleep disturbance. Future research investigating the impact of anxiety treatment on sleep disturbance (and vice versa) may provide greater clarity on the nature of anxiety in autistic youth, and the bi-directional influences at play in the anxiety-sleep relationship. The role of arousal dysregulation in anxiety and sleep disturbance is highlighted in this study. Arousal dysregulation is a common factor affecting both anxiety and sleep, and frequently experienced by autistic youth. Components of evidence-based sleep, and anxiety treatments target arousal regulation, but the integration of sleep and anxiety treatment into one package is still rare. Further research exploring the mechanisms underlying the relationship between anxiety and sleep would promote greater understanding of optimal treatments for anxious autistic individuals experiencing sleep disturbance.

Study Strengths and Limitations

The implementation of a single case multiple baseline A-B research design is a strength of this current study. Single case research designs (SCRD) allow participants to act as their own control, enabling treatment effects in individual participants to be detected when comparing active treatment data of outcome variables with baseline data (Kazdin, 2019). Additionally, the use of a multiple baseline enables behaviour change to be measured across

participants with similar behaviours, whilst also allowing for the complexities of a heterogeneous population such as autistic individuals represent (Shadish & Sullivan, 2011). Additionally, SCRD is able to identify the occurrence of 'real world' clinical change within individual participants, for example alleviation of distress, and attainment of goals related to quality of life (Blampied, 2013). The addition of FBA-based interventions is also a strength of this study. The use of FBA enabled treatment packages to be individualised according to the needs of the participant and their family (Jin et.al., 2013; McLay et.al., 2019), aligning with the principal of minimal sufficiency in treatment (Sanders et.al., 2014). Other study designs may use a placebo, or standardised treatment package with components which may be redundant or unhelpful for some participants' individual sleep difficulties. The utilisation of FBA in this current study ensured treatments were appropriately designed to target sleep issues relevant to participants' individual presentation, and modified in pace and content as required, in being responsive to the unique needs of the child and their family throughout treatment.

Limitations are also important to note in this current study. Firstly, despite the strength of SCRD, there is a large amount of missing data. This includes gaps in sleep diary data obtained by participants, resulting from various disruptions to family routines such as family illness or holidays away from home, and technological issues with instrumented measures, significantly restricting the capacity for inter-observer data (IOA) calculations. In Study One, the family did not consent to the use of instrumented measures (videography or actigraphy), therefore it was necessary to rely on parent and participant reported sleep diary data, without the addition of IOA calculation. In Study Two, IOA data was only able to be calculated for 3% of nights for Oscar, and 2% of nights for Gemma (significantly lower that the recommended minimum of 25% IOA calculation) due to technological issues occurring with actigraph devices. Although parent and self-reported sleep diary data was able to be

triangulated with other sources such as parent and self-reported questionnaires and clinical interviews, the low proportion of IOA data reduces the reliability of sleep diary data. Also, LTFU data was only able to be obtained for 2 of the 3 treatment completers, which limits the ability to provide a robust evaluation of the maintenance of treatment effects. Further, this researcher was not the lead clinician for the cases represented in this study, and there were different lead clinicians implementing interventions for each participant. This contributed to variations on psychometric assessment and post treatment measures utilised, which affected the consistency of data collected pre to post-treatment. Finally, the small number of participants who completed intervention means findings must be interpreted with caution, and inferences may not generalise to other anxious autistic youth experiencing sleep disturbance.

Clinical Implications

This current study presents a tapestry of factors contributing to sleep disturbance, which is a common challenge for a significant portion of anxious autistic children, and highlights the utility of FBA-informed behavioural interventions for supporting autistic individuals. FBA ensures individual challenges directly related to autism characteristics can be identified, and effectively addressed throughout treatment. For example, employing a high level of participant involvement and parent support, and introducing structured, concrete steps gradually at a pace suited to the child and family. The high level of therapist responsiveness, and successive treatment modifications implemented in response to individual needs (particularly in Study One), contributed significantly to positive outcomes in this study, and demonstrates the effectiveness of FBA-informed behavioural interventions for ameliorating sleep disturbance in anxious autistic youth. Further research implementing FBA-informed interventions would enrich the valuable evidence regarding effective treatments tailored to the needs of this population.

This research draws attention to the need for further development of autism-specific assessment tools, particularly for the evaluation of internalising problems such as anxiety or depression, where autism characteristics such as limited interoceptive abilities and reduced motivation to report internalising experiences may impact the accuracy of data reported by autistic individuals. Recent research has seen the development of autism-specific measures for evaluating anxiety and other internalising disorders. The utilisation of more autism specific measures such as the Anxiety Scale for Children – Autism Spectrum Disorder (ASC-ASD), or the Parent-Rated Anxiety Scale for Youth with Autism Spectrum Disorder in future autism research, may increase the reliability and validity of anxiety data for autistic individuals, potentially increasing accurate detection and understanding of anxiety in autistic youth, and improving targeted treatment for this population.

Future Directions

Despite limitations, the findings from this current study highlight a number of possible avenues worthy of future research. The evidence base demonstrating the effectiveness of behavioural sleep intervention for the treatment of sleep disturbance in autistic children has grown in recent years. However, evaluation of specific mechanisms which may be distinct to the adolescent population, (such as hormonal fluctuations, and increasing social pressures), and their influence on anxiety and sleep during this developmental stage is rare. Future research aimed specifically at the autistic adolescent population may promote positive future outcomes which endure into the adult years for autistic individuals. Overarching questions come to light through this research project. Can anxiety treatment alone improve both anxiety and sleep disturbance, and can sleep treatment alone alleviate both sleep disturbance, and anxiety? Future research which aims to isolate treatment components targeting anxiety and sleep disturbance, and evaluate their specific effects on co-occurring anxiety and sleep disturbance may well provide greater clarity around

the bidirectional influencers at play. Research which sheds light on whether stand-alone sleep or anxiety treatment can be effective in alleviating both conditions, or whether further development of integrated sleep and anxiety treatment is necessary, is sorely needed. This offers potential for optimising treatment of co-occurring anxiety and sleep disturbance, commonly experienced by autistic youth. The development of minimally sufficient treatments which are acceptable and effective for autistic adolescents, provide vital support for individuals and families in this population, towards enjoying full participation in life as they choose. Additionally, continued research investigating the mechanisms driving sleep disturbance in anxious autistic children prior to puberty, offers the opportunity for expanding effective treatments which may ameliorate sleep disturbance and anxiety before the onset of adolescence, potentially improving outcomes and quality of life for autistic adolescents, into adulthood.

Conclusion

This study provides evidence of the effectiveness of FBA-informed behavioural sleep interventions for treating sleep disturbance in anxious autistic youth. In addition, this current study suggests behavioural sleep interventions may be effective for ameliorating sleep disturbance (including the sleep-interfering aspects of anxiety) in anxious autistic youth. However, behavioural sleep treatment alone may not be sufficient for reducing autistic individuals' experience of anxiety outside of the sleep context. The evidence base continues to grow regarding the utility of FBA-informed behavioural treatments for ameliorating sleep problems in autistic children. However much is still to be understood regarding the exact role internalising disorders such as anxiety may play in the occurrence, and maintenance of sleep disturbance in autistic adolescents. Individuals in the autistic population are prone to experiencing both anxiety and sleep disturbance. This study contributes to the literature demonstrating the utility of individualised treatments which are responsive to the unique and

complex needs of anxious autistic youth with sleep disturbance. Further research which increases our understanding of the mechanisms underlying the complex relationship between anxiety and sleep in autistic individuals would provide a worthy contribution to improving the lives and well-being of those in the autism community.

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Appendix A: Parent Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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

CONSENT FORM FOR PARENTS/ CAREGIVERS

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

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

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

- ☐ I understand that I am able to request a copy of the results of this research, should I wish to do so, and that these results will be provided for me

- ☐ I allow videotaping of my child's sleep behaviour to be completed by the researcher and understand that this videotape will be used for data gathering purposes only. I also understand that I have the right to request that video footage is destroyed at any stage.

- ☐ I consent to others, listed below, being involved in the implementation of the intervention

Name: _____

Date: _____

Signature: _____

Others I consent to implementing intervention:

Name:_____

Name:_____

Name:_____

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

Please return this form to Juliana Edwards by email, at juliana.edwards@pg.canterbury.ac.nz, OR

post to Juliana Edwards, Pukemanu Centre, University of Canterbury, Rehua Building Level 2, Private Bag 4800, Christchurch, 8140.

Appendix B: Young Person's Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An investigation into the efficacy of treatments for sleep disturbance in young people with autism

Young Person Consent Form

My name is _____.

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

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

I know if I want to stop at any time or if I do not want to be a part of this project anymore that is fine. I can tell Juliana or my caregivers.

Date: _____

Young Person's Signature: _____

Please return this form to Juliana Edwards:
juliana.edwards@pg.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: Audiovisual Recording Consent Form/Parent

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 your privacy will be protected:

- The purpose of recording is to gather data for the research project
- Audiovisual recording will only be done with your knowledge and consent
- You can withdraw your consent to audiovisual recording at any time, without having to provide a reason for changing your mind
- The audiovisual file will only be seen by the researchers
- The audiovisual recording will be securely stored at the University of Canterbury for a minimum of ten years

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

Signed: _____

Date: _____

Appendix D: Audiovisual Consent Form-Young Person

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

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

VIDEO/ ACTIGRAPH RECORDING CONSENT FORM

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

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

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

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

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

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

Only people involved in the project can view the recordings.

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

Signed:

Date:

Appendix E: Parent Caregiver Information Sheet

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

Information for Parents/Caregivers

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

Dear Parent/ Caregiver,

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

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

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

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

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

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

For the purpose of this project, an intern psychologist//Masters student will be working closely with you to conduct the necessary assessments and formulate interventions. A research assistant/Masters/PhD student who also works as a part of our sleep team, may look at some of the information that we collect, such as video recordings and actigraph data.

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

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

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

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

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

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

Appendix F: Young Person Information Sheet

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An investigation into the efficacy of treatments for sleep disturbance in young people with autism

Young Person Information Sheet

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

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

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

There may be a video camera in your bedroom sometimes. This will help me to understand what you do when you are awake and asleep. Only your caregivers and other people working on this project will be able to see this video.

We may ask you to wear an actigraph. An actigraph is worn on your wrist like a watch and it tells us when you are asleep and when you are awake.

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

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

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

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 G: Young Person's Sleep Diary

_____ Sleep Diary	**Monday**	**Tuesday**	**Wednesday**	**Thursday**	**Friday**	**Saturday**	**Sunday**
Date							
My mood rating 1 to 5:							
What time did you go to bed last night?							
How long did it take you to fall asleep? (in minutes)							
Did you fall asleep: 1. Easily 2. After some time 3. With difficulty							
What kept you awake? (e.g. thoughts, environment, etc.)							
If thoughts kept you awake, please rate: 1 to 5 Blue or Grey							
How many times did you wake in the night?							
How long were you awake during the night?							

What disturbed your sleep?							
How do you feel this morning: 1. Refreshed 2. OK 3. Tired							
What time did you wake up in the morning?							

Appendix H: Post Treatment Discussion Template

Post Treatment Feedback Discussion

The purpose of these questions is to get your feedback on how you found the sleep programme, the positives, challenges, changes it made to you and your family, and your experiences/thoughts on the sleep study overall.

1.) **Tell me about the intervention?**

2.) **How did you find the intervention process over all? (What were the strengths/positives, challenges).**

3.) **What is it you did, that you feel made a difference?**

4.) **Anything more – anything you think that didn't really make a difference?**

5.) **Comparing life before intervention to life now, what's changed for you and the family?**

6.) **Any suggestions for how the process could be improved?**

7.) **Any final words/comments**